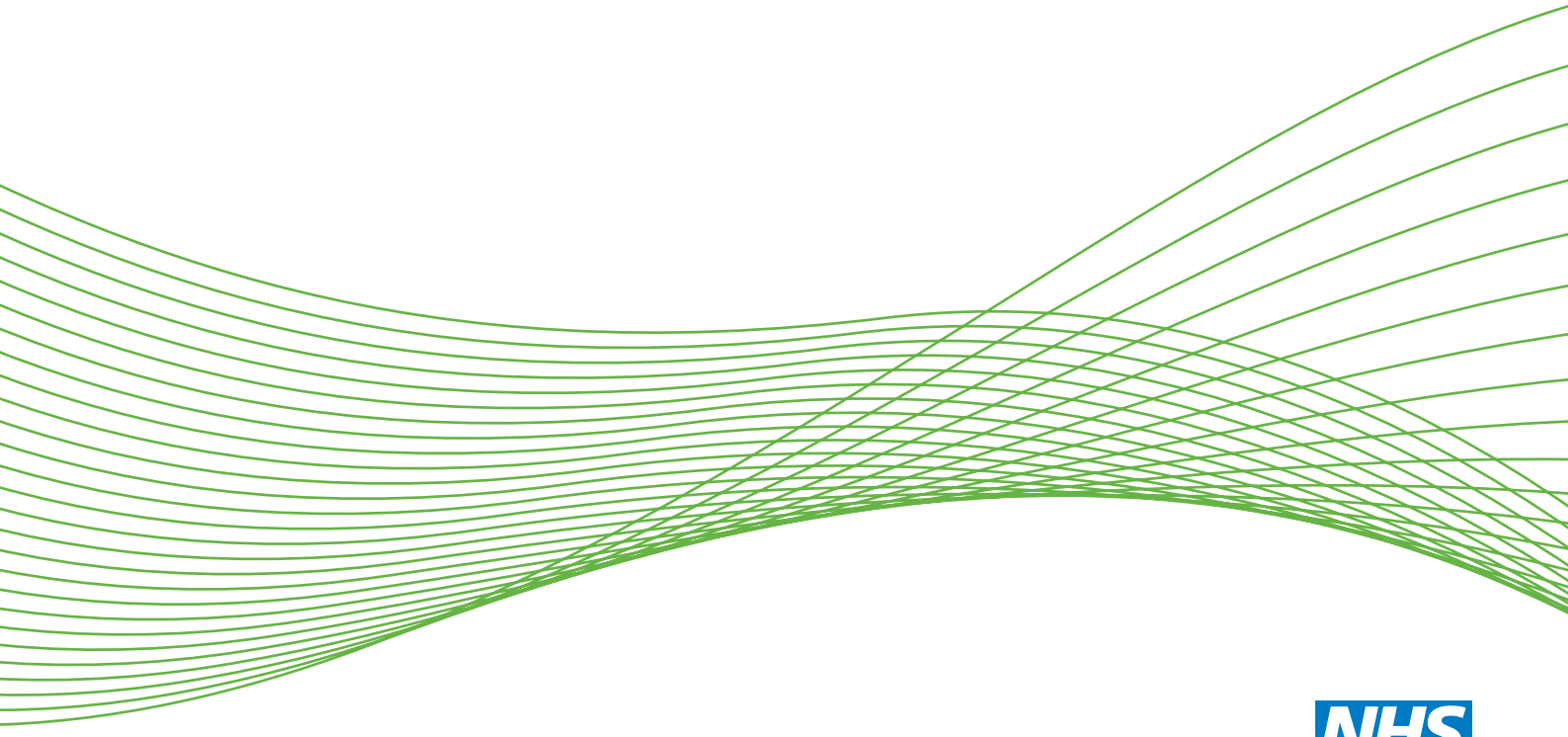
HEALTH TECHNOLOGY ASSESSMENT

VOLUME 19 ISSUE 68 AUGUST 2015
ISSN 1366-5278

The management of Otitis Media with Effusion in children with cleft palate (mOMEnt): a feasibility study and economic evaluation

Iain Bruce, Nicola Harman, Paula Williamson, Stephanie Tierney, Peter Callery, Syed Mohiuddin, Katherine Payne, Elisabeth Fenwick, Jamie Kirkham and Kevin O'Brien



National Institute for Health Research

DOI 10.3310/hta19680

The management of Otitis Media with Effusion in children with cleft palate (mOMEnt): a feasibility study and economic evaluation

Iain Bruce,[1] Nicola Harman,[2] Paula Williamson,[2,3] Stephanie Tierney,[4] Peter Callery,[4] Syed Mohiuddin,[5] Katherine Payne,[5] Elisabeth Fenwick,[6] Jamie Kirkham[3] and Kevin O'Brien[2]*

[1]Central Manchester University Hospitals NHS Foundation Trust, Royal Manchester Children's Hospital, Manchester, UK
[2]The Healing Foundation Cleft and Craniofacial Clinical Research Centre, School of Dentistry, University of Manchester, Manchester, UK
[3]Department of Biostatistics, Institute of Translational Medicine, University of Liverpool, Liverpool, UK
[4]School of Nursing, Midwifery and Social Work, University of Manchester, Manchester, UK
[5]Manchester Centre for Health Economics, Institute of Population Health, University of Manchester, Manchester, UK
[6]ICON Health Economics, ICON plc, Oxford, UK

*Corresponding author

Declared competing interests of authors: none.

Disclaimer: this report contains transcripts of interviews conducted in the course of the research and contains language that may offend some readers.

Published August 2015
DOI: 10.3310/hta19680

This report should be referenced as follows:

Bruce I, Harman N, Williamson P, Tierney S, Callery P, Mohiuddin S, *et al*. The management of Otitis Media with Effusion in children with cleft palate (mOMEnt): a feasibility study and economic evaluation. *Health Technol Assess* 2015;**19**(68).

Health Technology Assessment is indexed and abstracted in *Index Medicus*/MEDLINE, *Excerpta Medica*/EMBASE, *Science Citation Index Expanded* (SciSearch®) and *Current Contents®*/ Clinical Medicine.

Health Technology Assessment

HTA/HTA TAR

ISSN 1366-5278 (Print)

ISSN 2046-4924 (Online)

Impact factor: 5.116

Health Technology Assessment is indexed in MEDLINE, CINAHL, EMBASE, The Cochrane Library and the ISI Science Citation Index.

This journal is a member of and subscribes to the principles of the Committee on Publication Ethics (COPE) (www.publicationethics.org/).

Editorial contact: nihredit@southampton.ac.uk

The full HTA archive is freely available to view online at www.journalslibrary.nihr.ac.uk/hta. Print-on-demand copies can be purchased from the report pages of the NIHR Journals Library website: www.journalslibrary.nihr.ac.uk

Criteria for inclusion in the *Health Technology Assessment* journal

Reports are published in *Health Technology Assessment* (HTA) if (1) they have resulted from work for the HTA programme, and (2) they are of a sufficiently high scientific quality as assessed by the reviewers and editors.

Reviews in *Health Technology Assessment* are termed 'systematic' when the account of the search appraisal and synthesis methods (to minimise biases and random errors) would, in theory, permit the replication of the review by others.

HTA programme

The HTA programme, part of the National Institute for Health Research (NIHR), was set up in 1993. It produces high-quality research information on the effectiveness, costs and broader impact of health technologies for those who use, manage and provide care in the NHS. 'Health technologies' are broadly defined as all interventions used to promote health, prevent and treat disease, and improve rehabilitation and long-term care.

The journal is indexed in NHS Evidence via its abstracts included in MEDLINE and its Technology Assessment Reports inform National Institute for Health and Care Excellence (NICE) guidance. HTA research is also an important source of evidence for National Screening Committee (NSC) policy decisions.

For more information about the HTA programme please visit the website: http://www.nets.nihr.ac.uk/programmes/hta

This report

The research reported in this issue of the journal was funded by the HTA programme as project number 09/167/02. The contractual start date was in December 2011. The draft report began editorial review in May 2014 and was accepted for publication in August 2014. The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HTA editors and publisher have tried to ensure the accuracy of the authors' report and would like to thank the reviewers for their constructive comments on the draft document. However, they do not accept liability for damages or losses arising from material published in this report.

Published by the NIHR Journals Library (www.journalslibrary.nihr.ac.uk), produced by Prepress Projects Ltd, Perth, Scotland (www.prepress-projects.co.uk).

Abstract

The management of Otitis Media with Effusion in children with cleft palate (mOMEnt): a feasibility study and economic evaluation

Iain Bruce,[1] Nicola Harman,[2] Paula Williamson,[2,3] Stephanie Tierney,[4] Peter Callery,[4] Syed Mohiuddin,[5] Katherine Payne,[5] Elisabeth Fenwick,[6] Jamie Kirkham[3] and Kevin O'Brien[2]*

[1]Central Manchester University Hospitals NHS Foundation Trust, Royal Manchester Children's Hospital, Manchester, UK
[2]The Healing Foundation Cleft and Craniofacial Clinical Research Centre, School of Dentistry, University of Manchester, Manchester, UK
[3]Department of Biostatistics, Institute of Translational Medicine, University of Liverpool, Liverpool, UK
[4]School of Nursing, Midwifery and Social Work, University of Manchester, Manchester, UK
[5]Manchester Centre for Health Economics, Institute of Population Health, University of Manchester, Manchester, UK
[6]ICON Health Economics, ICON plc, Oxford, UK

*Corresponding author kevin.o'brien@manchester.ac.uk

Background: Cleft lip and palate are among the most common congenital malformations, with an incidence of around 1 in 700. Cleft palate (CP) results in impaired Eustachian tube function, and 90% of children with CP have otitis media with effusion (OME) histories. There are several approaches to management, including watchful waiting, the provision of hearing aids (HAs) and the insertion of ventilation tubes (VTs). However, the evidence underpinning these strategies is unclear and there is a need to determine which treatment is the most appropriate.

Objectives: To identify the optimum study design, increase understanding of the impact of OME, determine the value of future research and develop a core outcome set (COS) for use in future studies.

Design: The management of Otitis Media with Effusion in children with cleft palate (mOMEnt) study had four key components: (i) a survey evaluation of current clinical practice in each cleft centre; (ii) economic modelling and value of information (VOI) analysis to determine if the extent of existing decision uncertainty justifies the cost of further research; (iii) qualitative research to capture patient and parent opinion regarding willingness to participate in a trial and important outcomes; and (iv) the development of a COS for use in future effectiveness trials of OME in children with CP.

Setting: The survey was carried out by e-mail with cleft centres. The qualitative research interviews took place in patients' homes. The COS was developed with health professionals and parents using a web-based Delphi exercise and a consensus meeting.

Participants: Clinicians working in the UK cleft centres, and parents and patients affected by CP and identified through two cleft clinics in the UK, or through the Cleft Lip and Palate Association.

Results: The clinician survey revealed that care was predominantly delivered via a 'hub-and-spoke' model; there was some uncertainty about treatment strategies; it is not current practice to insert VTs at the time of palate repair; centres were in a position to take part in a future study; and the response rate to the survey was not good, representing a potential concern about future co-operation. A COS reflecting the opinions of clinicians and parents was developed, which included nine core outcomes important to both health-care professionals and parents. The qualitative research suggested that a trial would have a 25% recruitment rate, and although hearing was a key outcome, this was likely to be due to its psychosocial consequences. The VOI analysis suggested that the current uncertainty justified the costs of future research.

Conclusions: There exists significant uncertainty regarding the best management strategy for persistent OME in children with clefts, reflecting a lack of high-quality evidence regarding the effectiveness of individual treatments. It is feasible, cost-effective and of significance to clinicians and parents to undertake a trial examining the effectiveness of VTs and HAs for children with CP. However, in view of concerns about recruitment rate and engagement with the clinicians, we recommend that a trial with an internal pilot is considered.

Funding: The National Institute for Health Research Health Technology Assessment programme. This study was part-funded by the Healing Foundation supported by the Vocational Training Charitable Trust who funded trial staff including the study co-ordinator, information systems developer, study statistician, administrator and supervisory staff.

Contents

List of tables

List of figures

List of boxes

List of abbreviations

AOM	acute otitis media
CENTRAL	Cochrane Central Register of Controlled Trials
CG60	clinical guideline 60
CI	confidence interval
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CLAPA	Cleft Lip & Palate Association
COMET	Core Outcome Measures in Effectiveness Trials
COS	core outcome set
CP	cleft palate
CRANE	Craniofacial Anomalies Network
CYPC	Children and Young Persons Council
DN	do nothing
EconLit	American Economic Association's electronic bibliography
ENT	ear, nose and throat
EVPI	expected value of perfect information
EVPPI	expected value of partial perfect information
GP	general practitioner
GRADE	Grading of Recommendations, Assessment, Development and Evaluation
HA	hearing aid
HRQoL	health-related quality of life
HTA	Health Technology Assessment
ICER	incremental cost-effectiveness ratio
mOMEnt	management of Otitis Media with Effusion in children with cleft palate
NB	net benefit
NICE	National Institute for Health and Care Excellence
OME	otitis media with effusion
pEVPI	population-level expected value of perfect information
PSA	probabilistic sensitivity analysis
QALY	quality-adjusted life-year
QoL	quality of life
R&D	research and development
RCT	randomised controlled trial
SAG	Study Advisory Group
SD	standard deviation
SMG	Study Management Group
SSC	Study Steering Committee
VOI	value of information
VT	ventilation tube
WTP	willingness to pay

Plain English summary

Clinicians are unsure regarding the best way to treat children with cleft palate (CP) and 'glue ear'. They can give these children hearing aids or carry out an operation to put a small plastic tube (grommet) in their eardrum, which helps them to hear. The aim of our study was to find out the best method of studying how well these treatments work.

The study had several stages. In the first stage we surveyed the clinicians working in cleft lip and palate centres to find out their methods of treatment. This was followed by interviews with patients and their parents to find if they were willing to take part in a study and to discover the results that are most important to them. We also carried out a computer-based survey and had meetings with clinicians and parents of children with CP to find out the results of the treatment that are important to them. Finally, we carried out an analysis to find out how much should be spent on research into the treatment of glue ear.

We found out that further research is necessary. This should be a clinical trial carried out in the cleft lip and palate centres in the UK, measuring the results of treatment important to clinicians and patients. We had some concerns about potential recruitment rate and these can be addressed by designing a trial incorporating an initial study to evaluate recruitment. We could then decide whether or not to move to a full trial 6 months after initial recruitment has started.

Scientific summary

Background

Cleft lip and palate are among the most common congenital malformations, with an overall incidence of around 1 in 700 individuals. Approximately 90% of children with cleft palate (CP) have a history of non-trivial otitis media with effusion (OME). OME ('glue ear') is the accumulation within the middle-ear space of a mucoid or serous fluid. Although the exact mechanism for the development of OME is not fully understood, dysfunction of the Eustachian tube connecting the middle-ear space to the postnasal space is thought to be of fundamental importance. The function of the Eustachian tube is to equalise pressure either side of the tympanic membrane, avoiding the development of negative pressure in the middle ear. In children, the Eustachian tube does not work as efficiently, with the resultant tendency towards the development of negative middle-ear pressure and the accumulation of fluid within the middle-ear space (OME). This tendency towards Eustachian tube dysfunction is further increased in children with CP due to dysfunction of the muscles originating from the palate which act to open the Eustachian tube orifice.

There are several approaches to the management of OME in children with clefts and they include watchful waiting, the provision of hearing aids (HAs) and the insertion of ventilation tubes (VTs). However, the evidence underpinning these strategies is not clear and there is a need to determine (i) the optimum study design to investigate which treatment is the most appropriate for children with CP; and (ii) whether or not the costs of running a trial are outweighed by the potential benefit of resulting information.

Objectives

i. To identify current UK practice for the treatment of OME in children with CP.
ii. To capture patient and parent opinions on willingness to take part in the trial and to identify their needs regarding the content and form of information required to make a decision on whether or not to participate.
iii. To develop a core outcome set (COS) for use in future trials of OME in children with CP.
iv. To evaluate if the extent of existing decision uncertainty about OME care for children with CP justifies the cost of further research.
v. To determine feasibility and identify the optimum study design to add to knowledge about the treatment of OME in children with CP.

Methods

Clinician survey

A survey of current clinical practice for the treatment of OME in children with CP with or without cleft lip was carried out. This was directed at collecting information on the following main areas: (i) the method of provision of care; (ii) the clinical practice; and (iii) the caseload of patients. This was sent to the 16 UK cleft centres.

Qualitative research

A qualitative methodology was adopted to explore in depth (a) parents' views about their willingness for a child to take part in a potential trial comparing VTs and HAs, and (b) outcomes of OME management considered important by parents and children.

Parents were recruited from two cleft centres in northern England. They were eligible to take part if they had a non-syndromic child with CP (including cleft lip and palate) between 0 and 11 years of age, who had a current or past diagnosis of OME. Families with particularly difficult social circumstances (e.g. domestic violence, recent bereavement) were not approached. Children aged 6–11 years were interviewed, if they were happy to talk to the researcher. Data collection continued until the sample was diverse in terms of the children's age, gender and type of treatment received for OME, and it was judged that data saturation had been reached.

Interviews were recorded with parents' consent and transcribed verbatim for analysis, with identifying features removed during this process (including names of health-care professionals). The data were analysed using framework analysis.

Development of a core outcome set

This involved the following stages: (i) a systematic review of the literature to identify a list of outcomes previously reported in studies of the treatment of OME in children with CP; (ii) a Delphi exercise to gather information on the outcomes of importance to health professionals; (iii) an online survey of parents and children with CP; and (iv) a consensus meeting.

The search strategy was applied to the Cochrane Central Register of Controlled Trials (CENTRAL), EMBASE, MEDLINE, and the Cumulative Index to Nursing and Allied Health Literature (CINAHL) (January 2006 to April 2011).

Multiple databases were utilised to maximise the sensitivity of the search. CENTRAL comprises only studies that are deemed to be controlled trials by a team of reviewers. EMBASE, MEDLINE and CINAHL include published research of various study designs. The advantages conferred by using CENTRAL in addition to the other databases are that trials from other sources of research (e.g. journals not indexed in MEDLINE and conference proceedings) are hand-searched, and controlled trials from these are included. This improves the chances of identifying all relevant studies.

Economic analysis

This involved the following stages:

i. A systematic search of the literature was conducted to identify published decision-analytic model-based economic evaluations of treatment options for the management of OME in children with CP. The search strategy was designed to retrieve relevant studies from MEDLINE, EMBASE and the American Economic Association's electronic bibliography. These databases were searched from the date of their inception to January 2014.
ii. A de novo economic model was structured and populated to estimate the incremental costs and quality-adjusted life-years (QALYs) of four potential strategies for managing children with CP and OME.
iii. Value of information analyses were performed to quantify the potential value of future research.

Results

Clinician survey

We identified lead clinicians for each of the 16 centres and received complete surveys from 10 (62%). Partial responses were received from 14 centres (87%). Two centres refused to complete any part of the survey. The survey revealed that most centres (12/13) have a 'hub-and-spoke' clinics infrastructure, with the number of 'spoke' clinics ranging from 2 to 14. In this method of delivery of care, patients are seen in the centre ('hub') by clinicians who decide on the optimum treatment. This is then delivered in

clinics/hospitals that are nearer the patient's home (spoke). The clinical practice showed adherence to the National Institute for Health and Care Excellence Guideline Development Group guidelines and practitioners prescribed both HAs and VTs. The information on caseload which we obtained was not accurate and this was supplemented from a centrally held database (Craniofacial Anomalies Network). This suggested that the caseload for each of the centres showed some uniformity, with most seeing between 35 and 60 new referrals per year. Three centres received between 90 and 130 referrals per year, and four had 35 or fewer referrals per year.

Qualitative research

Interviewees held strong opinions about treatment. Only 25% of parents were willing to enter their children into a trial. This reflected the fact that most parents were not in equipoise, and were concerned about specific risks or benefits of either VTs or HAs. Furthermore, parents required comprehensive and detailed information about HAs and VTs. In addition to information on safety procedures in a trial, the following appear to be important: a clear explanation of clinical equipoise; a need for the investigators to understand patients'/parents' previous experience of treatment (bearing in mind that the burden of care for a child with a cleft is very high); ensuring that the study is introduced by clinicians with whom the parent and child are familiar and trust; and highlighting how the study will enhance knowledge and help others in the future. Addressing these issues may optimise trial recruitment.

When we evaluated outcomes that were important to parents and children, we found that they stressed the significance of speech and language development, educational outcomes and establishing social networks. Their concerns were not solely related to hearing difficulties but were associated with having a cleft. As a result, although hearing was the key outcome, this was largely because of its consequences on social and educational development, and psychological well-being. Findings from this part of the project fed into the development of a COS.

Core outcome set

The systematic review of the literature identified 49 papers which were assessed for outcomes used. Outcomes were grouped into relevant domains and individual outcome and domain names were agreed with input from the Study Advisory Group. A final list of 45 individual outcomes, together with an additional four outcomes identified through free-text responses, were included in the Delphi. The scores provided in each round of the Delphi survey and the survey of parents and children were analysed against predefined consensus criteria. The results were then presented at a face-to-face consensus meeting attended by both health professionals and parents. At this meeting the delegates discussed and voted on whether or not the outcomes should be included in a COS. This resulted in nine outcomes being agreed for inclusion.

Economic analysis

There were limitations in the current evidence base for the management of OME in children with CP. When the treatment alternatives were considered, it appeared that the surgical insertion of VTs was likely to be the most cost-effective option, but the need for additional information from a future study is needed to inform this treatment choice. The expected value of perfect information was approximately £5.24M for a population of children with CP in England, Wales and Northern Ireland, assuming the willingness-to-pay threshold of £20,000 per QALY and a decision horizon of 10 years, suggesting that further research work in this area is potentially worthwhile. However, the expected value of partial perfect information analysis indicated significant uncertainty surrounding the estimates of hearing-level parameters used for quantifying the QALYs. Interpretation of this economic analysis should be undertaken with caution as, with no definitive guidelines identified for the treatment of OME in children, the clinical pathway used to structure the economic evaluation was developed using assumptions based on available published evidence.

Conclusions

There is a need for further study of the management of OME in children with CP. This research should be a randomised trial based in eight of the UK cleft centres. The trial should compare the effectiveness of VTs with that of HAs. Children will enter the trial when they are 2 years old and will be followed for 3 years. An initial calculation suggests that the trial should enrol a sample of at least 90 children. The outcomes should be based on the COS that has been developed, with a primary outcome of hearing. However, there is uncertainty about the required sample size and likely recruitment rate for a trial.

As a result, we recommend that additional data should be obtained from a note review of hospital records to inform the sample size calculation.

Concerns about recruitment rate could be addressed by designing a trial with an internal pilot. The aim of the internal pilot would be to check the recruitment rate and include a qualitative component to establish barriers to recruitment and optimise recruitment methods. For example, the qualitative component of our study suggested that parents were concerned about the safety of their child, were not in equipoise and were not clear on the relative risks and benefits of the potential interventions. Progression to the main trial would be reviewed at 6 months after recruitment has started.

Funding

Funding for this study was provided by the Health Technology Assessment programme of the National Institute for Health Research. This study was part-funded by the Healing Foundation supported by the Vocational Training Charitable Trust who funded trial staff including the study co-ordinator, information systems developer, study statistician, administrator and supervisory staff.

Chapter 1 Introduction

Background

Otitis media with effusion in children with cleft palate

Cleft lip and palate are among the most common congenital malformations, with an overall incidence of around 1 in 700 individuals.[1,2] Approximately 90% of children with cleft palate (CP) have a history of non-trivial otitis media with effusion (OME).[1–3] OME (also known as 'glue ear') is the accumulation within the middle-ear space of a mucoid or serous fluid. Although the exact mechanism for the development of OME is not fully understood, dysfunction of the Eustachian tube connecting the middle-ear space to the postnasal space is thought to be of fundamental importance. The function of the Eustachian tube is to equalise pressure either side of the tympanic membrane, avoiding the development of negative pressure in the middle ear. In children, the Eustachian tube does not work as efficiently, with the resultant tendency towards the development of negative middle-ear pressure and the accumulation of fluid within the middle-ear space (OME). This tendency towards Eustachian tube dysfunction is further increased in children with CP as a result of dysfunction of the muscles originating from the palate which act to open the Eustachian tube orifice.[4,5]

A prospective longitudinal study following children between the ages of 1 and 5 years demonstrated that the overall prevalence of OME was 75% in children with cleft lip and palate compared with 19% in children without clefts.[2] This difference in prevalence of OME between children with and without clefts was also significant at individual time points throughout the study period. As well as being more common in children with CP, OME is likely to persist longer in children with CP. A retrospective longitudinal study of adolescents with various types of CP has demonstrated a decrease in prevalence of abnormal middle ears over time, with the decline in OME in patients with isolated CP occurring between 13 and 16 years of age.[6] Other studies have shown a similar decline in the prevalence of OME in late adolescence.[7,8] A questionnaire-based study of the natural history and outcome of middle-ear disease in children with CP reported that ear problems (ear infections and/or hearing loss) were most prevalent in the age range 4–6 years, only settling in adolescence, with 26% of the 13- to 15-years age group reported to have experienced ear problems in the preceding year.[9] However, 24% were still reported to have below-normal hearing when reaching early adulthood (16 years and above). Therefore, the prevalence of OME from early childhood into adolescence is an important factor when considering the optimum treatment strategy for OME in children with CP.

Otitis media with effusion commonly presents with hearing loss, but may also cause language delay, poor educational progress, recurrent ear infections, otalgia, behavioural deterioration, imbalance, tinnitus and hyperacusis. OME may also have a negative impact on quality of life (QoL) in affected children, with hearing being considered important at key stages in the development of language and behavioural and social relationships.[10]

Management options for otitis media with effusion in children with cleft palate

The diagnosis of OME requires a focused history, including information on the clinical features of OME and the general health and developmental status of the child. Clinical examination should include otoscopy, examination of the upper respiratory tract, tympanometry and an age-appropriate hearing test.

There are several possible approaches to the management of persistent OME in children with CP, which can be broadly divided into surgical, non-surgical and combination treatment.[4] The surgical treatment of persistent OME consists of the insertion of ventilation tubes (VTs, also known as grommets) into the tympanic membrane, which, while patent and in situ, prevent the development of the differential pressure

between the surrounding environment and the middle-ear space, thought to be an important factor in the pathogenesis of OME. VTs have recognised complications, which include persistent tympanic membrane perforation, ear infections and early extrusion.[1] Adjuvant adenoidectomy is not recommended in children with CP owing to the risk of velopharyngeal competence. Hearing aids (HAs) provide an alternative non-surgical treatment option for OME, with the aim of amplifying the sound delivered to the middle ear, compensating for the 'dampening' of the sound signal as it crosses the middle-ear space to reach the cochlea. HAs may also lead to ear infections and may not be considered cosmetically acceptable by a proportion of children and parents. Compliance with HAs in children with CP with or without cleft lip and OME has been reported to be only 52% (16/31 patients).[4] However, the same study reported otological complications in 5% (2/44) of children managed non-surgically and 38% of those treated with VTs, with the authors subsequently advocating VTs only in children not compliant with HAs or those who develop recurrent ear infections.[4] Combination treatment describes the scenario in which the chosen treatment strategy changes between surgical and non-surgical (or vice versa) owing to persistence or recurrence of symptoms.

In a systematic review directed at the early routine insertion of VTs for the management of OME in children with CP, the authors identified 18 eligible studies (case series, retrospective cohorts, prospective cohorts and randomised studies), but only one of these was a randomised clinical trial. This randomised trial had several significant methodological limitations which critically limited interpretation. The authors concluded that the majority of studies were small or of poor quality, with many having no formal sample size calculation, with the resultant risk of being underpowered to demonstrate a clinically important effect of treatment.[1]

When we consider outcomes, we see that studies have used diverse measures, mostly selected from clinicians' point of view and with limited consistency between studies. As OME can impair hearing at stages thought to be important in the development of language and behavioural and social relationships before the start of school, it could be suggested that outcomes relevant to these issues should be used in future studies. It is clear, therefore, that if further research into this treatment is to be commissioned, those outcomes relevant to parents and patients should be considered.

Guidelines for the management of otitis media with effusion in children with cleft palate

In 2008, the National Institute for Health and Care Excellence (NICE) published clinical guideline 60 (CG60), entitled *Surgical Management of Otitis Media With Effusion in Children*, which included a section specific to children with CP.[11] The guideline highlighted the particular problems posed by OME in children with CP, which included early onset, prolonged clinical course and higher rate of recurrence. For children in general, the guideline recommends that

> *Children with persistent bilateral OME documented over a period of 3 months with a hearing level in the better ear of 25–30 dBHL or worse averaged at 0.5, 1, 2 and 4 kHz (or equivalent dBA where dBHL not available) should be considered for surgical intervention.*

The recommendations for children with OME and CP were:

1.8.1 The care of children with cleft palate who are suspected of having OME should be undertaken by the local otological and audiological services with expertise in assessing and treating these children in liaison with the regional multidisciplinary cleft lip and palate team.

1.8.2 Insertion of ventilation tubes at primary closure of the cleft palate should be performed only after careful otological and audiological assessment.

1.8.3 Insertion of ventilation tubes should be offered as an alternative to hearing aids in children with cleft palate who have OME and persistent hearing loss.

The guideline also concluded that the evidence for a benefit of VT insertion in CP was lacking and that the optimal treatment for OME in children with CP had not been determined. In the absence of strong evidence, clinicians were recommended to base the management of OME in children with CP on the needs of the individual. Although the needs of each patient should be central to the decision-making process, there clearly remains a need to determine which treatment strategy is the most appropriate for these children.

Commissioning brief and objectives

The management of Otitis Media with Effusion in children with cleft palate (mOMEnt) study has been funded through a Health Technology Assessment (HTA) programme-commissioned call (project number 09/167/02) to address the uncertainty in the treatment of OME and to address the question 'What is the most appropriate way to manage OME in children with CP?' by completing a feasibility study.

Randomised controlled trials (RCTs) provide the highest level of evidence in the evaluation of health care. However, trials are expensive and require considerable additional effort from health-care staff and patients, which may create particularly high barriers to recruitment and successful completion of a study. From evaluating previous surgical trials, it appears that there are several challenges for a potential trial of care of OME in children with CP.[12] These concern feasibility, choice of comparator treatment, selection of relevant outcomes, and surgical compliance and skill. Furthermore, from the patient's point of view there may be difficulties with equipoise as surgical and non-surgical treatments have different risks.

The aim of our research was to provide information on the feasibility of carrying out a RCT or strong prospective cohort studies of the management of OME in children with CP. The project involved a set of studies and a value of information (VOI) study with the aim of identifying the optimum study design to add to knowledge of the treatment of OME in children with CP.

The study had the following components:

1. *Study Advisory Group (SAG)* We formed a SAG comprising clinical and methodological experts with nominations from the Craniofacial Society of Great Britain and Ireland. This included audiologists, speech and language therapists, and ear, nose and throat (ENT) surgeons. The SAG had the following specific roles: (i) to regularly provide advice for the study; (ii) to have an input into the design of the clinical survey and Delphi study; (iii) to advise on key parameters to explore the VOI analysis; and (iv) to have full input into the final exercise on feasibility.
2. *Clinician surveys* Surveys of clinicians were carried out to (i) identify the current UK practice for the treatment of OME in children with CP; and (ii) evaluate the feasibility of performing a RCT, or other relevant type of study, of VTs in comparison with 'usual methods' for the treatment of OME in children with CP.
3. *A qualitative project* The qualitative research project was designed to capture patient and parent opinions on willingness to take part in the trial, and to identify their needs for the content and form of information required to provide or withhold informed consent. Opinions on outcomes were also explored and data collected contributed to the core outcome set (COS) development.
4. *The development of a COS* A COS for a potential trial was developed. This would reflect the values of both providers and consumers of care.
5. *VOI analysis* This component was a VOI analysis that provided information on whether or not the extent of existing decision uncertainty about OME care for children with CP justifies the costs of the proposed research.
6. *Evaluation* The final part of the project was an evaluation of the data collected in the above components, in order to make recommendations on the feasibility of a potential study design.

Chapter 2 Clinician survey

Aim and objectives

The aim of the clinician survey was to collect data on the current clinical practice in cleft lip and palate centres in the UK, using a survey.

The main objective of the clinician survey was to collect information that would enable a decision to be taken on the feasibility of carrying out a trial or cohort investigation. As a result, within this report we are only including information on the following:

1. clinical provision and practice
2. method of delivery of care (centralised or 'hub and spoke')
3. caseload of the centres of children with non-syndromic CP.

A copy of the survey form is provided in *Appendix 1*.

Methods

We developed a survey form with the input of the SAG. This involved the preparation of drafts and a face-to-face discussion with the group, followed by development of the final form by e-mail 'discussion'. The form was piloted in two cleft lip and palate units and further changes were made. The survey form is included in *Appendix 1*.

We then approached the clinical directors of each of the cleft lip and palate networks and asked them to identify the lead ENT/audiology clinicians who could complete the forms. They were sent the survey electronically. We utilised several methods to obtain a high response rate to the survey. These included:

1. encouraging the clinical directors to discuss the study with the lead clinicians
2. contacting the clinicians by e-mail several times
3. telephone contact with the clinicians to discuss the form and the study.

When centres were unable to provide the full data set requested, we subsequently asked them to provide information on the three most important questions that were relevant to the decision regarding the potential study design (*Table 1*).

Information on the caseload of the centres of children with non-syndromic CP was collected from two sources. Firstly, the cleft network co-ordinators were approached and asked to provide data for their centre; secondly, the Craniofacial Anomalies Network (CRANE) database, a national database that includes data on the caseload and treatment outcomes of cleft centres in England and Wales, was consulted.

TABLE 1 Key questions for information needed to develop the potential study design

Question number in survey	Question text
2.5	What tests do you routinely use to diagnose and guide the subsequent management of OME?
2.13	What is your view on the optimum age for inserting VTs?
3.1	Do children attend cleft clinics outside your trust?

Results

The response rate for the survey is provided in *Table 2*.

The full data are included in *Appendix 2*.

The key responses received from each centre are outlined below for the associated survey questions.

Clinical provision and practice

2.1 Does your cleft service have dedicated audiology input based at your centre?

The majority of centres (9/10) completing this question indicated that they had access to a named health-care professional who undertook age-appropriate hearing testing in children with CP.

2.4 How often do children with cleft palate receive routine audiological assessment at your cleft centre? If assessment varies by age please give frequency of routine audiological assessment and age ranges

Details received would suggest that although testing regimes vary, most children undergo at least four hearing assessments by the age of 5 years (in addition to Universal Newborn Hearing Screening). Children may be seen more regularly, depending on clinical need.

2.6 At primary cleft palate repair, how is the decision made to insert ventilation tubes or not, and who is involved in the decision-making process?

The responses received indicate that it is not standard practice in the centres surveyed to sanction the insertion of VTs at primary cleft repair.

2.8 After what period of time would a conductive hearing loss (> 25–30 dBHL) trigger 'active' intervention (referral for/decision to insert ventilation tubes or prescribe hearing aids) at your centre?

Most centres would need evidence of persistence of OME over at least a 3-month period to recommend HAs or VTs. The response to this question would suggest adherence to the recommendations contained in NICE CG60[11] regarding a 3-month period of 'watchful waiting' prior to making a decision to recommend an intervention for OME.

2.9 Please describe the decision-making process to provide hearing aids or to insert/refer to ear, nose and throat for consideration of ventilation tubes as the first-line treatment for persistent otitis media with effusion. Please include any involvement of parents and/or the child

The responses indicated that patient choice is an important factor in the decision-making process, again adhering to NICE CG60,[11] which recommends that 'treatment and care should take into account children's needs and preferences together with those of their parents or carers'.

TABLE 2 Response rate to the clinician survey

Number of centres invited to complete the survey	16
Number of full responses received	10
Number of partial responses to an abbreviated questionnaire	14

2.5 What tests do you routinely use to diagnose and guide the subsequent management of otitis media with effusion?

All centres had access to a comprehensive set of audiological tests that would enable accurate assessment of frequency-specific hearing thresholds from approximately 6 months of age (visual reinforced audiometry 6 months to 3 years) through to adulthood (play audiometry 3–5 years, pure tone audiometry 4–5 years+). Tests were also available in certain centres that would enable threshold assessment in children < 6 months of age (auditory brainstem response) and assessment of other aspects of hearing, including the perception of speech.

This would indicate that all of the centres replying to the clinician survey were in a position to participate in a subsequent study to determine the most effective treatment for OME in children with CP.

2.13 What is your view on the optimum age for inserting ventilation tubes?

There was a spread of ages given in the responses received, with several centres indicating that clinical need was the more important determinant (*Table 3*).

The results for this question should be interpreted cautiously, as the wording of some answers suggested that the question asked for a minimum age for inserting VTs, as opposed to an optimum age. Although there was variance in practice, the majority of centres considered that the decision to insert VTs was either influenced by clinical need and not age (6/13 centres), or 1–4 years (4/13 centres). Only 3 out of 13 centres stated that the optimum age to insert VTs was under 1 year or over 5 years. It is likely that a subsequent study would include children (> 1 year old) of nursery, pre-school and school age. Therefore, with respect to age at VT insertion, the majority of centres would not be required to agree to a significant change in clinical practice.

Method of delivery of care: centralised or 'hub and spoke'?

Most centres (12/13) have a 'hub-and-spoke' clinics infrastructure, with the number of 'spoke' clinics ranging from 2 to 14. It should be noted that the one centre that stated it did not have any spoke sites indicated in response to a later question that it makes recommendations to clinics outside its cleft service.

The response to this question regarding structure of service was particularly relevant to the design and management of a future study, indicating that the majority of centres (12/13) had a hub-and-spoke infrastructure model. The number of spoke/outreach clinics varied and would have implications for any study design, especially randomisation, as well as standardisation of hearing testing and the need to obtain trust research and development (R&D) approval for multiple sites.

TABLE 3 Optimum age for VT insertion

Age	Number of centres ($n = 13$)
Under 1 year (at palatal repair)	2/13 (15%)
1–4 years	4/13 (31%)
5 years and above	1/13 (8%)
No optimum age/when clinical need dictates	6/13 (46%)

Caseload of the centres of children with non-syndromic cleft palate

Centres were unable to provide specific data on the number of non-syndromic patients only, and instead provided information on the total new referrals. This, unfortunately, appeared inaccurate according to the SAG. As a result, we have based our figures on yearly caseload on the data derived from the CRANE database. The advice from the CRANE database co-ordinator was that the caseload of non-syndromic CP/cleft lip and palate is approximately 75–80% of all registered cases. In generating these data, they assumed that this proportion was uniform across centres (*Table 4*).

CRANE does not collect information for Scottish centres and so we have used the data directly from the centres, as this seemed logical to the SAG. We have calculated an estimation of the number of patients per year who would enter a trial based on the number of patients who are likely to have OME (90%), then taking a conservative estimate of those who would meet trial eligibility criteria (50%), and finally factoring in the predicted consent rate, estimated from the qualitative research described in *Chapter 3* (25%).

TABLE 4 Yearly caseload of the centres represented by new referrals in 2012, and estimation of numbers who would be recruited into a trial

Centre	Number of new referrals (number of children with CP with or without cleft lip)	Estimate of numbers who would be recruited into a trial
Newcastle	65 (49)	6
Leeds	65 (49)	6
Liverpool	64 (48)	6
Manchester	69 (52)	7
Nottingham	93 (70)	8
Birmingham	121 (91)	10
Cambridge	87 (65)	8
North Thames	173 (130)	14
Oxford	45 (34)	4
Salisbury	53 (40)	5
Swansea	51 (38)	5
Bristol	65 (49)	6
South Thames	145 (109)	12
Belfast	31 (23)	3
Edinburgh	29 (22)	3
Glasgow	46 (35)	4
Total	1202 (902)	107

Discussion

The survey has provided useful information for the potential study design. However, the response rate was disappointing in that only 10 out of the 16 centres provided us with a full response. We made multiple efforts to engage with the clinicians; this included liaising with the clinical director of each network/centre, multiple e-mail contacts and reminder telephone calls. In spite of these efforts, our data set is not complete for all networks/centres.

The low response rate may be due to variation between centres in the method of delivery of care and structure of the service. For example, although 90% of those responding had access to an audiologist within their cleft team, this was not the case for all centres. Importantly, in those centres where there is no dedicated audiologist/ENT surgeon, the patients are referred to a general paediatric clinic. This made it difficult to identify the appropriate clinician to respond to the survey. We did make efforts to identify if these issues were relevant to centres that did not respond completely, but we found that information was very limited.

It is therefore clear that when designing a future study, the structure of the clinical team at each centre/network and the engagement with the current study are important considerations when identifying sites to participate. One option would be to only approach those sites that provided a good response to the present study.

The clinician survey has highlighted several key factors for the design and delivery of a subsequent study, and has suggested that UK cleft centres are in a position to participate in a study to determine the most effective treatment for OME in children with CP. The results indicate that centres would be able to nominate a lead ENT/audiologist for a study to act as local primary investigator and that the centres are able to perform age-appropriate hearing tests from 1 year of age through to adolescence. The survey also suggested that centres adhered to the 3-month 'watchful waiting' period prior to considering an intervention for OME, as recommended by NICE CG60,[11] and children were seen regularly for audiological assessment up to the age of 5 years. The majority of centres considered either the period from 1 to 4 years of age or any age based on clinical need to be the appropriate time for insertion of VTs. Therefore, a subsequent study is likely to be more readily acceptable to centres if it uses the criteria for intervention as recommended by NICE CG60, recruits patients within the first 5 years of life and concentrates testing within the same period to minimise additional clinic visits. The importance of parental opinion in the decision-making process regarding OME management was emphasised, and this has implications for the information provision contained in any study design.

The method of delivery of care was important for potential study design. It is clear that most of the cleft networks operated a hub-and-spoke infrastructure for clinics, in that the patients were seen at the centres, but their audiological/ENT care was provided in local clinics and hospitals. This has several important implications. Firstly, it would be difficult to engage peripheral clinicians with the random allocation of care as they may not be in equipoise, and the probability of protocol deviations would be high. Furthermore, obtaining trust R&D approval for multiple sites with potentially low caseloads would be problematic and inefficient. Finally, there will be the additional problem of standardising both audiological assessment and treatment away from the hub clinic. The potential numbers of eligible patients for recruitment were provided in *Table 4*, with the recruitment rate and required recruitment period being influenced by the study design and sample size.

Chapter 3 Qualitative interviews with parents and children with cleft palate

Background

There has been very little qualitative research on treatment or living with CP from the perspective of either parents[13] or children,[14] and none related to OME.

Aims

The aims of the qualitative interviews were to explore in depth (a) parents' views about their willingness for a child to take part in a potential trial comparing VTs and HAs, and (b) outcomes of the management of OME considered important by parents and children.

Methods

A qualitative methodology was adopted to enable individuals to recount experiences in their own words, highlighting what is important to them.[15] We focused initially on descriptions provided by participants, but, as the study progressed, took a more interpretive approach to data, in line with the principles of framework analysis.[16]

Participants

Parents were recruited from two cleft centres in northern England. They were eligible to take part if they had a non-syndromic child with CP (including cleft lip and palate) between 0 and 11 years of age, who had a current or past diagnosis of OME. This age range was selected because it is the common time period for children to experience OME, as reflected by the NICE[17] guideline on management of this condition, which is specific to the care of those aged under 12 years. Families with particularly difficult social circumstances (e.g. domestic violence, recent bereavement) were not approached. Children aged 6–11 years were interviewed, if they were happy to talk to the researcher. We felt that children younger than this would have difficulty expressing their thoughts on the research topic. Participants had to be able to converse in English. The interviewer was a researcher who did not have a clinical background and was not involved in participants' care.

A purposive approach to sampling was taken to ensure variation in terms of children's age, treatment experiences for OME and gender. A sampling matrix was developed for this purpose[18] to guide recruitment as it progressed. We intended to recruit parents of approximately 30 children with a range of treatment experiences, including VTs only, HAs only, both VTs and HAs, and neither VTs nor HAs (the watchful waiting group). Initially, any parent meeting the inclusion criteria was invited to take part. As recruitment progressed, practitioners were asked to identify specific individuals to ensure variation in the sample. Data collection continued until the sample was diverse in terms of children's age, gender and treatment experiences, and it was judged that data saturation had been reached.

Procedures

In unit A, a designated member of the cleft team screened clinic lists and medical notes on a regular basis for potential participants due to attend. Children who had a CP and a clinical history of OME but no concurrent syndrome were identified. The researcher was informed in advance when eligible participants had an appointment, and visited the unit on these dates. A member of the cleft team talked to the

identified parent and asked if he or she was happy to meet the researcher. If he or she agreed, the researcher introduced herself and gave the parent a copy of the participant information sheet. She also took a telephone number and called a day or two later to see if the parent was willing to be interviewed. As recruitment progressed, the clinic lists were not screened; rather, the researcher would attend on days when she might capture individuals missing from quota matrices (e.g. clinics for 10-year-olds).

In unit B, a designated member of the team screened clinic lists and patient notes to see whether or not eligible parents were due for an appointment, using the same criteria as for unit A. These individuals were sent a participant information sheet in the post. The researcher would visit clinic on dates when people identified as possible participants were attending. A member of the cleft team checked that parents were happy to talk to the researcher. If this was the case, she introduced herself and asked if they had received information in the post. When parents stated that they had and were happy to take part, a time and date were arranged for the interview. Sometimes parents said that they had not received information through the post or had not had time to read it. The researcher would give these parents a copy of the information sheet, taking a telephone number so that she could call them a day or two later to see if they were willing to be interviewed. As recruitment progressed, the practitioner screening clinic lists was advised to identify individuals who contributed to cells of the quota matrix that were lacking in numbers. For example, over time patients who had received HAs only were targeted.

Modified versions of information sheets, with simpler language and less text, were developed for children aged 6–7 years and a slightly more detailed version for 8- to 11-year-olds. These were given out at clinic in unit A or sent in the post for unit B along with study invitations to parents. Information sheets were piloted with families attending unit A in advance of data collection, and revised in light of their comments.

Data collection

In line with the qualitative methodology, semistructured interviews were conducted to gather data on views and experiences. Interviews took place at a time and place convenient to participants (mostly in their home, see *Results*) between March and August 2012. They were recorded with parents' consent and transcribed verbatim for analysis, with identifying features removed during this process (including names of health-care professionals).

A topic guide was developed for parents, based on relevant literature and discussion among the research team in relation to the project's aims (see *Appendix 3*). Interviews took the form of a conversation in which parents initially told the story of their child's OME, prompted by questions including:

1. When did you first notice a problem with your child's ears? What alerted you?
2. What information did you receive about different treatments for glue ear?
3. What made you choose [treatment] for your child?
4. How satisfied were you with treatment your child received?
5. What would you advise other parents about treatment for glue ear?

About midway through an interview, parents were invited to reflect on important results (outcomes) of treatment for OME, which were recorded on electronic 'sticky notes' on a tablet computer. Parents were able to move these around to demonstrate their importance; they were encouraged to elaborate on reasons for items they had listed and the order they placed them in. Towards the end of an interview, parents were introduced to the concept of a RCT comparing treatments for OME and asked for their thoughts on whether or not they would allow their child to be part of such a study.

The topic guide was revised as data collection progressed to incorporate additional topics raised during interviews. For example, parent 1 talked about the difficulties she found with obtaining a regular supply of batteries for her child's HA; hence, subsequent parents of children who had HAs were asked specifically about battery supplies. Likewise, parent 2 mentioned struggling to understand feedback she received from audiology after her child's hearing tests; this was added to the topic guide as an area for exploration in other interviews.

The first child to be interviewed was given the option of whether he wanted to talk to the researcher before or after his parent. He opted to go first. However, after interviewing his mother, the researcher had a better understanding of the child's condition and a greater awareness of his character, interests and likes. Therefore, subsequent interviews with children tended to be carried out after data collection with their parent(s). This approach had specific advantages; as well as allowing more details to be gathered about the condition's history, it enabled the child to (a) see their parent(s) interacting with the researcher, (b) become familiar with the researcher's presence and (c) observe the conversational tone and format of the interview.

Children were interviewed separately from parents to avoid the difficulty of disentangling individual perceptions in joint interviews and the potential for children to sense that they should agree with parents.[19] Parents were in the same room or an adjacent one when a child's interview was being conducted. Overall, children responded well to questions posed, but sometimes parents added comments to statements made or elaborated when a son or daughter struggled to verbalise his or her thoughts; this is something that others have noted to be helpful when gathering qualitative data from children.[20]

Interviewing parents and children separately was necessary because a different approach to data collection was used with the latter. There is a wealth of literature on how to conduct investigations with children that aims to offer an in-depth understanding of their experiences or views. Within such work, a recurring theme is the need for participatory techniques, including songs, drawings and stories, because children are said to communicate better through such media,[21] and the need for creativity in how data are collected.[22] We prepared a range of activities to engage children and to maintain interest among those with limited concentration.[23] Most were carried out on a tablet computer. For example, interviewees were shown a picture of a child and informed that this individual had just been told that he/she had glue ear. This indirect approach reduced the need for personal disclosure from the child straight away. They were then asked questions about how the child in the picture might feel about different treatments and to complete speech bubbles on the tablet computer to show what this child might be thinking. Activities were used as a starting point for discussion on areas relevant to the study's aims. They were piloted with a group of children without clefts on the topic of healthy eating, to see which appeared best at facilitating conversation with the researcher. Interviewees enjoyed playing on the tablet computer, which made data collection a fun event. All the children could use this device, even if they had not seen one before. Questions asked when carrying out activities included:

1. Can you tell me about any problems you've had with your ears?
2. How do you feel when you have to go and see the doctor about your ears?
3. What's the good thing about having grommets/HAs?
4. What's not so good about having grommets/HAs?

Analysis

Framework analysis was applied to interview data.[24] This allows for the sharing of information within a team, by summarising data into charts. It is suited to applied qualitative research that has specific questions and objectives,[25] and provides a clear record of how ideas moved from participants' words to final findings.[26] Framework analysis is divided into five stages: (1) familiarisation with the data (becoming immersed in material collected); (2) development of a thematic framework (identifying key issues in the transcripts), which involved constantly comparing emerging codes and categories with original data, across all cases; (3) indexing data (labelling key issues that emerge across cases); (4) devising a series of thematic charts (allowing the full pattern across cases to be explored and reviewed); and (5) mapping and interpreting data (looking for associations, providing explanations, highlighting key characteristics and ideas). It facilitates either theme-based or case-based analysis, or a combination of the two, through the development of charts that can be read across rows (cases) or down columns (categories). It also allowed us to explore data based on specific interviewee characteristics, such as the child's treatment experience or age.

Three researchers and three clinicians (surgeon, consultant in audiovestibular medicine, orthodontist) formed the analysis team. PC and ST led the process, meeting approximately once a week during data collection to

discuss what participants were saying, consider areas to follow up in later interviews and debate emerging ideas. The analysis team came together halfway through data collection. Before this meeting, each member was given four to six interview transcripts to review and identify potential codes, which were discussed as a team. ST used ideas from this meeting, and her knowledge of the entire data set, to develop a thematic framework in consultation with PC. This was shared with the team for their comments via e-mail before being used to index all interview data within the qualitative computer package NVivo 9 (QSR International, Warrington, UK). At the descriptive stage of analysis, separate thematic frameworks were developed for parents and children, to ensure that children's views were not lost among those of parents, whose more articulate expression could have dominated a single thematic framework for the entire data set.

Once all transcripts had been indexed using the thematic frameworks, ST charted data, again in NVivo 9. This involved summarising what participants had said in relevant cells of a chart (*Table 5*). PC checked 10% of transcripts and agreed how ST had indexed data overall; any disagreements were resolved through discussion between ST and PC. Charted data were sent to the analysis team in advance of a second meeting to talk about these summaries and to start interpreting data, a process that ST and PC continued in follow-up analysis sessions. To illustrate aspects of the analysis, we have included direct quotations from participants in this chapter. We have not used names, to avoid identification. Numbers are employed when referring to the sample, reflecting the order of data collection, with 'C' denoting a child interviewee, 'P' a parent, (m) a mother and (f) a father.

TABLE 5 Part of one of the charts produced for the analysis, on the category 'service provision', which had the four subcategories listed

	Sequence of care	Glue ear vs. other aspects of cleft care	Hearing tests	GP's role in treating glue ear
P25 4-year-old child Male Unilateral CLP VTs and HA Centre A	Tests when born suggested hearing was fine. Then at 18 months diagnosed with glue ear. Told from early on hearing could be problem but hard to take everything in then – focus on feeding. Happy when first tests came back OK – felt one less thing to worry about. Did not realise hearing could become a problem later on. Felt 'devastated' when told son had hearing problems. With HAs can hear as long as are in. So in bed and in morning cannot hear until put in	Had to make decision whether to have second set VTs or HAs. With cleft no decision to make – just go with what doctors advise	Just went for routine hearing test – did not think there was a problem but told there was. 'Devastated' with the news because thought would interfere with his speech	
P12 4-year-old child Male Unilateral CLP Watchful waiting Centre B	Aware a number of problems associated with cleft, including hearing, even before the birth. Told at time about VTs that it was just a little operation that could help with this	Child went through so much with palate operations – at start did not really see ears as major, especially since everyone gets ear infections. But now is having ear infections all time and been through all major palate operations, mum's concerned with ears and how these might affect child at school	Not had a hearing test for about 1 year. At last test, clinicians were impressed with child's hearing and could not see a problem but since then he has had a number of ear infections. Coped well with hearing tests because some play involved	Goes to doctors as soon as child seems to have ear infection. GP gives antibiotics. Tends to clear up but then returns. Feels nothing else has been offered to stop infections recurring. Going to speak to GP next time child has infection to see if VTs would help with this as it has affected child's sleep. But not had an infection for about 4 months

CLP, cleft lip and palate; GP, general practitioner.

Ethical considerations

Ethical approval for the qualitative interviews with parents and children was obtained from the National Research Ethics Service North East Committee – Greater Manchester East (reference 11/NW/0586). Approval was also obtained to further contact participants with an invitation to take part in the final consensus meeting described in *Chapter 4*.

All parents gave informed written consent to their participation and the use of quotations from their interviews for dissemination purposes. They also consented to the involvement of children aged ≥ 6 years. Assent was obtained from children, who were made aware from the outset that there were no wrong or right answers, that data collection was a confidential process and that the researcher was not coming to provide treatment, but just to ask about their views and experiences. They were given the option of saying 'no' to taking part, even if their parents had consented to their involvement. To put children at ease, at the start of the interview the researcher showed them the digital recorder. She gave them the chance to take on the role of interviewer, inviting them to ask her any question they wanted or to choose a question from a selection she had prepared. This gave them the opportunity to understand how the recorder worked and introduced them to the conversational tone and form of the interview. Participants (parents and children) had the opportunity to ask questions prior to starting data collection and at the end of the interview. When the interview finished, children were asked to indicate how they felt by selecting one of a range of cartoon faces showing different emotions (e.g. happy, sad, angry, confused). There was a general sense of happiness at being listened to and being able to possibly help other children.

Rigour

Based on guidelines for producing good-quality qualitative research, we employed the following strategies.[27,28]

- Reflexive notes were made during data collection by ST. In these, she recorded contextual information relating to where interviews took place and emerging ideas relating to analysis.
- Data were sought from a diverse group of individuals, in terms of factors thought to be pertinent to experiences of OME (e.g. age, type of treatment).
- More than one person was involved in the analysis; the analysis team comprised members with different experiences in terms of the care of children with CP.
- We looked for disconfirming data while developing themes to deepen the analysis and to ensure all aspects of transcripts were considered.

Results

Interviews were conducted with 37 families (five from minority ethnic groups). Twenty-eight were recruited from unit A and nine from unit B. This represented a 71% response rate among those invited to participate, as shown in the flowchart in *Figure 1*. After comparing narratives from those in units A and B, no obvious differences were identified. Therefore, data from both sets of interviewees were combined within the analysis.

Twelve parents were interviewed as couples, while one father and 30 mothers were interviewed on their own. The mean age of parents was 34.9 years [standard deviation (SD) 6.7 years]. Data were collected from 22 children, comprising 13 boys and 9 girls; two children aged 6–11 years did not want to take part, so only their mothers were interviewed. The mean age of children interviewed was 8.8 years (SD 1.3 years). The type of treatment and cleft experienced by children is illustrated in *Tables 6* and *7*. The most difficult group to identify was children with experience of HAs only, especially those in the younger age group. It appeared that VTs had often been inserted at a young age during an anaesthetic for another cleft procedure. In addition, children who had received HAs only were often syndromic and, therefore, not eligible to participate.

Interviews with parents lasted between 20 and 65 minutes (average 40 minutes). Those with children ranged from 10 to 40 minutes (average 20 minutes). Most were conducted at a participant's home but five interviews with parents and three with children were conducted in clinic, at the parents' request.

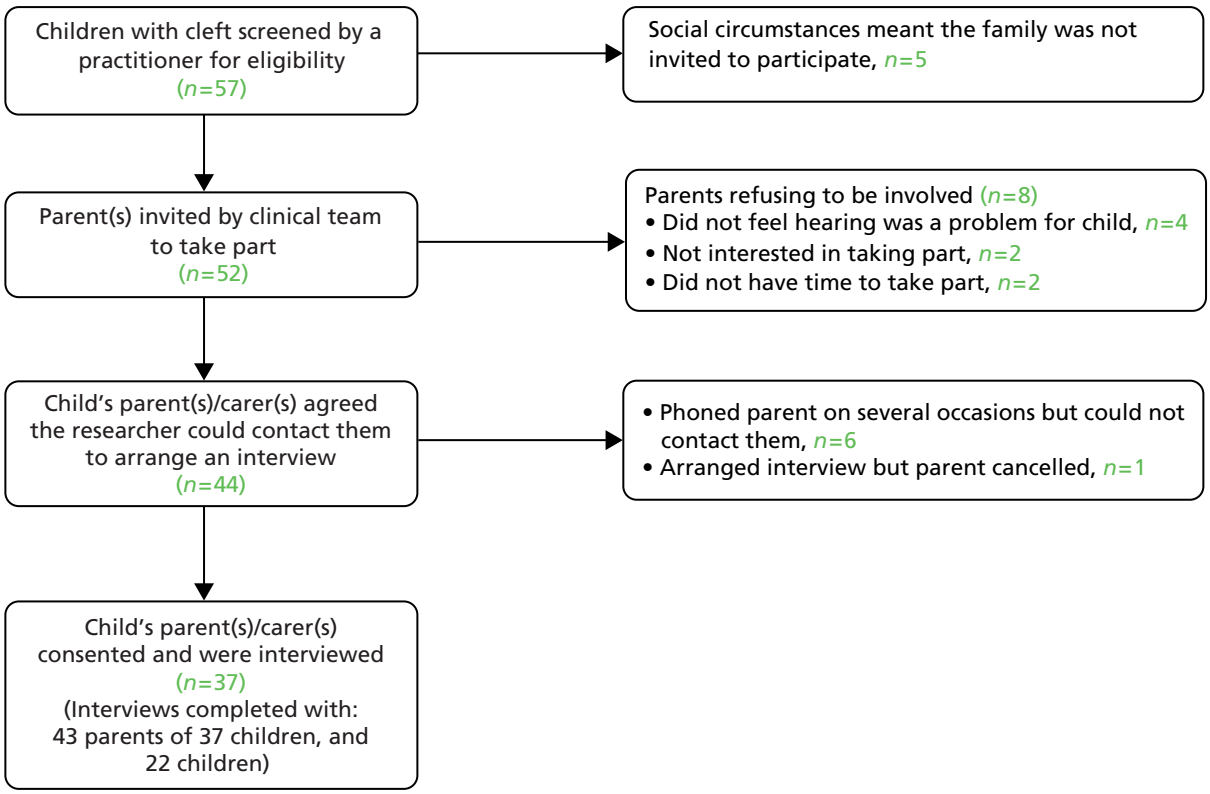


FIGURE 1 Recruitment to the study.

TABLE 6 Participating families with a child aged 0–5 years (no children interviewed)

	Parent(s) interviewed		Cleft type			Treatment experience			
Interview number	Mother	Father	UCLP	BCLP	CP	VTs	HAs	Both	WW
2	✓			✓					✓
4	✓			✓		✓			
6	✓	✓	✓			✓			
8	✓				✓	✓			
11	✓		✓						✓
12	✓		✓						✓
13	✓		✓					✓	
15	✓	✓			✓			✓	
19	✓	✓			✓				✓
20	✓		✓						✓
21	✓				✓	✓			
23	✓				✓				✓
25	✓		✓					✓	

BCLP, bilateral cleft lip and palate; UCLP, unilateral cleft lip and palate; WW, watchful waiting.

TABLE 7 Participating families with a child aged 6–11 years (C29 and C30 not interviewed)

	Parent(s) interviewed		Cleft type			Treatment experience			
Child number	Mother	Father	UCLP	BCLP	CP	VTs	HAs	Both	WW
1	✓		✓					✓	
3	✓				✓			✓	
5	✓				✓			✓	
7	✓			✓				✓	
9	✓			✓				✓	
10	✓				✓			✓	
14	✓				✓	✓			
16	✓				✓				✓
17	✓	✓			✓	✓			
18	✓				✓			✓	
22	✓	✓			✓	✓			
24	✓		✓					✓	
26	✓		✓			✓			
27	✓			✓			✓		
28	✓	✓			✓	✓			
29	✓				✓	✓			
30	✓				✓		✓		
31	✓		✓			✓			
32	✓				✓	✓			
33	✓				✓		✓		
34	✓		✓				✓		
35		✓	✓						✓
36	✓		✓			✓			
37	✓			✓			✓		

BCLP, bilateral cleft lip and palate; UCLP, unilateral cleft lip and palate; WW, watchful waiting.

All but one interview was digitally recorded and transcribed verbatim; one mother requested that her interview was not recorded but was happy for the researcher to take written notes.

For parent interviews, the team identified 139 initial codes through the familiarisation stage of analysis. These were clustered into 10 categories (service provision, family and social life, VTs, HAs, ear infections, communication, child's education, parents' involvement, outcomes from treatment, views of being part of a trial), each of which had subcategories. For child interviews, 54 initial codes were collapsed into six categories (everyday life and hearing, VTs, HAs, ear infections, clinical encounters, school); again, each of these had subcategories. *Table 8* shows how data from initial codes relating to parents' views of HAs were combined to create four subcategories. Categories and subcategories formed the indexing scheme that structured the summarising of data into charts. These charts were then used to describe and interpret interviewees' words.

As mentioned above, two main areas were explored during the interviews: willingness to enter a child into a RCT and outcomes of importance for children and parents following management of OME. Given that these are distinct topics, results and a discussion of what was found relating to involvement in a trial is followed by results and a discussion of what was found about perceptions of outcomes.

TABLE 8 Illustration of development of the category 'HAs' and five subcategories from transcripts

Initial codes	Subcategories	Examples from transcripts
• Impact on hearing • Impact on speech	Impact on hearing (to include impact on speech)	*. . . when she put them in his ears for the first time and turned them on his face he was like that [pulls a face] and it was only about a year ago I think, maybe 18 months ago that he heard a microwave ping for the very first time 'cause obviously it's all trial and error with hearing aids, getting the right frequencies . . .* *P1 (m)* *As soon as we got that [HA] it did, it did improve his speech. It has improved his speech since we got it 'cause he was hardly talking. He's still not, I wouldn't say he's at the age of a 3-year-old speech-wise cause his little friends in school are all talking a lot better and are a lot more clearer as well.* *P15 (m)*
• Reminder of child's hearing problems • Image • Mark child out as different • Bullying and social stigma	Visibility	*I know a lot of parents who wouldn't have hearing aids because of social reasons, people pick up on oh he's deaf or whatever, whereas grommets they're not seen, nobody has a clue.* *P2 (m)* *She was the only one with hearing aids so everybody loved them and we sent books in to school so she could read them with her friends to show them that this is what I'm going to get and everybody was dead excited and all of her friends keep saying 'I want hearing aids'.* *P13 (m)*
• Seen as a sign of hearing deteriorating • Just one more thing for child to deal with • Children coping better than parents thought • Not treating underlying problem of fluid build-up	Parents' beliefs about HAs	*. . . it [HA] looks like she's relying on something, yeah rather than trying hard like for herself but obviously if it does affect her hearing during the class and she does need one then as I said you can't see it from outside, I think it's just for us as her parents, we were worried that it may not be comfortable for her, that's all.* *P11 (m)* *. . . if you put a grommet in it's opening the tube so the tube, the fluid can drain, whereas these [HAs] are just, well it's just making the hearing a little bit better. It's not, so it's still bunged up. It's still, the eardrum is still as flat as anything. It's not solving anything . . . it's just assisting.* *P27 (m)*
• Getting child to wear • Parents' normalising HAs	Getting child to wear	*It was just whether he would tolerate them, you know, for a long time because he's a wee bit of a fussy wee boy [laughs]. He doesn't like anything really that interferes with him and I just was worried that perhaps maybe he wouldn't wear them or maybe he would just wear them for a week and then decide that these weren't for him.* *P4 (m)* *. . . he only wears the one, he only wears it at school. He doesn't wear it at home or anything, he doesn't really need it at home, he can manage watching the telly or listening to us. So I've never made him wear it at home.* *P33 (m)*

TABLE 8 Illustration of development of the category 'HAs' and five subcategories from transcripts (*continued*)

Initial codes	Subcategories	Examples from transcripts
• Production of HAs • Frequency of HA appointments • Customising • Replacements and repairs • Different types of HAs	Supplies and maintenance	*I soak them twice a week in warm water and . . . we have to make sure there's not water in the tubes, so it's using a puffer and puffing the water out. It's not too bad.* *P3 (m)* *He doesn't bother about the hearing aids 'cause they're snazzy aren't they. At the minute he's got like red and yellow in and last time he had stickers in them and so he can do what he wants with them really . . . The batteries, through no fault of anyone, you just forget and you'd think oh I need to get batteries. They're so tight. You think well, you go in and say can I have some batteries, for instance, [child] has school, here and his dad's. So we leave them at all places but they'll give you like one packet and you're like oh great [sarcastic tone], you know, but no getting them is fine.* *P37 (m)*

Analysis strand 1: parents' views about their child's participation in a potential trial

This section describes parents' comments about whether or not they would allow their child to be part of a trial comparing VTs and HAs, and factors influencing their decision. It covers views of randomisation and explores possible barriers to recruitment. We focus on the opinions of mothers and fathers rather than those of children because they would make the ultimate decision of whether or not to participate. In addition, it was felt that children would struggle to understand the concepts of randomisation and equipoise. Parents were asked to state whether or not they would allow their child to take part in a trial to test the best method of treating OME. However, one mother (P9) was not asked because she expressed negative views about VTs with such emotion that it was inappropriate to explore if she would enter her son into a study where there was a 50% chance of receiving this treatment. An outline of how the topic was approached within the interview is shown in *Box 1*.

BOX 1 An example of the way in which the topic of being in a trial was broached with parents

Interviewer: *At the moment it's not clear what treatment is best for glue ear. We would like to do a trial comparing two different treatments. The best way to do a fair test between two types of treatments is for there to be an equal chance of children receiving treatment A or treatment B. This could be done by a computer programme or by rolling a dice – for example, if an even number comes up the child receives treatment A and if an odd number comes up they receive treatment B. If a parent agreed to let their child be part of this type of trial it wouldn't be a doctor who decided what treatment they received or the parent, and the child would have an equal chance of receiving treatment A or B. What they did receive would be down to chance. What are your views of letting your child be part of such a trial if [child's name] got either treatment A or treatment B by chance?*

(Invariably parents would ask which treatments at this point, so the researcher mentioned VTs vs. HAs.)

Follow-up questions: (a) What made you say [yes, no, unsure]?; (b) Is there anything that would change your view?; (c) What would you want to know before you decided?

Parents' willingness to enter their child into a trial comparing ventilation tubes and hearing aids

In nine parent interviews, participants stated that they would allow their child to be part of the trial, whereas in 19 the answer was negative, and in eight, participants were unsure what they would do. *Table 9* groups interviewees based on their response to taking part in a trial ('no', 'unsure', 'yes') and summarises key factors influencing their decision-making as recorded in interview transcripts. Patterns which emerged on how individuals responded are shown in *Table 10*.

Most interviewees were reticent about entering their child into the proposed trial, although they recognised the need to advance scientific knowledge. Some of their reluctance stemmed from concerns about not being able to choose treatment and a risk of their child not being allocated to the most effective arm. Furthermore, several parents had pre-existing views about the benefits or drawbacks of VTs or HAs, as suggested in the following interview extracts:

> *Urm, possibly not, just because if she then fell in the grommets group, she would have to have an operation and I wouldn't want her to go under general again just for grommets. So probably not.*
>
> *P7 (m)*

> *I've had the results and I've witnessed it and he was a changed child. He could hear perfectly well. I mean he went for his hearing test after his grommets and he was passing them with flying colours . . . So no I wouldn't be happy with that and I wouldn't want a hearing aid because it's there, it's on view, children will poke at it and say 'what's that in your ear?' and it's the sheer embarrassment for a child, I would say no, absolutely no way.*
>
> *P8 (m)*

Hence, interviewees saying 'no' or 'unsure' did not see the two treatments as sufficiently equivalent to accept randomisation and did not appear to feel that risks associated with one intervention were warranted. Some parents expressed fears that there could be social consequences of HAs, including the potential for bullying:

> *Just the stigma, the stigma with hearing aids isn't it. It's, you don't want anything that anyone can say to, to the chances of your child being bullied, being picked on and you know yourself it's like kids and it's just one more that they can, he's not a very confident child, you know you worry that he, that's why we were concerned whether his speech, would that cause bullying and stuff like that. It's just a stigma isn't it really, it's like anything.*
>
> *P16 (m)*

Others had strong views about physical risks that they associated with VTs (e.g. causing damage to the ear or infections):

> *I think she was about 1 when she had her first set of grommets that they said would help sort out the glue ear and that unfortunately made things worse . . . I think the hearing aids have actually helped more than the grommets ever did because we haven't had an ear infection for over a year . . . They [VTs] never worked. She'd be ill, she'd be off school. We'd get phone calls saying she wasn't in but it wasn't our fault.*
>
> *P24 (m)*

Participants were divided in terms of their favoured treatment, including parents of children in the watchful waiting group (*Table 11*). Hence, even when interviewees had no personal experience of VTs or HAs, they could still hold strong opinions about these approaches, informed by conversations with friends or relatives, media outlets or social norms. Parents with children who had received VTs only tended to prefer this approach.

TABLE 9 Parents' views about being asked to be part of a trial and reasons for their response

	Parents saying 'yes'									Parents saying 'no'																		Parents who were 'unsure'							
	Interviewee identifier																																	
Reason for response	1	12	18	19	22	26	27	28	31	4	6	7	8	10	11	14	15	16	17	20	21	23	30	32	33	34	36	37	2	3	5	13	24	25	29	35
Trust in medical professionals	✓			✓						✓							✓														✓			✓		
Benefits to child of being in trial					✓				✓																								✓		✓	
Altruism/advance knowledge	✓	✓	✓	✓		✓	✓	✓																						✓	✓	✓	✓			✓
Want control/choice/options[a]											✓				✓	✓	✓	✓	✓			✓	✓		✓	✓		✓	✓		✓	✓				
Child not getting best treatment										✓	✓						✓	✓	✓		✓	✓							✓							
Preference stated for VTs										✓			✓											✓										✓		
Preference stated for HAs																	✓						✓							✓				✓	✓
Drawbacks related to VTs												✓		✓						✓			✓		✓	✓		✓		✓	✓	✓	✓		✓	✓
Drawbacks related to HAs[b]															✓	✓			✓					✓			✓							✓		

a Includes not liking the idea of the child being a research object.
b For example, getting child to wear HAs, maintaining them and potential bullying.

TABLE 10 Patterns of responses regarding taking part in a future trial

Those saying 'no'	Those who were 'unsure'	Those saying 'yes'
• Wanted control/choice/options • Drew on previous experience when expressing a preference for either VTs or HAs, or concerns about VTs or HAs • Worried their child may not get the best treatment if randomised • Did not mention potential benefits for child of being in a trial • Did not refer to altruistic reasons for taking part	• Wanted control/choice/options • Drew on previous experience when expressing a preference for either VTs or HAs, or concerns about VTs or HAs • Acknowledged a clash between altruistic beliefs and a wish to protect their child • Considered potential benefits for the child of being in a trial	• Did not mention wanting control/choice/options • Did not appear to draw on previous experience; they did not express a preference for VTs or HAs, and did not focus on possible negative aspects of VTs or HAs • Tended to express a wish to help others or to advance knowledge

TABLE 11 Parents' preference for one treatment or another (divided based on treatment received by their child)

Interviewee	Prefer VTs	Prefer HAs	No preference	Not clear
Child had VTs				
P4	✓			
P6	✓			
P8	✓			
P14	✓			
P17	✓			
P21	✓			
P22			✓	
P26			✓	
P28			✓	
P29		✓		
P31				✓
P32	✓			
P36	✓			
Child had HAs				
P27		✓		
P30		✓		
P33		✓		
P34		✓		
P37			✓	

TABLE 11 Parents' preference for one treatment or another (divided based on treatment received by their child) (*continued*)

Interviewee	Prefer VTs	Prefer HAs	No preference	Not clear
Child had VTs and HAs				
P1			✓	
P3			✓	
P5			✓	
P7		✓		
P9		✓		
P10		✓		
P13		✓		
P15		✓		
P18			✓	
P24		✓		
P25	✓			
Child in watchful waiting				
P2	✓			
P11	✓			
P12	✓			
P16				✓
P19			✓	
P20				✓
P23			✓	
P35		✓		

Just one person in this group expressed a preference for HAs; P29 did not think it was fair to put her child under anaesthetic when the VTs kept falling out. Most parents with a child who had experienced HAs only described being encouraged to try them by a health-care professional. They expressed a preference for these devices in part because they eliminated the need for anaesthetic, which they worried could be required on several occasions if VTs fell out. These parents talked about their own struggles witnessing their son or daughter being anaesthetised. They also mentioned their child's difficulties with surgery:

> *. . . he had a bad reaction . . . he was being sick . . . it was just horrible . . . I didn't want to put him through another operation no matter how big or small.*
>
> *P33 (m)*

> *. . . we've had a lot of battles with her going for surgery . . . she goes to the play specialist a couple of months before surgery . . . cannulas . . . they are the major issue with her.*
>
> *P34 (m)*

It was notable that though interviewees with children who wore HAs said they had been worried about teasing, these fears had not been realised; most stated that their child coped better wearing HAs than they had expected. As for those whose children had tried both treatments, HAs tended to be preferred as a result of poor previous experiences with VTs (e.g. falling out, repeated insertions, ear infections that parents attributed to VTs). Of those receiving both, most children had received VTs followed by HAs; just C13 and C18 had HAs first. Only P25 in this group of parents preferred VTs because, unlike HAs, they allowed for a constant improvement in hearing while in place:

> *So sort of when you go to bed they're not in and when you get up on a morning they're not in so obviously you've got that lull of whereas it's like putting contact lenses in, you can constantly see rather than put your glasses on you know that type of thing, that was the only thing in my head to compare it to . . . the grommets give you a more rounded hearing cause it's always there as opposed to just when they're in.*
>
> *P25 (m)*

Views on the presentation of information about trial participation

Interviewees stated that prior to deciding whether or not to allow their child to be part of the proposed trial, they would like to talk to a researcher about it and wanted written information which they could take home and reflect on with family members. They also suggested that information should be provided to children if they were old enough. Participants stated that the information they would like to help make a decision included:

- *General* That neither treatment would make the child's situation worse, potential benefits and drawbacks of each treatment and what might happen if either did not work.
- *HAs* What this would involve for parents and how often they would have to take their child to get new moulds fitted.
- *VTs* What might go wrong, how many sets would be inserted, the chances of them falling out and any after effects.

Some parents were clear that how the idea of randomisation was presented could affect their willingness to contemplate their child's involvement:

> *I don't know if kind of like, I know it's, to me kind of like roll of a dice sounds a bit like a board game . . . you're thinking about kind of like your child's kind of like welfare and to think of a dice, you're thinking I don't know whether or not I like the idea of that, whereas if you've got kind of like a computer generated list . . . then that's fine.*
>
> *P5 (m)*

This is consistent with previous research, which has noted that explaining randomisation in terms of pulling names out of a hat or coin tossing may influence willingness to be part of a trial, leaving individuals feeling as if the approach is haphazard.[29]

Conditions for agreeing to participation

This section moves on from the description of beliefs, experiences and knowledge to consider how individuals could be clustered based on factors influencing their willingness to allow their child to be part of a trial comparing VTs and HAs. By reflecting on participants' responses, we grouped them according to key factors shaping their decision-making:

- 'protecting': not wanting to put their child at undue risk of harm (physical or psychosocial)
- 'fixing': believing that one treatment was more appropriate and/or convenient
- 'following': being persuaded by the views of professionals
- 'helping': wanting to advance knowledge and assist patients in the future.

The response of those characterised as 'protecting' was influenced mainly by previous experience, either direct or vicarious. These individuals were concerned about perceived physical or social risks associated with either VTs or HAs. Some had witnessed their child having several sets of VTs and some believed that these had caused permanent damage inside the ear. Others were reluctant to agree to the possibility of HAs, seeing these devices as an additional burden on top of scars from surgery and speech difficulties:

> *Just because urm I think I had so many other things, so many other problems as well then to sort of . . . be picked out for grommets or hearing aids and think it's going back to the hearing aids issue for us, I think we would say no.*
>
> *P36 (m)*

Whereas parents classed as 'protecting' rejected either HAs or VTs, those defined as 'fixing' articulated a preference for one of these treatments and a sense of knowing what was best for their son or daughter. Some valued the opportunity to capitalise on their child undergoing an anaesthetic for a palatal closure to have VTs inserted at the same time. Others felt that HAs were preferable because they could avoid the need for repeated VT insertions.

Data from parents whose response was shaped predominantly by a wish to protect or fix implied that accepting uncertainty about which option is most effective (clinical equipoise) was a necessary but not sufficient condition. Agreement to trial participation could also require what we refer to as 'parental equipoise'. This relates to the wider impact a treatment may have on everyday life. For example, those defined as 'fixing' felt that one approach to managing OME was more suitable in terms of its bearing on their child's psychosocial well-being. This could be expressed as a preference for the expedient solution of inserting VTs while the child was anaesthetised for another procedure. Alternatively, HAs could be preferred as an acceptable solution to hearing loss that avoided the need for surgery. Those classed as 'protecting' were not in parental equipoise because they had significant beliefs about potential adverse consequences of either VTs or HAs.

If parents expressed concerns or preferences for VTs or HAs, they were unable to agree to participation. When these were not overriding factors influencing decision-making, some individuals agreed to their child participating in a trial because they trusted practitioners. We have described this as 'following'. Alternatively, those we designated as 'helping' talked about being motivated mainly by a wish to progress knowledge and assist others in a similar situation. These interviewees did not voice strong views about VTs or HAs and, in that sense, appeared to accept there was sufficient clinical and parental equipoise to allow their child to take part in a trial. In their narratives they reflected on the widespread benefits of participation for future generations of patients.

Figure 2 summarises conditions associated with agreeing to be in the proposed trial, which were derived from interview data. It highlights that those saying 'no' or 'unsure' expressed views associated with 'protecting' or 'fixing' which were not articulated by those saying 'yes', who were willing to follow a practitioner's suggestion or to help advance knowledge. It also underlines differing moral drivers shaping people's decisions. Those characterised as 'protecting' exhibited the socially expected role of safeguarding their child. Likewise, interviewees described as 'fixing' demonstrated parental authority by suggesting a need to improve their child's circumstances in what they felt was the most efficient way possible. 'Following' could result from a sense of loyalty to staff involved in patient care, if they are the people asking parents to take part. 'Helping' suggested a wish to assist others in a similar position in the future.

These different conditions associated with decision-making ('protecting', 'fixing', 'following', 'helping') highlight possible barriers to and enablers of recruitment. They suggest that allaying concerns around a perceived need to protect children from risks, and/or addressing beliefs that one treatment has particular benefits making it more suitable for the child, might be essential prior to recruitment. If parents can be reassured about the safety of interventions proposed, and that there is no definite advantage to either, they may be more likely to move on to 'following' or 'helping' and, therefore, saying 'yes' to

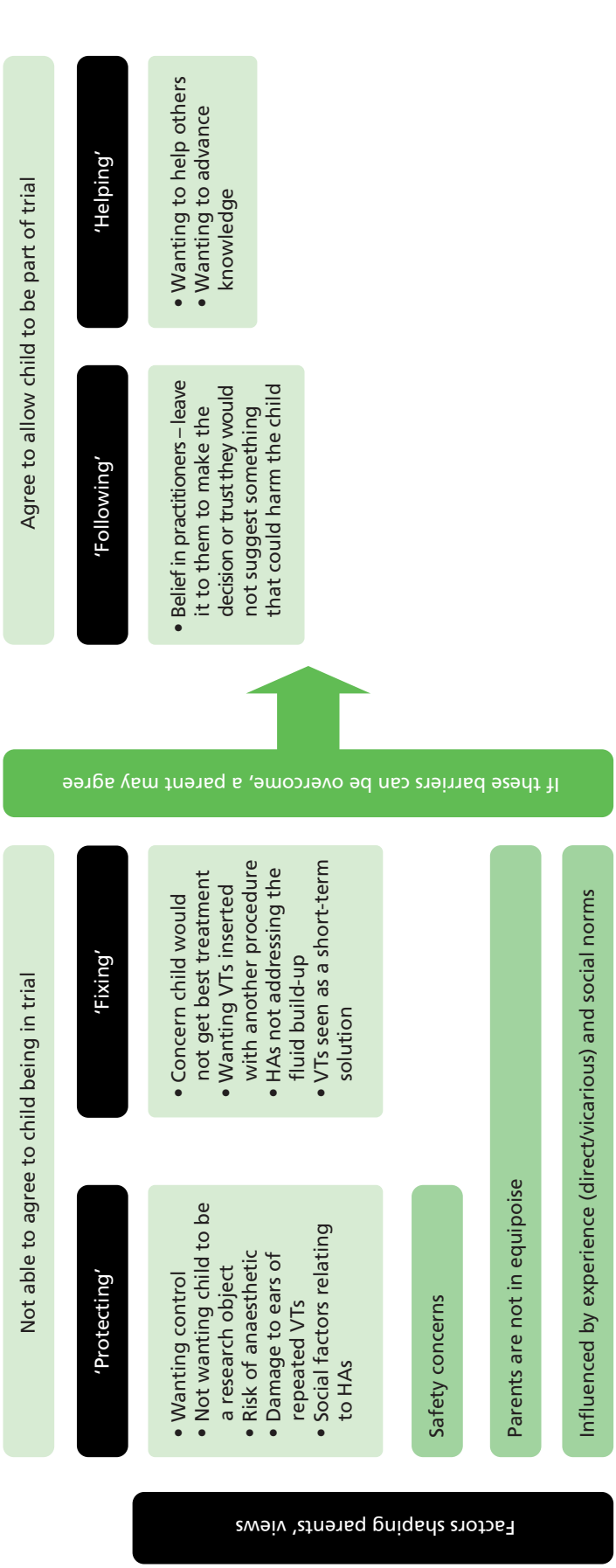


FIGURE 2 Conditions associated with agreeing to be part of a trial.

randomisation. Further research could test how these drivers of decision-making relate to recruitment, which could allow for individualised approaches to trial presentation based on whether a parent's view, at recruitment, was most reflective of 'protecting', 'fixing', 'following' or 'helping'.

Nature of the two interventions being compared

We have described how most interviewees were resistant to the idea of entering their child into the proposed trial because they did not regard it as a fair comparison. As noted by P20:

> *I don't think I'd be too happy . . . because they're two extremes, you've got the grommets, which is an operation, or the hearing aid, which isn't an operation and to me it's, I'd be, you'd be scared to be put in the grommets group . . .*
>
> *P20 (m)*

To test the influence this perceived lack of equivalence had on parents' response to our question about trial participation, after 10 interviews we explored participants' views of their child being involved in a study of a hypothetical medication for OME. As shown in *Table 12*, they appeared much more receptive to this treatment route. Those whose initial response was characterised by a drive to protect agreed if convinced that the medication was safe, as did parents classed as 'fixing', if they saw it as an attractive solution to the child's hearing problem. In general, participants described medication as a treatment that was easier to accept than surgery or HAs. They talked about taking medication being a 'normal' activity compared with the possibility of social stigma associated with HAs or the invasive nature of VTs. In addition, this option was seen as being more in parents' control, with any side effects rectified by stopping the medication:

> *. . . you have to try new things because we're never gonna learn are we and we're never gonna progress, we've got to try new things and it's a medicine. If he was ill from it, it would either come out of his bottom or through his mouth and I'd stop giving it him.*
>
> *P37 (m)*

The main hurdle that parents saw with this type of treatment was how the medication tasted, which might put the child off taking it.

Only one person agreeing to a child's participation in a trial comparing VTs and HAs would not allow her son to be part of a study examining medication for OME. Her rationale related to a concern about potential side effects, and in that sense 'protecting' came to dominate her decision-making:

> *. . . the grommets will go in his ear but they come out after so long and . . . they're not gonna damage him, you know, there's nothing major that that's gonna do to his body but obviously him taking a medication, I don't know whether he could be allergic to something that's in the medication . . . I wouldn't really want to put him through that if there was no real, 'cause obviously they couldn't promise me that something wasn't gonna happen to [child].*
>
> *P12 (m)*

Two parents who said 'no' to the trial of VTs and HAs also said 'no' to a study of a hypothetical medication for OME. Their reasons related to a concern about side effects and not wanting to try a new treatment route for this condition.

Discussion: strand 1

Interviewees diverged in their opinions of VTs and HAs, with some favouring the former while others preferred the latter. They often held strong opinions about treatment, but also reflected the uncertainty that surrounds best practice in managing OME in those with CP. Our data show that recruitment could be difficult in research comparing VTs and HAs, a common obstacle with trials[30] and a key reason for their failure.[31] Only one-quarter of those interviewed said that they would enter their child into such a study,

TABLE 12 Parents' views on their child taking part in a study comparing VTs and HAs, and on their involvement in a study of a hypothetical medication for OME

Willingness to consent to trial of surgical or medical intervention	Parent identifier[a]																					
	11	12	13	14	15	16	17	18	19	20	21	22	23	24	26	27	28	31	32	33	34	37
Yes to trial comparing VTs and HAs		✓						✓	✓			✓			✓	✓	✓	✓				
Yes to a study of medication for OME			✓	✓	✓	✓	✓	✓	✓	✓		✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

a Not all parents were asked; we only included this question about medication from P11 onwards and in five cases interviewees were not asked because children needed feeding or were getting restless.

although parents' explanations for their responses are potentially more useful than this figure because they identify concerns that, if addressed, might facilitate a higher rate of recruitment.

Other authors have talked about 'individual' or 'patient' as opposed to 'collective' equipoise.[32] We used the term 'parental equipoise' to highlight that interviewees thought more broadly than a treatment's clinical effectiveness, considering its impact on a child's and the family's psychosocial well-being. Most participants were not in parental equipoise, a condition that is necessary, we suggest, before agreeing to enter a child into a trial comparing VTs and HAs. Individuals were distributed across a continuum that included phases of decision-making shaped primarily by a need to protect, fix, follow or help. Data suggested that a lack of parental equipoise would need to be addressed if it were not to be a significant hurdle to trial recruitment.

Parents whose responses we have described as 'protecting' were not in equipoise because of the risks they associated with treatments. Some were particularly concerned to avoid their child having general anaesthetics. Negative views about VTs could also be based on personal or vicarious experience of multiple insertions, ear infections and/or concerns about long-term damage inside the ear. Therefore, HAs were preferred in some cases because they were non-invasive. Alternatively, parents might worry about their child's or others' responses to HAs, owing to social norms with regard to visible difference and disability. The response of interviewees indicates how a range of parental concerns may need to be addressed in order to recruit to a trial, including social experiences as well as clinical benefits and risks.

'Protecting' could be seen as focusing on perceived negative implications of a treatment, whereas 'fixing' suggested that individuals were making a positive choice of one way of managing OME over another. Parents described as 'fixing' believed that a particular treatment would be of greater benefit to a child and/or was more convenient than the alternative. Some considered the risks associated with VTs to be minimal, particularly if the insertion took place while a child was anaesthetised for another procedure. In contrast, HAs may be seen as requiring a longer commitment from parents, ensuring that children wore them, that batteries were replaced and that the HAs were not lost or broken. However, if VTs had not been an effective treatment, for example because they fell out soon after insertion, or if parents had seen the child flourish with HAs, they may believe this to be the most appropriate intervention for their son or daughter.

Parents who did not have strong beliefs about the benefits or risks of either VTs or HAs could be prepared to enter their children into a trial. In the case of those whom we characterised as 'following', experience again played an important role in their decisions; if they had a positive rapport with practitioners, whom they had trusted to guide them through previous treatments for CP, they may be happy to accept the request to be involved in a trial. This is not to say that those described as 'protecting' or 'fixing' mistrusted clinicians, but what was clear from their narratives were strong beliefs about treatments based on experience (whether personal or vicarious) and possibly limited knowledge to make an informed decision. If such beliefs could be addressed, there may be potential for these individuals to be more willing to agree to participation.

Altruism was an important motivation for interviewees agreeing to the potential trial, as reported in other studies.[33,34] However, the fact that parents decide about participation for a child leads to a bioethical debate about whether or not one can act altruistically on behalf of another.[19] Parents may be concerned about making the 'right decision', seeing the need to protect their child from harm as fundamental.[35] As a consequence, although some studies have found that parents are motivated to consent to a trial for altruistic reasons,[36,37] it can be conditional on reassurances about concerns for a child's safety and well-being.[35] This reflects findings that 'protecting' or 'fixing' were potential barriers to trial involvement.

Randomisation was a concept that many interviewees struggled to comprehend. The term itself may imply a haphazard approach to treatment allocation,[29] with some people believing it is an unethical way of deciding how a condition will be managed.[38] Parents we interviewed talked about their dislike of the term 'rolling a dice', feeling that it devalued treatment choices and decision-making. Misunderstanding of

randomisation shows that clear information is required when asking mothers and fathers to involve their child in a trial. Shilling *et al*.[19] reported that parents they interviewed liked being able to take away information on which to reflect and to share with family members. Likewise, in our study, parents wanted verbal details about the study to be accompanied by written material which they could discuss with others. However, previous research underlines that not everyone will read this information.[39] In addition, parents vary in terms of how much information they want about a trial.[40,41] A wish for more information has been associated with increased anxiety and less sense of control,[42] but if parents feel inadequately informed it can dissuade them from participating.[43] Anxiety may be moderated by trust in medical research and the relationship parents have with practitioners.[35] Hence, confidence in a clinical team may be a factor influencing agreement to trial involvement.[19,44] A higher proportion of our interviewees may have said 'yes' to their child's participation in a potential study comparing VTs and HAs if asked by a member of the cleft team, rather than a researcher they had only met for this qualitative study. Conversely, they may have felt more able to communicate their reservations to the researcher, without any fear of upsetting or letting down someone involved in the long-term care of their son or daughter.

Conclusion to strand 1

The diversity of views expressed in interviews indicates that parents did not share a consistent preference for either VTs or HAs. Data suggested that because many parents were not in equipoise when talking about VTs and HAs, recruitment to a trial comparing the two could be problematic. In order to perceive these two management approaches as equivalent, parents require information about how they work and potential drawbacks. Parental equipoise is a term we used to reflect the wider range of considerations, over and above clinical effectiveness, which might influence views on entering a child into a trial. The continuum of parental equipoise presented above ('protecting', 'fixing', 'following', 'helping') highlights potential factors driving decision-making around participation, and implies that specific issues may need to be considered when planning a trial comparing VTs and HAs:

- stressing the safety procedures within the trial set up
    - especially if parents are characterised as 'protecting'
- emphasising and clearly explaining equipoise (recognising that parental equipoise is wider than clinical equipoise)
    - especially if parents are characterised as 'protecting' or 'fixing'
- exploring people's previous experience/understanding of the two treatments
    - especially if parents are characterised as 'protecting' or 'fixing'
- being careful about who introduces the study to mothers and fathers
    - especially if parents are characterised as 'following'
- highlighting how results will advance knowledge for future generations
    - especially if parents are characterised as 'helping'.

As well as equipoise, parents' understanding of randomisation, and how this process of allocation is described (e.g. throwing a dice, computer generated), could influence recruitment. Parents may believe that practitioners will select the treatment that is best for their child, which could make the concept of uncertainty and random allocation difficult to accept. Prior to recruitment, time may need to be set aside to talk about how randomisation is performed and reasons for its use in this type of research.

Analysis strand 2: important outcomes for parents and children

This section presents parents' and children's views of important outcomes in the management of OME. As mentioned above, there was a specific section during interviews when they were invited to focus on this topic. However, overall narratives were also explored and interpreted for data relating to outcomes that researchers could assess in clinical trials.

Defining outcomes

The Oxford English Dictionary[45] suggests the noun 'outcome' refers to the way a thing turns out; a consequence. It is a term familiar to researchers when defining the end point measured in a study to examine whether or not a treatment is effective but is less common in everyday conversation in relation to health. This makes it a potentially difficult topic to explore with service users. Nevertheless, research has shown that collecting people's perspectives on outcomes can be a fruitful endeavour. For example, in the field of arthritis care, feeling less fatigue was found to be more important to those with the condition than traditional areas measured, such as joint tenderness and stiffness.[46]

Parents' views when talking specifically about outcomes following treatment for otitis media with effusion

Towards the middle of an interview, parents were asked what results or improvements they would look for following treatment of OME. If they had difficulty responding, the researcher posed the question 'What do you think grommets or hearing aids should do for a child with glue ear?' In our research, two parents found it difficult to distinguish processes from outcomes, listing 'good aftercare' as an important result of treatment (see *Table 13*). However, eventually most individuals were able to say something about important outcomes (only P35 could not, stating it was too difficult because his daughter had not experienced VTs or HAs). *Figure 3* shows the responses on electronic 'sticky notes' to this part of the interview for P15 and P16. The left-hand picture from the interview with P15 shows that this mother and father could not identify a key outcome, rating hearing and speech as being equally important (which was also the case for P26: see *Table 13*).

FIGURE 3 An example of sticky notes from P15 and P16 (left-hand sticky notes relate to important outcomes; those on the right relate to a question about drawbacks of treatment received by their child).

Table 13 presents outcomes referred to by parents when they were asked to focus on this topic within their interview. Green squares indicate the outcome(s) they defined as most important. This tended to be hearing but not for everyone; some people rated communication or seeing their child less frustrated as key, although both were dependent on improved hearing. Parents whose son or daughter had not received treatment (the watchful waiting group) referred to a narrower range of outcomes compared with other interviewees (see *Table 13*). Hearing, in particular, was listed as key by this group, with only P12 rating something else as more important; she explained that her son had experienced repeated ear infections, so a primary result of treatment should be to address this problem. Similarly, P9 proposed this to be her key outcome, after her son had endured recurring ear infections, which she attributed to repeated VTs. This mother formed part of a group whose child had received VTs and HAs. Reducing pain was listed frequently as an outcome by these interviewees (see *Table 13*), because a son or daughter had often had HAs following a bad experience with VTs falling out on several occasions and believing they caused damage to the ear.

Those with a child who had received only HAs were similar to the no treatment group in their focus on hearing and communication as outcomes. In contrast, the VTs-only group listed a variety of potential outcomes (see *Table 13*). Members of this group talked about HAs not addressing what they believed to be the underlying problem of fluid build-up. They were the only individuals to refer to 'OME not returning' as an outcome, which suggests they viewed VTs as a cure for the condition. This could cause disappointment when OME recurred if VTs fell out soon after insertion:

> *Yeah it was a bit of a let down really that they'd, they'd said that it would be better if she had them [VTs] and then it came out more or less straight away . . .*
>
> *P6 (m)*

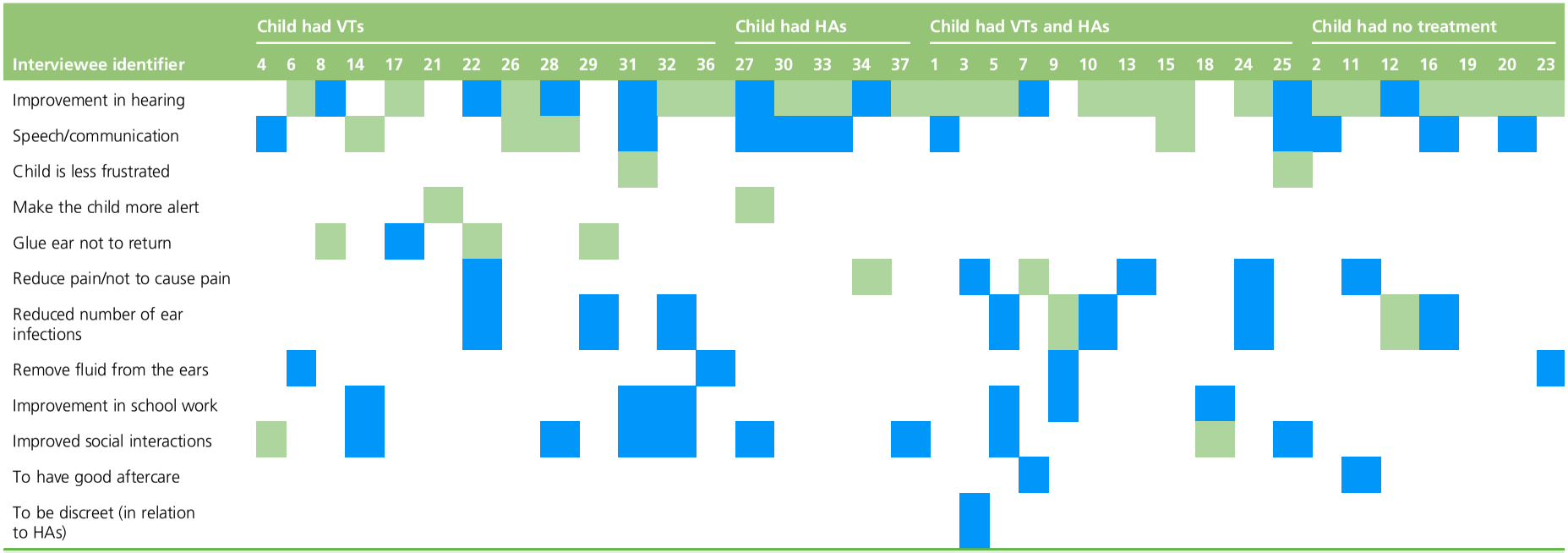
Education was mentioned by just a handful of parents in response to specific questions asking about outcomes (see *Table 13*). They listed 'improved concentration' and 'being able to follow a teacher's instructions' on their virtual 'sticky notes'. However, at other moments within the interview participants referred to their joy and relief at seeing a son or daughter advance at school once hearing loss had been addressed. Participants did not mention absence from school as an important outcome, yet during interviews they did raise the issue of time lost from school due to ear infections and for HA appointments:

> *. . . I've just had a right ticking off by the school because of his hearing. He has a green are good, amber is mediocre and red is warning and he's just got a warning because of his attendance and that is just because of the time spent in hospital and that's all because of his ENT appointments.*
>
> *P5 (m)*

Improved social interaction was a wish expressed by several parents for their child following treatment for OME. They wanted their son or daughter to be understood by others and for the child to understand others so they could communicate in social settings, with peers and at school. When unable to engage in this manner, children could remove themselves from socialising:

> *In the playground they would be playing and chasing each other and she wouldn't be able to hear and she would have to try and look at faces so she would withdraw herself, she withdrawed herself for years.*
>
> *P24 (m)*

TABLE 13 Outcomes mentioned by parents



Green shading = parent rated as most important; blue shading = mentioned but not the most important.

Age of children and outcomes

Given the wide age range represented in the study (0–11 years), potential differences in outcomes mentioned by parents for younger and older children were explored. Looking across ages, responses broadly followed the pattern outlined in *Table 14*, which suggests some differences based on age.

It is understandable that parents of preschool children tended to mention speech and language because at this age these skills are developing. There was some concern expressed about children being competent in speech and language prior to starting school so they could flourish in this setting. At school, children become less dependent on parents and begin developing their own social network. Hence, an ability to interact is important during this life stage. Parents of older children tended to worry about educational performance, perhaps because difficulties in this area were more evident at this age and concerns about coping at secondary school arose. Several of these parents had experienced OME in *their* child for several years and thought that hearing would have been 'normal' by this age (≥ 8 years). Hence, there was some anxiety among parents of older children that their son or daughter would have on-going difficulties with hearing.

Parental stress

Interview transcripts were replete with indications of parental anxiety and strain. For example, seeing a child in pain, usually due to ear infections or damage, was upsetting for parents, who felt helpless to relieve this discomfort. Parental stress could also stem from decision-making around whether to have VTs or HAs and whether or not to try VTs more than once. Some parents talked about the need to maintain HAs (e.g. cleaning them, ordering batteries), although this was depicted as a hassle rather than a major problem. What appeared to be more stressful was when a child lost a HA; parents then had a child who was unable to hear and they had the embarrassment of contacting an audiology service to request a new HA. Frustration exhibited by children when unable to hear caused parental disquiet in case others thought their son or daughter was 'just being naughty' (P22 m). It could also lead to disharmony at home, as children were moody with their siblings and parents disagreed about how to manage such behaviour:

> *. . . it [child's behaviour] was causing a lot of stress in our house . . . parents could be falling out with each other, saying 'well why are you not being firmer' . . .*
>
> *P18 (m)*

> *They [school] even made an appointment for us to see somebody . . . about her behaviour. So that made me quite anxious and worried and thought that we were, you know, she was really naughty and going off the rails but when we sort of realised that it was more cause of . . . her ears then I was angry at the school more after that because I'd, not taken it out on her but . . . I'd been a bit upset and taking it out on myself more than anything, thinking it was something that we were doing or that I was doing personally.*
>
> *P22 (m)*

TABLE 14 Outcomes described by parents based on their child's age

Parents of preschool children (age 0–4 years)	Parents of young primary school children (age 5–7 years)	Parents of older primary school children (age 8–11 years)
Focused on hearing, pain and ear infections	Focused on hearing, pain and ear infections	Focused on hearing, pain and ear infections
Focused also on speech and language	Focused on social interaction more than just speech and language	Focused on social interaction and educational performance
		Talked about a wish for glue ear to be removed

Physical and psychosocial outcomes

Parental interviews contained data on a mixture of physical and psychosocial outcomes (*Table 15*). Interview data suggest that when considering outcomes, parents saw the health problem (OME) and associated symptoms more broadly within the context of the individual child's life. For example, the physical problem of not hearing appeared to contribute to difficulties in the child's psychological functioning (e.g. frustration, fear, sadness) and social life (e.g. learning and communication, being unable to take part in activities like swimming). Physical outcomes were related, primarily, to VTs (e.g. infections

TABLE 15 Psychosocial and physical outcomes as depicted in interview transcripts of parents

Broad area	Specific outcomes	Reality of parents' experiences	Quotations from interviews
Social	Education	Once hearing had been addressed parents felt children blossomed educationally and were more able to concentrate	*. . . to be at school and not feel that you weren't hearing everything and being able to . . . take part fully in school . . .* *P18 (m)*
	Interaction	Speech and language improved after treatment for OME – parents felt once hearing was rectified, children were more able to be part of their social world and were aware of what was happening around them	*. . . she was very closed off from people at school . . . She didn't really talk to anybody and I didn't understand how difficult that was until we found out how much of a hearing loss she had. But since having the hearing aids she's, she'd made a few friends.* *P13 (m)*
Psychological	Child feeling less frustrated	Children became less frustrated when able to hear what others were saying	*Socially it [treatment] helps them and it helps them with any frustrations because it's annoying when you can't hear things properly all the time.* *P31 (m)*
	Child being more confident	Children seemed to become more self-confident in social and school settings – parents saw their child's personality come out when they could hear better	*. . . everyone was like 'wow he's come out of his shell hasn't he'.' Wow, where's his quiet side gone'. I said 'it's them bloody hearing aids and it were'.* *P37 (m)*
Physical	Rectify hearing loss (once and for all if possible)	VTs helped some children's hearing, if they did not fall out, as did HAs for others – some parents suggested treatment should mean the hearing problem would not return when discussing VTs	*The first thing that I wanted was for him to hear. Better hearing.* *P5 (m)*
	Reduce ear infections and blocked ears	Some parents felt VTs reduced ear infection frequency, others believed they caused this problem; with HAs there could be a difficulty of not being able to wear them with an ear infection and it was felt that HAs did not address the underlying fluid build-up	*I'm not expecting it [VTs] to stop him having ear infections completely but to kind of control it so you know it wasn't as often as he's getting them cause I mean I don't know how often kids get ear infections but to me it seemed like a lot to be having.* *P12 (m)*
	Pain-free	Parents suggested that HAs should be comfortable and that VTs should not cause pain, which some parents attributed to their insertion (e.g. earache)	*She wouldn't be ill for a start. She wouldn't have leaky ears and pain. It's just [with VTs], this is all it's been is pain, blood, sick. We've not had a result, there's never been a result.* *P24 (m)*

and perforations). Some interviewees felt that a resolution of hearing loss should not be sought if it was going to cause further significant discomfort for the child:

> *. . . she had two major ear infections with them [VTs] and they are now blocked so she has to go back and get them looked at. So she's had nothing but trouble with them . . . The infections and the pain and I don't think you know her being in pain outweighs her hearing, if that makes sense.*
>
> *P13 (m)*

Parents described the change in their child once hearing loss had been addressed in very emotive terms, emphasising, once again, that OME could affect numerous areas of a child's social functioning and psychological well-being:

> *He just was like in this bubble and it was just, I just, to me I can just picture the ears just opening up [when VTs were inserted] and everything getting in . . . So really it was just participating in more things perhaps, being involved a little bit more in everything that was going on around him, being able to give him the chance to do it all . . . He was even more enthusiastic and just a completely different boy to me really [laughs].*
>
> *P4 (m)*

Their narratives often centred on 'normality'. They wanted their son or daughter to have the same opportunities in life as their peers and to reach their potential at school, noting that OME could prevent this from occurring. It was implied that successful treatment would help the child become confident and content by allowing them to hear what was taking place around them, so they did not feel like an outsider. When OME was present, parents suggested it could mean their son or daughter was disconnected from the social world, resulting in the child becoming agitated:

> *Oh, he was not a very nice toddler [laughs]. He was quite difficult as a toddler, like we'd go to like places, he was alright at pre-school, he was better for other people but when it was like, especially when he was really little . . . when other kids are all starting to speak and he couldn't communicate properly with other children. So he'd get, he'd like lash out, you know, he'd try and talk to them but obviously he wasn't making the right sounds.*
>
> *P33 (m)*

A desire for the child to have the same life chances as their peers and not to miss out educationally relates to a concept underpinning parental interviews around achieving developmentally appropriate independence. For example, parents worried if their child was reliant on siblings for friends and social activities, and encouraged them to pursue their own interests. Interviewees appeared to value a progression towards autonomy and talked about seeing the child's personality emerging as a positive. *Box 2* provides some exemplar quotations from parents on this topic.

BOX 2 Illustrative extracts when interviewees talked about their child and age-appropriate independence

> *. . . without his hearing aids he would be very limited. There's no way I would let him, I mean at the minute if he wants to play out and he'll play out just in front of the house. Without hearing aids it wouldn't happen because he wouldn't hear the cars coming down . . .*
>
> *P3 (m)*
>
> *. . . she was more clingy than any of the other children would have been. In lots of situations she didn't like being left . . . when she first started [school] she was very quiet, she wasn't even answering the register . . . so there was a tiny bit of concern at one stage . . .*
>
> *P18 (m)*
>
> *The teacher would have to come and say 'you can do it, you know how to do it', and go through it with her, 'no, no I can't do it', rather than trying herself, she'd rather the teacher come and be with her and try and help her . . . But at the minute they say she's trying so much better and she'll give it a go rather than saying straight away 'I can't do it'. She'll give it a go and quite often she'll get it right, she knows what she's doing.*
>
> *P22 (m)*

Children's views of outcomes

The term 'outcomes' could have been difficult for children to understand so they were invited to say what they thought was 'good' and 'not so good' about VTs or HAs. Responses to this question are listed in *Table 16*, which shows that children varied in their views, with some struggling to provide an answer because they had received treatment (usually VTs) several years previously or were in the watchful waiting group.

Hearing was referred to by children as a primary result of treatment for OME because it allowed them to understand others and to join in activities. However, one 10-year-old girl said an improvement in hearing had to be balanced against problems associated with interventions such as HAs:

> C10: *They [HAs] make you hear properly. But I'd rather no hearing. I'd rather, I'd rather not hear properly than wear those things. I hate them so much.*
>
> ST: *What makes you hate them so much?*
>
> C10: *Everything about them. Sometimes they can get way too loud and I can't really turn them down . . . and then sometimes they're really, really low when you turn them on and then they stay like that . . . I'd be sitting there [in the classroom] with the hearing in my ear blocking all the sounds from my right ear cause, I'd be like, 'what, what, what did you say sir?'*

For participants who wore HAs, being involved in their design was a positive factor associated with these devices. Conversely, when discussing negative aspects of HAs, children mentioned their visibility; that they could fall out when playing; having to change batteries; an inability to alter the volume setting; and they could be uncomfortable in their ear. The main issues they associated with VTs included having to undergo an operation; a belief that they could cause infections; and feeling their presence inside the ear. In *Box 3*, ideas relating to children's expectations of treatment for OME have been summarised into four main areas, alongside illustrative quotations. More views were expressed (positive and negative) about HAs compared with VTs, possibly because children had experienced them recently or currently wore these devices, whereas VTs may have been inserted several years prior to interview.

TABLE 16 Children's views of treatment for OME

Identifier	Treatment	What's good about VTs or HAs?	What's not so good about VTs and HAs?
1	VTs and HAs	Nothing	People touch it (HAs)
			It's irritating (HA)
			Sometimes my ear hurts (HAs aggravates)
			Tickles when getting moulds made (HAs)
3	VTs and HAs	Helps me listen (HAs)	Small children (noisy in restaurants) (HAs)
		Miss school (for appointments) (HAs)	Annoying when batteries go (HAs)
		Likes playing with putty when getting HAs made	
5	VTs and HAs	Nothing	Felt tight inside the ear (VTs)
7	VTs and HAs	Excellent because they are pretty (HAs)	Annoying because they fall out (HAs)
9	VTs and HAs	Hearing is louder when in (HAs)	You have to be put to sleep and don't know what they [doctors] are doing (VTs)
			They [doctors] force you to have it (VTs)
10	VTs and HAs	They make you hear properly (HAs)	I hate wearing hearing aids
			They are annoying cause they itch (HAs)
			Can't change the levels on HAs
14	VTs	Hearing improves (VTs)	Irritating (could feel the VTs were there)
		Confidence (when had VTs knew there was nothing to worry about the operation)	
16	Watchful waiting	Not asked because had not had treatment	
17	VTs	Not asked because child did not have much to say on the topic	
18	VTs and HAs	Hearing is better (HAs)	Not always comfortable, especially when get a new mould (HAs)
			Have to take them in and out (HAs)
			Batteries – make a noise when running out (HAs)
22	VTs	It helps you to hear (VTs)	Painful ear just after surgery (VTs)
24	VTs and HAs	They are useful and they help you to understand people (HAs)	Putting them in was painful (needle) and they cause pain (VTs) (had recurrent ear infections)
26	VTs	Can hear better (said this about HAs but had not had them)	Annoying cause you have to wear them (HAs – even though he did not have personal experience of these devices)
27	HAs	They help you hear and do more (HAs)	You have to keep changing batteries (HAs)
			Can hurt when getting the mould made (HAs)
28	VTs	Hear better (VTs)	Nothing
31	VTs	Rest of the interview suggested child would struggle with the task	
32	VTs	Child struggled with giving opinions	
33	HAs	Rest of the interview suggested child would struggle with the task	

TABLE 16 Children's views of treatment for OME (*continued*)

Identifier	Treatment	What's good about VTs or HAs?	What's not so good about VTs and HAs?
34	HAs	Hear better (HAs)	Tickles when getting moulds made (HAs)
35	Watchful waiting	Child struggled because had no experience of treatment	
36	VTs	Child struggled because had no experience of HAs and had VTs a number of years ago	
37	HAs	Hear better (HAs)	Running out of batteries (HAs)
		You get to pick your own colours (HAs)	Tickles when getting moulds made (HAs)

Shaded cells indicate no response, with explanation.

BOX 3 Quotations from children about expectations of treatment for OME

1. Help with hearing

It's a bit hard to explain but they [HAs] help by letting me hear people more and I can urm, if they wasn't invented I'd just be completely deaf like that and I wouldn't even hear anything.

C3

I could tell [hearing had improved] because urm I could like mum didn't have to repeat things over and over again [after he had VTs] like she did sometimes because I just wasn't, I was concentrating on something else but urm before urm she had to do it quite a lot . . .

C14

2. To be aesthetically pleasing

Every time I have new hearing aids my friends say 'oh they look nice' [laughs] . . . I think they're a bit pretty.

C7

3. Not to cause discomfort, pain or irritation

Urm, when it [VTs] goes in it feels too tight . . . Inside my ear . . . Soon as I'd had the operation.

C5

I find it OK but if the, one of the wires goes too far in I find it, it urm hurts a bit . . . Well they put the mould in [for the HA], then they put this wire in and they put like a mic over it or something.

C27

4. Not to interfere with activities

. . . when the batteries go . . . I feel like tearing it [HA] out because when you're doing it, it's always there, just in the back of your mind, when you're playing a game and then it just pops into the back of your head you go hooray, I think it might be charged up again, put it back on and it goes on and then a few moments later goes off.

C3

. . . the bad thing is it [HA] does fall out sometimes at school . . . That was a surprise that was cause I didn't know it was gonna fall out.

C18

Combining children's and parents' views

Children and parents placed an importance on hearing as an outcome. Yet, as mentioned above, it was not referred to in isolation and interviewees did not focus on hearing as simply a physical improvement that could be measured. Instead, they associated it with psychosocial functioning, such as:

- independence (freedom for the child to go and do things on their own)
- performance at school (more attentive, able to read because they could hear words)
- able to interact and connect with the social world (being understood and being able to understand what others were saying)
- less frustration because of better communication, resulting in improved behaviour and self-confidence
- engagement in everyday activities (e.g. swimming, cinema)
- impact on home life (less shouting, TV less loud).

Views of outcomes from children and parents are merged in *Figures 4* and *5*. *Figure 4* depicts physical outcomes mentioned within interviews, whereas *Figure 5* centres on psychosocial ones. *Figures 4* and *5* illustrate a pathway from hearing to physical and psychosocial outcomes, respectively, based on data from interviewees. White rectangles in these figures indicate outcomes mentioned by parents only, whereas shaded ones represent those referred to by children and parents. Children's views were not inconsistent with those of parents, but mothers and fathers did have more to say about outcomes that could be used to assess the effectiveness of interventions for OME.

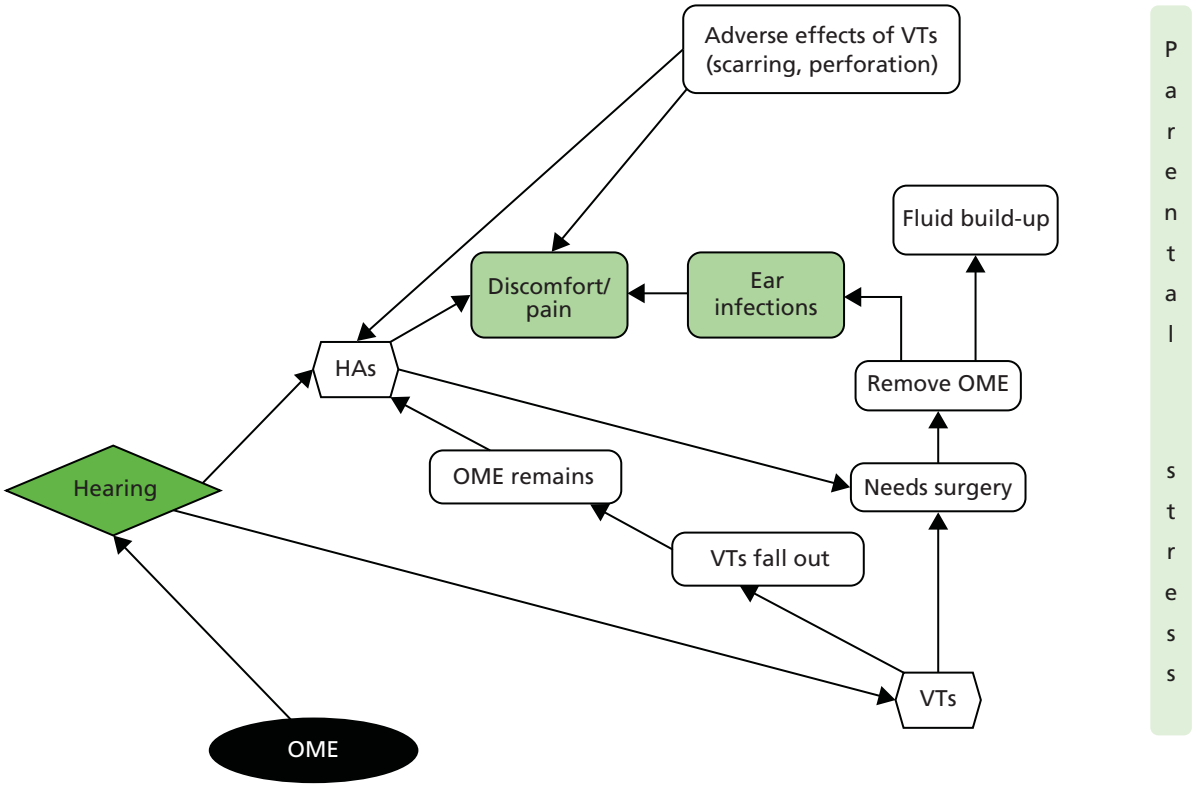


FIGURE 4 Physical outcomes described in interviews (shaded boxes = mentioned by parents and children; white boxes = mentioned by parents only).

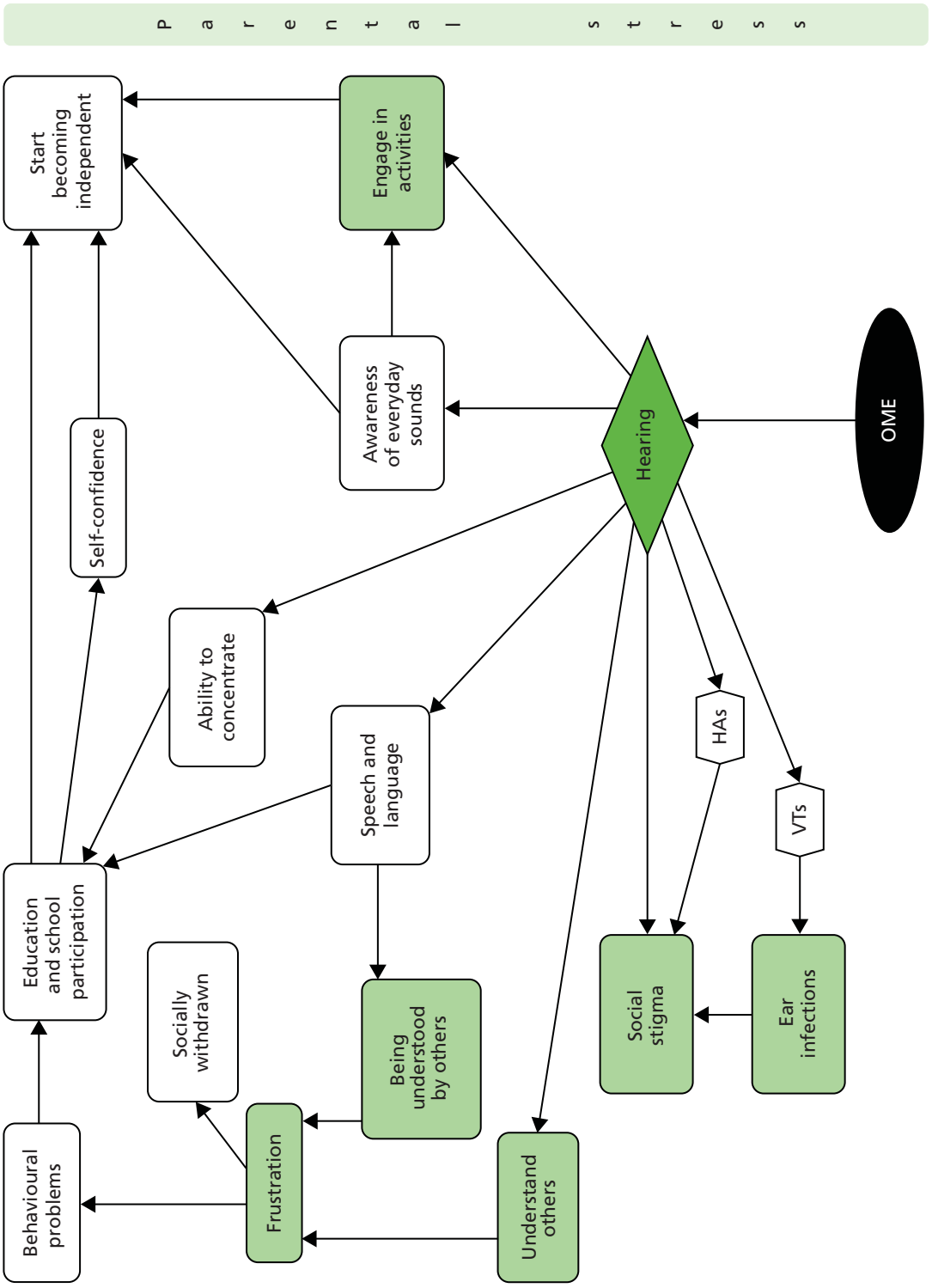


FIGURE 5 Psychosocial outcomes described in interviews (shaded boxes = mentioned by parents and children; white boxes = mentioned by parents only).

Treatment-specific outcomes

Certain aspects of managing OME were specifically connected to a treatment. These are highlighted in *Figure 6*, which shows that VTs, in particular, were associated with physical outcomes. For example, parents often referred to VTs as bringing additional benefits compared with HAs by addressing fluid build-up inside the ear. In some interviews, parents believed that VTs reduced the frequency of ear infections experienced by their child. Conversely, others blamed VTs for constant pain:

> *I think him having the grommets has caused more agony and ear infections and sleepless nights . . . He's had very many sleepless nights to the point where you give paracetamol but the child still says 'I can't sleep, it's painful' and you as a parent don't have anything to do other than be there for your child, console them . . .*
>
> *P9 (m)*

Ventilation tubes were also associated with procedural issues and, to a lesser degree, social consequences. In contrast, HAs were related mainly to social consequences (see *Figure 6*). Although parents expressed concerns that HA would result in their child being bullied, in reality this fear did not appear to be realised; children who wore HAs were said to adapt very well to these devices and did not report the social stigma that parents anticipated. However, as children got older they appeared to become more self-conscious about having a HA. One boy, aged 8 years, talked about his frustration when classmates tried to touch his HAs:

> C1: *They just talk to me all the time about them and everyone touches them.*
>
> ST: *Everybody touches them, at school . . . How do you feel about that?*
>
> C1: *I feel weird.*
>
> ST: *Why do you feel weird?*
>
> C1: *Because everyone keeps touching them.*

Discussion: strand 2

Interviews started with participants talking about experiences of OME and then midway through they were invited to focus discussion on the topic of outcomes. Therefore, most parents and children identified outcomes directly by listing them on a tablet computer and indirectly through describing how this condition and its management affected their lives; outcomes were explored within the context of this broader reflection on experiences, meaning that although reference to outcomes was sometimes explicit, it also involved drawing out from participants what were important consequences of OME based on their overall narratives about living with this condition.

The key outcome emerging in participants' narratives was hearing. This was referred to by children *and* parents, who saw it as more than a restoration of physical functioning. Instead, hearing was described as important largely because of the psychosocial consequences associated with its loss, such as relationships with others and ability to progress in life. This could be summarised as the child's 'connection with their social world'. *Figure 5*, in particular, delineates how psychosocial variables were linked to hearing. However, some parents and children implied that hearing should not be sought at any cost; comfort and absence of pain were felt to be more important in some cases than being able to hear everything that was occurring. Hence, a degree of hearing that allowed children to function in the social world without excessive discomfort was a happy medium that certain interviewees depicted as acceptable.

Data suggest that particular outcomes may become increasingly important over time, with parents of older children being more concerned about social interaction and education; whereas earlier, focus on speech and language development may be a priority. Others have noted that hearing loss can leave individuals

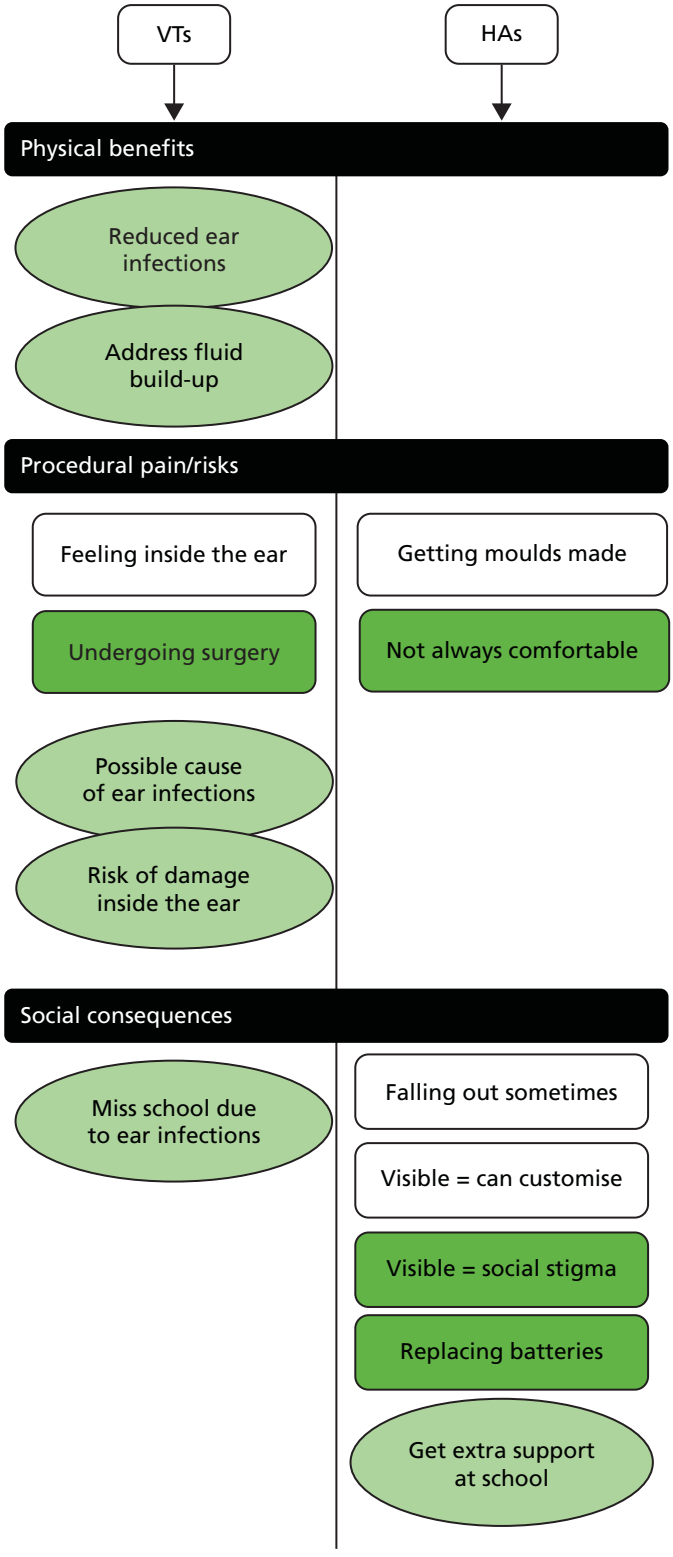


FIGURE 6 Outcomes associated with treatment described by parents and children (green rectangle = mentioned by parents and children; green oval = mentioned by parents only; white rectangle = mentioned by children only).

behind educationally.[47] Education was only mentioned by parents; children were more concerned with a wish to fit in with friends and not missing out on valued activities (e.g. swimming and playing). Parents identified consequences for children's self-esteem and confidence as important outcomes, which could be related to a notion of experiencing a 'normal' childhood and reaching their potential. This highlights that outcomes discussed during interviews did not necessarily relate to a discrete endpoint, implying that a distinction may be required between immediate and longer-term consequences of treatment for OME.

Conclusion to strand 2

Hearing was a key outcome referred to by parents and children. This was probably to be expected, given the nature of the problem and aims of treatment. Yet although hearing was central to interviewees' concerns, this was largely because of its psychosocial consequences. Hence, rather than absolute hearing at one moment in time, participants talked about the impact of not hearing on social development and psychological well-being; interviewees were looking for a health gain in terms of hearing to engender improvements in the child's functioning in their social world and to bring them psychological comfort, including self-confidence, a move towards independence and the opportunity to develop their personality. Interviewees talked about significant psychosocial difficulties that stemmed from hearing loss, including poor social interaction and educational performance. Furthermore, both physical and psychosocial outcomes contributed to parental stress, with hearing difficulties adding to anxiety and pressures already associated with caring for a child who had a cleft.

Strengths and limitations of the study

Findings were strengthened by interviewing a range of participants in terms of the child's age, gender and treatment received for OME. We involved fathers as well as mothers and recruited from more than one site. We talked with children, allowing their voice to be heard on important outcomes. Interviews addressed an event that may have happened several years previously (i.e. VTs insertion), which could result in recall bias. In addition, parents were asked to comment hypothetically on entering their child into a potential trial. We cannot be certain that their response would be the same if they were asked by a member of their team to be part of a real investigation. Parents we talked to who drew on personal experiences of treatment may not be representative of those invited to enter their children into a future trial at the first intervention for OME. However, interviewees included those whose children had only experienced watchful waiting, which indicates that even if someone has no direct experience, they may still have views about one intervention or another through listening to others.

Parents taking part had agreed to be interviewed. It could be argued that they might be predisposed to say 'yes' to trial participation. However, we spoke to people with a range of views about allowing their child to enter a trial comparing VTs and HAs. We interviewed 71% of those invited to take part, a high response rate for this type of study. It is not clear whether or not those refusing to be interviewed were put off by the more involved nature of data collection compared with other studies (e.g. completing a questionnaire), although one mother did refuse because she only wanted to be interviewed by telephone due to her busy family life. Two children declined to take part because they did not like talking to strangers but their mothers were interviewed. To encourage children to talk, we used a tablet computer to perform activities. We explored what they thought it was like for other children to undergo an operation or to wear a HA and asked them to comment on what might be good and not so good about each treatment. Children said at the end of their interview that they had enjoyed being listened to by a researcher and doing activities.

We took a systematic approach to the analysis, involving a mixed team of researchers and clinicians who cared for children with CP. This helped to add to the depth of the analysis and allowed for the checking and challenging of data interpretation. Using the computer package NVivo 9 meant a clear trail of how the analysis progressed was kept, from initial coding through to categorising and development of key ideas reported above.

Chapter 4 Development of a core outcome set for use in clinical trials of treatment for otitis media with effusion in children with cleft palate

Background

Clinical trials should have defined primary and secondary outcomes that answer questions generated by the main hypotheses. However, when we consider outcomes that may be used in studies of the treatment of OME in children with CP, it appears that these are numerous and diverse, and include outcomes such as chronic otitis media, OME, hearing loss, eustachian tube function, behaviour, receptive language and side effects of treatment to name a few. Furthermore, some of these outcomes may be influenced by other factors associated with clefting, for example the effect of the palatal cleft on speech.

Heterogeneity of outcomes across trials has been illustrated by a recent review which aimed to identify COSs for trials of treatment of childhood conditions. The authors categorised outcomes measured in clinical trials for a variety of paediatric conditions into six broad domains: disease activity, physical consequence of disease, functional status, social outcome and QoL, side effects of therapy and health resource utilisation.[48] Importantly, in this review, they did not retrieve any studies relating to OME.

In summary it appears that the domains in which the most tangible benefits from the users and providers of care for children with CP and OME are unknown and traditionally researchers have used diverse outcomes. This situation could lead to the following potential problems.

Heterogeneity between studies

This may be illustrated by considering the findings of a recently published systematic review directed at the early routine insertion of VTs for management of OME in children with CP. In this review the authors evaluated the literature up to 2006 (including RCTs, controlled clinical trials, case series and historical cohort studies).[1] Eighteen studies satisfied the inclusion criteria. When the outcomes were evaluated the studies were shown to have used varied primary outcome measures including hearing loss, tympanosclerosis, parental satisfaction with treatment, speech and language, and OME. Furthermore, there was inconsistency in the method of assessment for some outcomes; in particular speech and language which was assessed by undefined speech and language therapist assessment, a study specific scale or using the Reynell Developmental Language Scale. This limited consistency between studies, leading to marked heterogeneity, may result in difficult interpretation and comparison of findings and hinder potential meta-analysis.[48,49]

Outcome reporting bias

Another relevant factor is outcome reporting bias. This occurs when only a selection of results for measured outcomes are reported in a study, and the choice of which to report is based on the results. For example, a tendency to report only significant or positive findings results in a biased representation of the results of a trial. There is overwhelming empirical evidence that this phenomenon occurs.[50]

Core outcome sets

One strategy that has been suggested to overcome these issues is the development of a COS which should be measured and reported in all RCTs of a specific condition.[48,49,51–54] As a result, the risk of outcome reporting bias and heterogeneity is reduced for those outcomes in the set, and the potential for carrying out a meta-analysis for key outcomes is increased.

The outcome measures that could potentially be used to evaluate OME treatment are numerous and diverse, and may also be affected by specific factors in clefting. There is currently no COS available for clinical trials of the management of OME in children with CP.

Aim and objectives

Aim

The aim of this part of the project was to contribute to the development of a COS, relevant to studies of the treatment of OME in children with CP.

Objectives

Specific objectives were:

1. to systematically review the literature to identify the outcomes that had been previously reported in studies of the treatment of OME
2. to prioritise outcomes from the clinician perspective
3. to prioritise outcomes from the perspective of patients who can express their views, and parents
4. to compare outcomes of importance to health professionals and parents/patients
5. to integrate patient/parent and health professional outcomes into a combined COS.

Systematic review and development of a list of outcomes previously reported

Aim

To identify the outcomes that had been used in previous research evaluating the management of OME in children with CP.

Methods

The systematic review was carried out using two sources of publications:

1. Studies of the early placement of VTs for children. This was done by updating the search from a previous systematic review.[1]
2. Studies of other surgical interventions for OME in children with and without cleft obtained from relevant Cochrane reviews.

Population

Children, with and without CP, aged < 18 years with OME.

Intervention

Any surgical intervention used to manage OME.

Comparison
Untreated control or other surgical interventions.

Outcome
Any outcome that was reported.

Criteria for considering studies for updating the Bristol review[1] and Cochrane reviews identified through Cochrane Central Register of Controlled Trials search
We included the following types of studies:

- systematic reviews with/without meta analyses
- RCTs
- controlled clinical trials
- case series
- prospective cohorts
- retrospective cohorts.

Search methods for identification of studies
An identical search strategy to Ponduri *et al.*[1] was used and applied to the Cochrane Central Register of Controlled Trials (CENTRAL), EMBASE, MEDLINE, and Cumulative Index to Nursing and Allied Health Literature (CINAHL) (January 2006–April 2011).

The detailed search strategies applied are given in *Appendix 4*.

Multiple databases were utilised to maximise the sensitivity of a search. CENTRAL comprises only studies which are deemed to be controlled trials by a team of reviewers; EMBASE, MEDLINE and CINAHL include published research of various study designs. The advantages conferred by using CENTRAL in addition to the other databases is that trials from other sources of research (e.g. journals not indexed in MEDLINE and conference proceedings) are hand-searched, and controlled trials from these are included. This improves the chances of identifying all relevant studies.

Eligibility of studies
Two researchers (MOS and KOB) independently assessed the abstracts of studies resulting from the searches using a screening proforma. Full copies of all potentially relevant studies and those appearing to meet the inclusion criteria, or for which there were insufficient data in the title and abstract to make a clear decision, were then obtained.

The full-text papers were assessed independently by two review authors (MOS and KOB) and any disagreement on the eligibility of included studies resolved through discussion. Where resolution was not possible, a third review author (IAB) was consulted.

For the purpose of this study there was no synthesis of outcome data from the RCTs, and hence a critique of the methodological quality of the studies was not necessary.

Data extraction

Two reviewers (NH and IAB) independently extracted the data. NH and IAB then reviewed the extracted data together to assess consensus and to ensure that all outcomes have been identified. Disagreement was resolved through discussion, where resolution was not possible, a third review author (KO) was consulted. The following data was extracted from each study:

1. study type
2. author details
3. year and journal of publication
4. intervention(s) under investigation
5. whether the study population was exclusively paediatric and CP or mixed (children and adults, with or without CP)
6. age and number of children included in the study population
7. inclusion and exclusion criteria.

Outcomes:

1. the outcomes which were measured, including the method of measurement
2. the time points at which they were measured
3. if stated, the designated primary outcome
4. designated secondary outcome(s).

Data analysis and presentation

For analysis purposes the data was initially tabulated so that for each study outcomes were listed.

The outcome domains were then determined following a review of the extracted outcomes by the authors (IAB and NH). The outcomes were then grouped under these domains. Finally, the outcome domains and included outcomes were reviewed by the SAG to assess suitability of domain name and grouping of outcomes.

Systematic review: results

Figure 7 illustrates the generation of the eligible studies. The search retrieved 85 potentially eligible studies, after screening titles and abstracts, all but nine studies were deemed to be irrelevant. After further analysis of the full texts, one further study was excluded as it was undertaken to determine the frequency that children with CP pass their newborn hearing test.[55] In addition, full texts of the 18 studies identified in the previous review[1] were retrieved. Six Cochrane systematic reviews relating to OME were also retrieved to examine outcomes measured in studies of other interventions for OME in children with and without cleft.[56–61] This yielded another 24 studies.

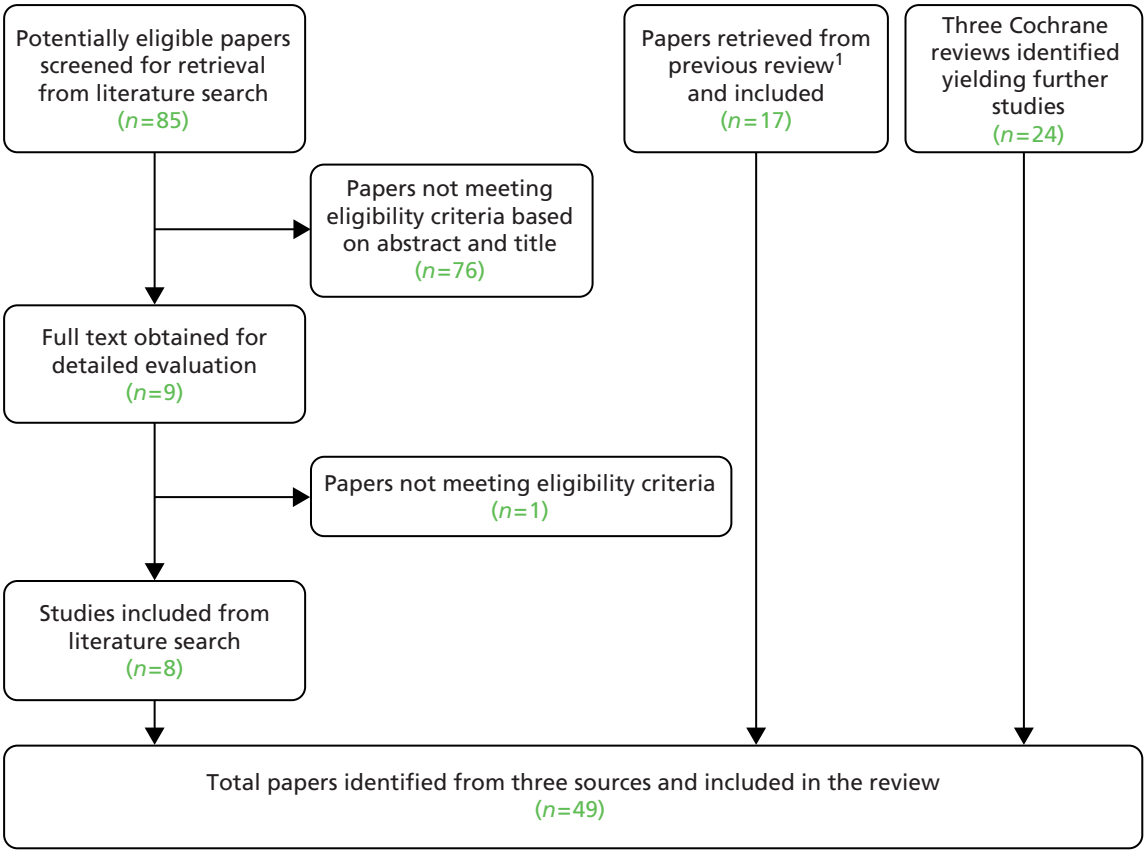


FIGURE 7 Retrieved studies flow chart.

Description of included studies

The characteristics of included studies together with outcomes measured are given in *Table 17*. Ponduri *et al.*[1] reported 18 studies of children with CP and 17 have been included in this review.[62,63,65–78] One paper of the original 18 identified[110] was excluded from the present study as the results included are reported in a later paper by the same author.

A further eight studies of children with CP have been included.[5,79–85] In addition, the search was expanded to studies of children without CP or where the presence or absence of a cleft was not specified, this yielded a further 24 studies.[86–89,91–109]

Studies did not always clearly state the primary outcome and when this was the case the primary outcome was inferred from the sample size calculation, the study title and/or the results presented first in the results section. Papers which did not explicitly identify a primary outcome are detailed in *Table 17*. The primary outcome was determined independently by NH and IAB, and reviewed with any disagreement discussed further, there was disagreement for only two papers with further discussion required to agree the primary outcome. Papers with disagreement are noted in *Table 17*.

Each outcome measured was listed by study, only individual outcomes were included (e.g. where an outcome was measured using different methods this was counted as one outcome but the methods of measurement noted).

TABLE 17 Characteristics of included papers including outcomes used

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
1	Møller (1982)[62]	Retrospective cohort	12 months–4 years	70	Children with CP aged 12–156 months at study entry	CP with bilateral VT, unilateral VT, anterior quadrant VT	Incidence of otorrhoea	Incidence of otorrhoea within 1 year of VT	Primary outcome not specified in paper
							Necessity to remove VT	Necessity to remove VT	
							Middle ear ventilation	Position of tympanic membrane (measure of middle ear ventilation)	
							Tympanosclerosis	Tympanosclerosis	
							Middle ear ventilation	Retraction	
							Necessity for new VT	Necessity for new VT	
							OME[a]	Otoscopic findings (OME)	
2	Potsic *et al.* (1979)[63]	Retrospective cohort	5 years	69	Children with CP	No treatment vs. VT when needed vs. VT early insertion	Hearing impairment[a]	Pure tone audiometry	Primary outcome not specified in paper
							Hearing impairment[a]	Speech audiometry	
							OME	Otoscopic findings	Hearing impairment included as an outcome with two methods of assessment
3	Paradise and Bluestone (1974)[64]	Case series	32 months	138	Children with CP aged < 24 months. Pittsburgh USA	n/a	Incidence of otorrhoea	Otorrhoea – 6 months post operation	Primary outcome not specified in paper
							Duration of otorrhoea		OME included as an outcome with two methods of assessment
							OME[a]	Pneumatic otoscopy	
							OME[a]	Microscopy	
							Perforation	Perforation	
							Tympanic membrane on otoscopy	Fullness or bulging of the tympanic membrane	
							Tympanic membrane on otoscopy	Erythema or colour of the tympanic membrane	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
4	Møller (1981)[65]	Prospective cohort	36 months	261	Children with CP aged 1 month–20 years (mean 7 years). Western and Northern Norway	28 bilateral VT, 40 unilateral VT	Hearing impairment[a]	Pure tone audiometry	Primary outcome not specified in paper
							Middle ear pressure	tympanometry	Disagreement between reviewers on primary outcome requiring further discussion. Consensus reached that hearing impairment was considered the primary outcome
							OME	Otoscopic findings	
							Scarring of the tympanic membrane	Otoscopic findings	
							Atelectasis of the tympanic membrane	Otoscopic findings	
							Tympanosclerosis of the tympanic membrane	Otoscopic findings	
							AOM	Otoscopic findings	
							Chronic perforation	Otoscopic findings	
							Stapedial reflex	Tympanometry	
5	Smith *et al.* (1994)[66]	Case series	65 months	81	Children with CP, age not stated. North Carolina, USA	–	Eustachian tube dysfunction[a]	Tympanometry	Primary outcome not specified in paper
							Number of VTs until normal tympanometry	Tympanometry/note review	
							Average time to extrusion of VT	Note review	
							Hearing	Pure tone audiometry	
							Perforation of the tympanic membrane	Otoscopy	
6	Hormann and Roehrs (1991)[67]	Prospective cohort	Ages 8 and 16 years	184	Children with CP. Hamburg, Germany ($n = 126$) and Ioqa, USA ($n = 58$)	Grommets vs. early grommets	Risk of otorrhoea	Risk of otorrhoea	Article in German. Abstract and Ponduri *et al.* 2009[1] review used
							Middle ear status	Unknown	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
7	Broen *et al.* (1996)[68]	Prospective cohort	3-monthly follow-up for 9–30 months	28 CP; 29 non-CP	Children with and without CP. Minnesota, USA	Cleft vs. non cleft	Hearing impairment[a]	Sound field audiometry	Primary outcome not specified in paper
							Hearing impairment[a]	Visual reinforcement audiometry	Disagreement between reviewers on primary outcome requiring further discussion. Consensus reached that hearing impairment was considered to be the primary outcome
							Hearing impairment[a]	Pure tone audiometry	
							Middle ear function	Tympanometry	
							Hearing screening failures		
							Number of VTs	Audiometry number of VTs	
									Hearing impairment included as an outcome with three methods of assessment
8	Frable *et al.* (1985)[69]	Case series	6-monthly follow-up for 10 years	36	Children with CP	CP children with bilateral VTs placed	Number of VTs	Note review	Primary outcome not specified in paper
							Tympanic membrane atresia[a]	Otoscopic findings	
							COM[a]	Otoscopic findings	
							Incidence of AOM	Parent report	
							OME	Otoscopic findings	
							Hearing loss	Audiometry	
9	Sheahan *et al.* (2002)[70]	Case series	Mean follow-up 6 years and 11 months	104	Children with CP. Dublin, Ireland	CP children treated for OME	COM[a]	Otoscopic findings	Primary outcome not specified in paper
							Cholesteatoma	Otoscopic findings	Cholesteatoma, retraction and perforation have been grouped in this paper and are considered to be signs of COM
							Retraction	Otoscopic findings	
							Perforation	Otoscopic findings	
							OME	Otoscopic findings	
							Hearing impairment	Pure tone audiometry	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
10	Hubbard *et al.* (1985)[71]	Retrospective study	60–132 months	48	Children with CP aged 3 months at study entry. Pittsburgh, USA	VT at 3 months vs. VT 30 months	Scarring of the tympanic membrane[a]	Otoscopy	Primary outcome not specified in paper
							Hearing loss	Pure tone audiometry	
							Hearing loss	Speech reception thresholds	
							Middle ear pressure	Pneumatic otoscopy	
							Middle ear pressure	Tympanometry	
							OME	Otoscopic findings	
							Hypernasality of speech	5-point scale completed by speech and language therapist	
							Consonant articulation	Test of articulation	
							Social maturity	Vineland Social Maturity Scale	
							Social maturity	Wechsler Intelligence Scale for Children	
							Self-esteem	Coopersmith Self-Esteem Inventory	
							Behaviour	Child Behaviour Checklist	
							Nasal resonance	Assessment by speech and language therapist	
							Perforation	Otoscopic findings	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
11	Gordon *et al.* (1988)[72]	Retrospective study	Follow-up 10–16 years post treatment	50	Children with CP. New Zealand	VTs vs. no treatment	Hearing impairment	Pure tone audiometry	Primary outcome not specified in paper
							Middle ear pressure	Tympanometry	
							Tympanosclerosis[a]	Otoscopy	
							Scarring of the tympanic membrane[a]	Otoscopy	
							Retraction	Otoscopy	
							OME[a]	Otoscopy	
							Otorrhoea[a]	Otoscopy	
							Cholesteatoma[a]	Otoscopy	
12	Robson *et al.* (1992)[73]	Retrospective	30–60 months' follow-up	74	Children with CP. Bristol, UK	VTs vs. no treatment	Otorrhoea	Otoscopy	Primary outcome not specified in paper
							Perforations of the tympanic membrane	Otoscopy	Communication disorder (verbal comprehension and expression) as an outcome with four methods of assessment
							Tympanosclerosis	Otoscopy	
							Episodes of otalgia		
							Middle ear pressure	Tympanometry	
							Atelectasis of the tympanic membrane	Otoscopy	
							Retraction of the tympanic membrane	Otoscopy	
							Velopharyngeal insufficiency	SLT assessment	
							OME	Otoscopy	
							Hearing impairment	Pure tone audiogram	
							Educational performance	Parent questionnaire (is child above/below average)	
							Level of speech therapy support required	Level of speech therapy support required	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
							Behaviour	Parent questionnaire (is child above/below average)	
							Communication disorder (verbal comprehension and expression)[a]	Cleft-related articulation at SLT assessment	
							Communication disorder (verbal comprehension and expression)[a]	Phonological problem at SLT assessment	
							Communication disorder (verbal comprehension and expression)[a]	Language difficulties at SLT assessment	
							Communication disorder (verbal comprehension and expression)[a]	Learning difficulties at SLT assessment	
13	Greig *et al.* (1999)[74]	Case series	60 months	36	Children with CP. London, England	CP children with bilateral VT tubes	Hearing improvement	Parental questionnaire	
							Change in otorrhoea	Parental questionnaire	
							Parental satisfaction with VT treatment[a]	Parental questionnaire	
							Speech improvement	Parental questionnaire	
							Receptive language	SLT assessment	
							Expressive language	SLT assessment	
							Speech development	SLT assessment	
							Global development	Unclear	
							Hearing impairment	Audiometry (not details given)	
							Nasal escape	SLT assessment	
							Cleft speech	SLT assessment	
							Perforation	Presume otoscopy	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
14	Shaw *et al.* (2003)[75]	Retrospective study	10 years	72	Children with CP. Liverpool, UK	Children grouped into LAHSAL cleft classification	Speech – resonance[a]	SLT assessment	Primary outcome not specified in paper
							Speech – articulation[a]	SLT assessment	
							Number of grommets	Number of grommets	
15	Freeland and Evans (1981)[76]	Retrospective study	Followed 6-monthly for 48 months	68	Children with CP. Oxford, UK	Recruited at birth with regular follow-up	OME	Otoscopic findings	
							Retraction	Otoscopic findings	
							Perforation	Otoscopic findings	
							Tympanosclerosis of the tympanic membrane	Otoscopic findings	
							Language development[a]	Reynell Developmental Language Scale	
16	Zheng *et al.* (2003)[77]	RCT	6-month postoperative (VTs), 20-month postoperative (control)	62	Children with CP. China	Palatoplasty ($n = 24$) vs. palatoplasty + VT ($n = 38$)	Presence of OME[a]	Unknown	Article in Chinese. Abstract and Ponduri *et al.* 2009[1] review used
							Hearing levels	Unknown	
17	Liu *et al.* (2004)[78]	Case series	2 weeks–18 months postoperatively	19	Children with CP. China	Unilateral VT vs. other ear as control with no VT	Complications	Unknown	Article in Chinese. Abstract and Ponduri *et al.* 2009[1] review used
							Hearing loss[a]	Audiometry	
							Middle ear status	Unknown	
18	Tanpowpong *et al.* (2007)[79]	Cohort	10 months	23	6 children with CP and 17 without CP. Bangkok	Myringotomy and VT insertion	Hearing impairment[a]	Pure tone audiometry	Primary outcome not specified in paper
							Middle ear pressure	Tympanometry	
							Otorrhoea	Otoscopy	
							Time to extrusion of VTs	Note review	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
19	Civelek *et al.* (2007)[5]	Retrospective	72-month follow-up	41	56 children with CP, 15 children without CP and history of VT insertion. Turkey	CP vs. non-CP with VTs	Perforation[a]	Otoscopic findings	Primary outcome not specified in paper
							Tympanosclerosis[a]	Otoscopic findings	
							Cholesteatoma[a]	Otoscopic findings	
							Retraction[a]	Otoscopic findings	
							Hearing impairment[a]	Pure tone audiometry	
							Middle ear pressure	Tympanometry	
							Velopharyngeal insufficiency	Speech assessment (method not specified)	
20	Phua *et al.* (2009)[80]	Retrospective	Minimum 2 years, maximum 15 years	234	Children with CP. Auckland, New Zealand	VT ($n = 45$) vs. no treatment ($n = 189$)	Hearing loss[a]	Pure tone audiogram	Primary outcome not specified in paper
							Recurrent AOM	Note review	
							Persistent OME	Otoscopic findings	
							Retraction	Otoscopic findings	
							Perforation	Otoscopic findings	
							Cholesteatoma	Otoscopic findings	
							Subjective hearing loss	Note review	
							Number of VTs		
21	Reiter *et al.* (2009)[81]	Retrospective	6 years	116	Children with CP. Germany	Divided age and type of cleft then ± VTs	Cholesteatoma[a]	Otoscopic findings	Primary outcome not specified in paper
							Hearing loss	Pure tone audiogram	
							Middle ear pressure	Tympanometry	
							Atelectasis of the tympanic membrane[a]	Otoscopic findings	
							Perforations of the tympanic membrane[a]	Otoscopic findings	
							Retraction of the tympanic membrane[a]	Otoscopic findings	
							OME	Otoscopic findings	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
22	Szabo *et al.* (2010)[82]	Retrospective	5 years	86	Children with CP. Connecticut, USA	All cases VT	Hearing[a]	Newborn hearing screening	Primary outcome not specified in paper
							Number of surgeries	Note review	
							Atelectasis of the tympanic membrane	Otoscopic findings	
							Perforations of the tympanic membrane[a]	Otoscopic findings	
							Retraction of the tympanic membrane	Otoscopic findings	
							Tympanosclerosis	Otoscopic findings	
							Scarring of the tympanic membrane	Otoscopic findings	
23	Hornigold *et al.* (2008)[83]	Long-term follow-up data of RCT	20 years post-VT insertion in original RCT	7	Children with CP who had participated in previous RCT. UK	VT insertion vs. control	Hearing loss	Pure tone audiometry	Primary outcome not specified in paper In this study primary outcomes were grouped in the paper as symptomatology
							Middle ear function	Tympanometry	
							Mucosal COM	Otoscopic findings	
							Cholesteatoma (squamous COM)	Otoscopic findings	
							Otorrhoea[a]	Patient interview	
							Subjective hearing loss[a]	Patient interview	
							Otalgia[a]	Patient interview	
							Vertigo[a]	Patient interview	
							Tinnitus[a]	Patient interview	
							Need for further surgery[a]	Patient interview	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
24	Merrick *et al.* (2007)[84]	Cohort study	Mean 8 years of age. Follow-up post-palatoplasty	100	50 children with CP, 50 children without CP. UK	VT ($n = 50$) vs. control ($n = 50$)	OME	Otoscopy	Primary outcome not specified in paper
							Speech intelligibility[a]	CP speech assessment audit form	
							Speech nasality[a]	Assessment audit form	
							Nasal air flow[a]	Assessment audit form	
							Consonant production[a]	Assessment audit form	
							Cleft type characteristics[a]	Assessment audit form	
25	Curtin *et al.* (2009)[85]	Prospective cohort	6-months post-palate repair	33	Children with CP. Stanford, USA	VT at 9 months with a 6-month follow-up	Incidence of otorrhoea[a]	Parent report	Hearing impairment as an outcome with three methods of assessment
							VT patency	Tympanometry	
							Hearing impairment	Behavioural audiometry	
							Hearing impairment	Sound field audiometry	
							Hearing impairment	Newborn infant hearing screen	
26	Mandel *et al.* (2002)[86]	RCT	4 weeks	111	Children without CP. Pittsburgh, USA	Bilateral VT vs. bilateral myringotomy vs. no surgery	Middle ear effusion[a]	Pneumatic otoscopy	Primary outcome not specified in paper
							Middle ear pressure	Tympanometry	
							Recurrence of OME following resolution	Note review	
							Incidence of AOM	Note review	
							Hearing impairment	Age-appropriate hearing test (procedures varied according to age)	
							Adverse events	Incidence of hyperactivity, increased appetite, vomiting, irritability, diarrhoea, abdominal discomfort, rash	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
27	Casselbrant *et al.* (2009)[87]	RCT	Up to 36 months	98	Children without CP. Pittsburgh, USA.	Myringotomy + VT ($n = 32$), adenoids with myringotomy + VT ($n = 32$), adenoidectomy with myringotomy ($n = 34$)	Percentage of time with OME[a]	Pneumatic otoscopy	Percentage of time with OME as an outcome with three methods of assessment
							Percentage of time with OME[a]	Tympanometry	
							Percentage of time with OME[a]	Otoscopy	
							Requirement for further surgery	Note review	
							Incidence of AOM	Outpatient assessment	
							Incidence of otorrhoea	Outpatient assessment	
							VT functional status	Tympanometry	
							Perforation of the tympanic membrane	Otoscopy	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
28	Koivunen *et al.* (2004)[88]	RCT	2 years	180	Children without CP. Oulu, Finland	Adenoidectomy ($n = 60$), chemoprohpylaxis ($n = 60$), placebo ($n = 60$)	Intervention failure[a]	Two acute episodes in 2 months or three in 6 months based on symptom diaries; or middle ear effusion for at least 2 months as assessed by study otolaryngologist using pneumatic otoscope	Necessity to visit doctor – definition of doctor not specified in paper, would assume that the outcome describes unplanned visits to the GP and not planned study visits to the consultant
							Incidence of AOM	Symptom diary	
							Necessity to visit doctor	Symptom diary	
							Requirement for antibiotics	Symptom diary	
							Days with rhinitis	Symptom diary	
							Days with earache	Symptom diary	
							Days with fever	Symptom diary	
							Incidence of adverse events	Symptom diary	
29	Matilla *et al.* (2003)[89]	RCT	Follow-up until age 2 years, mean follow-up 7 months	137	Children without CP. Helsinki, Finland	Bilateral VTs ($n = 63$) vs. bilateral VTs + adenoidectomy ($n = 74$)	Rate of AOM[a]	Otoscopy	Rate of AOM as an outcome with two methods of assessment
							Rate of AOM[a]	Symptomology	
							Rate of otitis media episodes caused by *S. pnemoniae*	Culture	
							Rate of otitis media episodes caused by *H. influenzae*	Culture	
							Rate of otitis media episodes caused by *M. catarrhali*	Culture	
							Middle ear pressure	Tympanometry	
							Number of days with otorrhoea	Follow-up card	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
30	Paradise *et al.* (1990)[90]	RCT and cohort	3 years	99 in RCT and 114 in cohort	Children without CP. Pittsburgh, USA	Adenoidectomy ($n = 99$) vs. control ($n = 114$)	Proportion of time with otitis media[a]	Interpolation of visit data. Pneumatic otoscopy and biweekly enquiries by nurse	Proportion of time with otitis media
							Number of VT insertions	Case note review	
							Number of days when experienced otalgia	Biweekly enquiries by nurse	
							Number of days receiving antibiotics	Biweekly enquiries by nurse	
31	Paradise *et al.* (1999)[91]	RCT	Up to 3 years	461 (304 in three-way trial and 157 in two-way trial)	Children without CP. Pittsburgh, USA	Adenoidectomy ($n = 100$) vs. adenotonsillectomy ($n = 180$) vs. control ($n = 181$)	Number of episodes of AOM[a]	Not specified	
							Proportion of time with otitis media	Interpolation of visit data. Pneumatic otoscopy and biweekly enquiries by nurse	
							Number of VT insertions	Case note review	
							Number of days when experienced otalgia	Biweekly enquiries by nurse	
							Number of myringotomies	Not specified	
							Number of days receiving antibiotics	Biweekly enquiries by nurse	
32	Rynneldagoo *et al.* (1978)[92]	Prospective controlled study	24 months	76	Children without CP. Sweden	Adenoidectomy ($n = 37$), control ($n = 39$)	Change in frequency of common cold	Unknown	Primary outcome unknown
							Change in frequency of purulent otitis media	Unknown	
							Change in frequency of serious otitis media	Unknown	
							Change in frequency of nasal obstruction	Unknown	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
33	Gates *et al.* (1987)[93]	RCT	24 months	491	Children without CP. Texas, USA	Bilateral myringotomy ($n = 107$) vs. bilateral VTs ($n = 129$) vs. adenoidectomy ($n = 130$) vs. adenoidectomy + bilateral VTs ($n = 125$)	Time with hearing loss	Unknown	Primary outcome unknown
							Time with OME	Unknown	
							Time to recurrence of OME	Unknown	
							Requirement for further VT insertion	Unknown	
34	Hammaren-Malmi *et al.* (2005)[94]	RCT	12 months	217	Children without CP. Helsinki, Finland	Adenoidectomy + bilateral VTs ($n = 109$) vs. bilateral VTs only ($n = 108$)	Number of AOM episodes in 12 months[a]	Patient diary and review by GP (primary care doctor)	IAB and NH agreed only one outcome in paper. This was noted under outcome measures in the paper but not explicitly as a primary outcome
35	Roydhouse (1980)[95]	RCT	36 months	169	Children without CP. Auckland, New Zealand	Bilateral VT + adenoids ($n = 50$), bilateral VT no adenoids ($n = 50$), control ($n = 69$)	Presence of OME	Unknown	Primary outcome unknown
							Requirement for repeated grommets	Unknown	
							Number of relapses	Unknown	
36	Black *et al.* (1990)[96]	RCT	2 years	149	Children without CP. Oxford, UK	Adenoidectomy, myringotomy and VTs ($n = 37$) vs. adenoidectomy and VTs ($n = 38$) vs. myringotomy and VTs ($n = 37$) vs. VTs ($n = 37$)	Hearing impairment[a]	Pure tone audiometry	Main outcomes stated as hearing loss, results of impedance tympanometry and parental views on child progress. Primary outcome not specifically stated
							Developmental progress	Parental opinion of child's progress	
							Presence of an abnormal tympanogram	Impedance tympanometry	
							Adverse side effects of treatment	Parental opinion (favourable, uncertain or unfavourable)	
							Requirement for further surgery	Note review	
							Parents opinion on treatment	3-point scale	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
37	Dempster *et al.* (1993)[97]	RCT	12 months	78	Children without CP. Glasgow, UK	Adenoidectomy + VTs ($n = 37$) vs. VTs only ($n = 35$)	Presence of OME[a]	Otoscopy	Presence of OME likely to be primary outcome but paper states two primary outcomes, presence of OME and hearing
							Presence of OME[a]	Tympanometry	
							Hearing[a]	Pure tone audiometry	
							Tympanosclerosis	Otoscopy	
							Perforation	Otoscopy	
							Retraction	Otoscopy	
38	Maw *et al.* (1999)[98]	RCT	18 months	182	Children without CP. Bristol, UK	Adenoidectomy ($n = 47$) vs. adenoidectomy and tonsilectomy ($n = 47$) vs. control ($n = 56$)	Hearing loss	Age-appropriate hearing test	Expressive language, verbal comprehension (grouped as language development)
							Expressive language[a]	Reynell Developmental Language Scale scores at 9 and 18 months	
							Verbal comprehension[a]	Reynell Developmental Language Scale scores at 9 and 18 months	
							Mental development	Griffiths Mental Development Scales	
							Middle ear pressure	Tympanometry	
							Presence of OME	Otoscopy	
							Requirement for further VTs	Note review	
39	Rach *et al.* (1991)[99]	RCT	6 months	43	Children without CP. Netherlands	VTs vs. no surgery	Language development – verbal expression[a]	Reynell Developmental Language Scale	Primary outcome not specified in paper
							Language development – verbal comprehension[a]	Reynell Developmental Language Scale	
							Middle ear pressure	Tympanometry	
							Duration of VT tube in situ	Tympanometry	
							Presence of OME	Tympanometry	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
40	Rovers *et al.* (2000)[100]	RCT	12 months	187	Children without CP. Netherlands	VT ($n = 93$) vs. no surgery ($n = 94$)	Expressive language[a]	Schlichting Test for Expressive Language	Primary outcome not specified in paper but study designed to detect a difference in language development
							Expressive language[a]	Lexi test	
							Comprehensive language[a]	Reynell Developmental Language Scale	
							Hearing loss	Visual reinforcement audiometry	
							IQ	Bayley Scale of Infant Development	
							OME	Otoscopy	
							OME	Tympanometry	
41	Johnston *et al.* (2004)[101]	Screening followed by RCT	Follow-up to age 3 years	429	Children without CP. Pittsburgh, USA	VT ($n = 216$) vs. delayed VT ($n = 213$)	Tympanoscerosis[a]	Otoscopy	Primary outcome not specified in paper Outcome, tympanic membrane abnormalities which includes tympanosclerosis, fibrosis, atrophy, retraction pocket, perforation and cholesteatoma
							Fibrosis[a]	Otoscopy	
							Atrophy[a]	Otoscopy	
							Retraction pocket[a]	Otoscopy	
							Perforation[a]	Otoscopy	
							Hearing impairment	Pure tone audiometry	
							Cholesteatoma[a]	Otoscopy	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
42	Paradise *et al.* (2001)[102]	RCT	Follow-up to age 3 years	429	Children without CP. Pittsburgh, USA	VT ($n = 216$) vs. delayed VT ($n = 213$)	Cognition[a]	General cognitive index of McCarthy Scales of Children's Abilities	Primary outcome not specified in paper
							Hearing loss[a]	Age-appropriate hearing tests	
							Behaviour[a]	Child Behaviour Checklist	
							Receptive language[a]	Peabody-revised picture vocabulary test	
							Expressive language[a]	Number of different words	
							Expressive language[a]	Percentage of consonants correct – revised	
							Expressive language[a]	Mean length of utterance in morphemes	
							Parental distress[a]	Parental stress index, short form	
							Duration of OME	Pneumatic otoscopy	
							Duration of OME	Tympanometry	
43	Maw (1983)[103]	RCT	12 months	222	Children without CP. Bristol, UK	Adenoidectomy ($n = 36$) vs. no surgery ($n = 33$) vs. adenotonsillectomy ($n = 34$)	Presence OME[a]	Pneumatic otoscopy	Primary outcome not specified in paper Although other methodologies listed these were to confirm OME as part of patient screening and were not used as an outcome

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
44	Zielhus *et al.* (1989)[104]	Screening followed by RCT	Up to age 4 years	43	Children without CP. Netherlands	VT ($n = 22$) vs. no treatment ($n = 21$)	Presence OME	Tympanometry	Primary outcome not specified in paper
							OME	Otoscopy	
							Verbal comprehension[a]	Reynell Developmental Language Scale	Verbal comprehension and verbal expression grouped as language development
							Verbal expression[a]	Reynell Developmental Language Scale	
45	Fiellau-Nikolajsen *et al.* (1980)[105]	RCT	6 months post operative	42	Children without CP. Aarhus, Denmark	Myringtomy with adenoids ($n = 20$) vs. myringotomy only ($n = 22$)	Middle ear pressure[a]	Tympanometry	Primary outcome not specified in paper but middle ear pressure considered to be the primary outcome by the reviewers
							Presence of OME	Otoscopy	
							Duration of OME	Otoscopy (repeated over time)	
							Hearing impairment	Pure tone audiometry	
							Presence of middle ear reflexes	Impedance audiometry	
							Middle ear pressure[a]	Tympanometry	
46	Nguyen *et al.* (2004)[106]	RCT	Minimum 12 months	63	Children without CP. Quebec, Canada	VTs and adenoidectomy ($n = 23$) vs. VTs only ($n = 40$)	Recurrence AOM[a]	Patient questionnaire	Primary outcome not specified in paper
							Recurrence of OME over 3 months[a]	Patient questionnaire	Outcome in paper considered to be the primary outcome was 'treatment failure' as defined by the three outcomes listed and marked with 'a'
							Reinsertion of VTs[a]	Patient questionnaire	
47	Paradise *et al.* (2007)[107]	RCT	Follow-up at ages 9–11 years	429 in original study. 391 in follow-up	Children without CP	VTs ($n = 216$) vs. delayed treatment ($n = 213$)	Literacy	Woodcock Reading Mastery tests	Primary outcome not specified in paper
							Literacy	Oral fluency – the number of words in a grade-level passage read correctly	Multiple methods of assessment for outcomes listed in paper

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
							Literacy	Spelling and writing samples subtests of the Woodcock–Johnson III Tests of Achievement	Follow up of 2003 and 2005 paper
							Phonological awareness	Ellison and Rapid Letter Naming subtests of the Comprehensive Test of Phonological Processing	
							Auditory processing ability	Children's version of the Hearing in Noise Test	
							Attention, impulsivity and psychological functioning	Disruptive Behavior Disorders Rating Scale	
							Attention, impulsivity and psychological functioning	Child Behaviour Checklist	
							Attention, impulsivity and psychological functioning	Impairment Rating Scale	
							Attention, impulsivity and psychological functioning	Social Skills Scale of the Social Skills Rating System	
							Intelligence and academic achievement	Wechsler Abbreviated Scale of Intelligence	
							Intelligence and academic achievement	Calculation subtest of the Woodcock–Johnson III Tests of Achievement	
							Attention, impulsivity and psychological functioning	Continuous Performance Test – visual	
							Attention, impulsivity and psychological functioning	Continuous Performance Test – auditory	

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
48	Paradise *et al.* (2005)[108]	RCT	Follow-up at age 6 years	429	Children without CP. Pittsburgh, USA	VT ($n = 216$) vs. delayed treatment ($n = 213$)	Intelligence and academic achievement	Wechsler Abbreviated Scale of Intelligence	Primary outcome not specified in paper
							Receptive vocabulary	Peabody Picture Vocabulary Test	Follow-up of 2003 paper[109]
							Behaviour	Parent-reported inventories – Child Behaviour Checklist	
							Phonologic memory	Non-word repetition task	
							Auditory processing and language	SCAN test, a measure of central auditory processing	
							Vocabulary diversity	Conversational sample – number of different words	
							Sentence length and grammatical complexity	Conversational sample – length of utterances in morphemes	
							Speech sound production	Conversational sample – percentage of consonants correct	
							Parent–child stress	Parent–reported inventories – Parental Stress Index	

continued

TABLE 17 Characteristics of included papers including outcomes used (*continued*)

Study number	Author (year)	Study type	Duration of follow up	Sample size	Participants	Interventions	Outcomes measured	Method of measurement	Comments
49	Paradise *et al.* (2003)[109]	RCT	Follow up at age 4 years	429	Children without CP. Pittsburgh, USA	VT ($n = 216$) vs. delayed treatment ($n = 213$)	Cognition	McCarthy Scales of Children's Abilities	Primary outcome not noted as primary but main outcome. However, this included more than one outcome
							Receptive language	Peabody Picture Vocabulary Test – Revised	
							Phonological memory	Non-word repetition test	
							Expressive language	Word diversity (number of difference words)	
							Expressive language	Sentence length and grammatical complexity (mean length of utterance in morphemes)	
							Expressive language	Speech sound production (Percentage of Consonants Correct – Revised)	
							Parent–child stress	Parental Stress Index	
							Behaviour	Child Behaviour Checklist	

AOM, acute otitis media; COM, chronic otitis media; GP, general practitioner; IQ, intelligence quotient; n/a, not applicable; SLT, speech and language therapy.
a Primary outcome.

Generation of an outcome list for use in the Delphi survey of health professionals

The number of outcomes measured in an individual study varied with a median of 6 outcomes (range 1–14 outcomes) per paper. All outcomes measured were reviewed by NH and IAB, and grouped into possible outcome and outcome domain headings. After the initial review a total of 43 outcomes were listed under 13 domain headings (*Table 18*). Outcomes related to resource use were considered to be outside of the scope of the COS and would instead be considered as part of the VOI analysis (see *Chapter 5*). Consequently, the outcomes 'necessity to visit doctor' and 'level of speech therapy support required' were not considered in the initial list of 45 outcomes.

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
Outcomes related to COM	COM	COM[a]	8
		COM[a]	9
		Mucosal COM	23
		Fibrosis	41
	Cholesteatoma	Cholesteatoma	9
		Cholesteatoma[a]	11
		Cholesteatoma[a]	19
		Cholesteatoma	20
		Cholesteatoma[a]	21
		Cholesteatoma (squamous COM)	23
		Cholesteatoma	41
	Tympanosclerosis	Tympanosclerosis	1
		Scarring of the tympanic membrane	4
		Tympanosclerosis of the tympanic membrane	4
		Scarring of the tympanic membrane[a]	10
		Scarring of the tympanic membrane[a]	11
		Tympanosclerosis[a]	11
		Tympanosclerosis	12
		Tympanosclerosis of the tympanic membrane	15
		Tympanosclerosis[a]	19
		Scarring of the tympanic membrane	22
		Tympanosclerosis	22
		Tympanosclerosis	37
		Tympanosclerosis	41

continued

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
	Atelectasis	Atelectasis of the tympanic membrane	4
		Tympanic membrane atresia[a]	8
		Atelectasis of the tympanic membrane	12
		Atelectasis of the tympanic membrane[a]	21
		Atelectasis of the tympanic membrane	22
		Atrophy	41
	Persistent tympanic membrane perforation	Perforation	3
		Perforation of the tympanic membrane	5
		Perforation	9
		Perforation	10
		Perforations of the tympanic membrane	12
		Perforation	13
		Perforation[a]	15
		Perforation[a]	19
		Perforation	20
		Perforations of the tympanic membrane[a]	21
		Perforations of the tympanic membrane[a]	22
		Perforation of the tympanic membrane	27
		Perforation	37
		Perforation	41
		Chronic perforation	4
		Tympanic membrane	3
		Tympanic membrane on otoscopy	3

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
	Persistent tympanic membrane retraction	Retraction	9
		Retraction	11
		Retraction of the tympanic membrane	12
		Retraction[a]	15
		Retraction[a]	19
		Retraction	20
		Retraction of the tympanic membrane[a]	21
		Retraction of the tympanic membrane	22
		Retraction	37
		Retraction pocket	41
Outcomes related to behaviour	Behaviour	Behaviour	10
		Behaviour	10
		Behaviour	12
		Behaviour	12
		Behaviour[a]	42
		Behaviour[a]	42
		Attention, impulsivity and psychological functioning	47
		Behaviour	48
		Behaviour	49
Outcomes related to ear symptoms	Otalgia	Episodes of otalgia	12
		Otalgia[a]	23
		Days with earache	28
		Number of days on which ear pain occurred	30
		Number of days when experienced otalgia	31

continued

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
	Otorrhoea	Incidence of otorrhoea	1
		Incidence of otorrhoea	3
		Duration of otorrhoea	3
		Risk of otorrhoea	6
		Otorrhoea	11
		Otorrhoea	12
		Change in otorrhoea	13
		Otorrhoea	18
		Otorrhoea[a]	23
		Incidence of otorrhoea[a]	25
		Incidence of otorrhoea	27
		Number of days with otorrhoea	29
		Number of episodes of secondary otorrhoea	30
	Vertigo	Vertigo[a]	23
		Hearing impairment[a]	2
		Hearing impairment[a]	4
		Hearing	5
		Hearing impairment[a]	7
		Hearing screening failures	7
		Hearing loss	8
		Hearing impairment	9
		Hearing loss	10
		Hearing loss	10
		Hearing impairment	11
		Hearing impairment	12
		Hearing impairment	13
		Hearing improvement	13
		Hearing levels	16
		Hearing loss	17
		Hearing impairment[a]	18
		Hearing impairment[a]	19
		Hearing loss[a]	20
		Subjective hearing loss	20
		Hearing loss	21

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
		Hearing[a]	22
		Hearing loss	23
		Subjective hearing loss[a]	23
		Hearing impairment	25
		Hearing impairment	26
		Time with hearing loss	33
		Hearing impairment[a]	36
		Hearing[a]	37
		Hearing loss	38
		Hearing loss	40
		Hearing impairment	41
		Hearing loss[a]	42
		Hearing impairment	45
	Tinnitus	Tinnitus[a]	23
Outcomes related to nasal symptoms	Nasal obstruction	Nasal air flow[a]	24
		Change in frequency of nasal obstruction	34
	Rhinitis	Days with rhinitis	28
Outcomes related to development	Developmental progress	Global development	13
		Developmental progress	36
		Mental development	38
	Intelligence and academic achievement	Educational performance	12
		IQ	40
		Intelligence and academic achievement	47
		Intelligence and academic achievement	48
	Psychosocial development	Self-esteem	10
		Social maturity	10
		Psychosocial development	47
		Literacy	47
	Phonological memory	Phonologic memory	48
		Phonological memory	49
	Cognitive development	Cognition[a]	42
		Cognitive development	47
		Cognition	49

continued

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
Outcomes related to middle ear status	Eustachian tube function	Middle ear ventilation	1
		Middle ear ventilation	1
		Middle ear pressure	4
		Eustachian tube dysfunction[a]	5
		Middle ear status	6
		Middle ear function	7
		Middle ear pressure	10
		Middle ear pressure	11
		Middle ear pressure	12
		Middle ear status	17
		Middle ear pressure	18
		Middle ear pressure	19
		Middle ear pressure	21
		Middle ear function	23
		Middle ear pressure	26
		Middle ear pressure	29
		Presence of an abnormal tympanogram	36
		Middle ear pressure	38
		Middle ear pressure	39
		Middle ear pressure[a]	45
	Stapedial reflex	Presence of middle ear reflexes	45
		Stapedial reflex	4
Outcomes related to speech and language	Speech	Speech sound production	48
	Speech intelligibility	Speech intelligibility[a]	24
	Receptive language skills	Receptive language	13
		Receptive language[a]	42
		Receptive vocabulary	48
		Receptive language	49
		Auditory processing ability	47
		Auditory processing and language	48
	Expressive language skills	Expressive language	13
		Expressive language[a]	38
		Language development – verbal expression	39
		Expressive language	40

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
		Expressive language[a]	42
		Expressive language[a]	42
		Verbal expression[a]	44
		Vocabulary diversity	48
		Expressive language	49
		Expressive language	45
	Speech and language development	Speech development	13
		Speech development	47
		Phonological awareness	47
	Parents perspective of speech	Speech improvement (parent reported)	13
	Language skills – comprehension and expression	Communication disorder (verbal comprehension and expression)[a]	12
		Language development	47
		Language development – verbal	39
		Sentence length and grammatical complexity	48
		Language development	15
		Verbal comprehension[a]	38
		Verbal comprehension[a]	44
		Comprehensive language	40
	Consonant production – cleft vs. non-cleft speech difficulties	Consonant articulation	10
		Consonant production[a]	24
		Speech – articulation[a]	14
	Cleft-related speech patterns	Cleft speech	13
		Cleft type characteristics[a]	24
	Speech signs of velopharyngeal insufficiency	Velopharyngeal insufficiency	12
		Velopharyngeal insufficiency	19
		Speech – resonance[a]	14
		Speech nasality[a]	24
		Hypernasality of speech	10
		Nasal resonance	10
		Nasal escape	13
		Speech – resonance[a]	14
		Speech nasality[a]	24

continued

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
Outcomes related to otitis media	AOM	AOM	4
		Incidence of AOM	8
		Recurrent AOM	20
		Incidence of AOM	26
		Incidence of AOM	27
		Incidence of AOM	28
		Rate of AOM[a]	29
		Rate of otitis media episodes caused by *H. influenzae*	29
		Rate of otitis media episodes caused by *M. catarrhalis*	29
		Rate of otitis media episodes caused by *S. pnemoniae*	29
		Episodes of suppurative otitis media[a]	30
		Number of days on which antimicrobial treatment was received	30
		Incidence of AOM[a]	31
		Number of days receiving antibiotics	31
		Number of AOM episodes in 12 months	34
		Change in frequency of purulent otitis media	34
		Recurrence AOM[a]	46
		Requirement for antibiotics	28
		Days with fever	28
	OME	OME[a]	1
		OME	2
		OME[a]	3
		OME	4
		OME	8
		OME	9
		OME	10
		OME[a]	11
		OME	12
		OME	15
		Presence of OME	16

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
		Persistent OME	20
		OME	21
		OME	24
		Middle ear effusion[a]	26
		Recurrence of OME following resolution	26
		Percentage of time with OME	27
		Percentage of time with OME[a]	27
		Percentage of time with OME[a]	27
		Proportion of time with OME[a]	30
		Proportion of time with otitis media	31
		Time to recurrence of OME	33
		Time with OME	33
		Change in frequency of serious otitis media	34
		Number of relapses	35
		Presence of OME	35
		Presence of OME[a]	37
		Presence of OME[a]	37
		Presence of OME	38
		Presence of OME	39
		OME	40
		Duration of OME	42
		Presence of OME	43
		Presence of OME	44
		Presence of OME	45
		Duration of OME	45
		Recurrence of OME over 3 months[a]	46
		Intervention failure[a]	28
	Temporary tympanic membrane perforation	This outcome was identified from outcomes listed as part of persistent tympanic membrane perforation	

continued

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
Outcomes related to VTs (grommets)	Requirement for repeated VTs	Necessity for new VT	1
		Number of VTs until normal tympanometry	5
		Number of VTs	7
		Number of VTs	8
		Number of grommets	14
		Number of VTs	20
		Number of surgeries	22
		Need for further surgery[a]	23
		Requirement for further surgery	27
		Number of tympanostomy tube procedures	30
		Number of VT insertions	31
		Requirement for further VT insertion	33
		Requirement for repeated grommets	35
		Requirement for further surgery	36
		Requirement for further VTs	38
		Reinsertion of VTs[a]	46
	Necessity to remove VTs	Necessity to remove VT	1
	Early extrusion or blockage of VTs	Average time to extrusion of VT	5
		Time to extrusion of VTs	18
		VT patency	25
		VT functional status	27
		Duration of VT tube in situ	39

TABLE 18 Provisional list of outcomes identified from systematic review and provided to the SAG for review (*continued*)

Outcome domain	Outcome	Outcome identified in individual study	Study number of papers retrieved from systematic review (see *Table 17*)
Side effects of treatment	Side effects of treatment	Complications	17
		Adverse events	26
		Incidence of adverse events	28
		Adverse side effects of treatment	36
Upper respiratory tract infections	Upper respiratory tract infection	Change in frequency of common cold	34
Parent and child stress	Parental stress	Parental distress[a]	42
	Child stress	Parent–child stress	49
		Parent–child stress	48
Parental satisfaction with treatment	Parental satisfaction with treatment	Parental satisfaction with VT treatment[a]	13
		Parents opinion on treatment	36

AOM, acute otitis media; COM, chronic otitis media; IQ, intelligence quotient.
a Primary outcome.

The draft list of outcomes was then circulated to the mOMEnt SAG. The SAG comprised cleft clinicians representing speech and language therapists ($n = 2$), cleft surgeons ($n = 1$), ENT surgeons ($n = 2$), audiologists ($n = 2$) and clinical psychologists ($n = 1$). Members of the SAG reviewed outcomes relevant to their clinical field. The SAG also commented on the suitability of the overall domain under which outcomes are grouped.

Following review by the SAG and further review by the Study Management Group (SMG) the following actions were taken (*Figure 8*):

- The SAG were asked if 'temporary membrane retraction' should also be included as an outcome as temporary retraction was included. All agreed that it was not necessary to include this in the list of outcomes.
- The outcome 'language and comprehension' was removed as this is duplicated by the individual outcomes of 'receptive language' and 'expressive language'.
- Speech' was grouped with 'speech and language development'.
- Speech intelligibility was listed as a separate outcome.
- Outcomes related to behaviour were split into 'externalising behaviour' and 'internalising behaviour' based on the domains used in the Child Behaviour Checklist.
- 'Intelligence and academic achievement' was split into two individual outcomes.

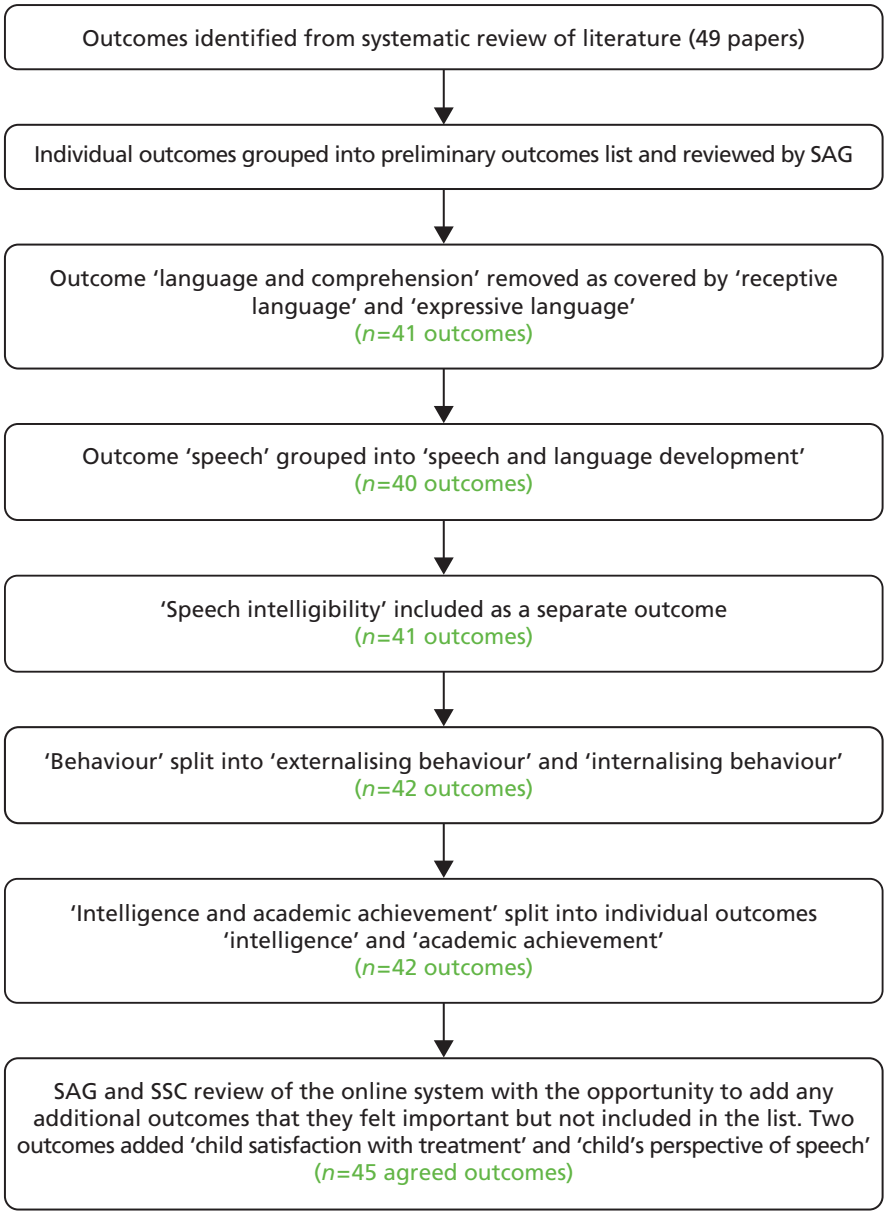


FIGURE 8 Development of final list of outcomes to be used in the Delphi survey of health professionals. SSC, Study Steering Committee.

Both the SAG and the Study Steering Committee (SSC) tested the online system with an opportunity to score each outcome and to add any additional outcomes that they felt were important. Two members of the SAG added outcomes related to HAs; however, these were outside of the scope of the COS, surgical management of OME, and were not included in the outcome list. The SSC noted two additional outcomes 'child's satisfaction with treatment' and 'child's perspective of speech' which were taken forward to the list of outcomes to be scored by clinicians.

The final list of outcomes used in round 1 of the clinician Delphi comprised 45 individual outcomes (43 identified from the systematic review of the literature and two by the SSC) grouped under 14 domains (*Table 19*). To aid all participants completing the Delphi an outcome tip was written for each outcome and suitability confirmed with the SAG.

TABLE 19 Domains, outcomes and outcome tips used in the health professional Delphi

Domain	Outcome	Tip (where there is no tip the outcome was considered to be understandable by all clinical roles)
Outcomes related to behaviour	Externalising behaviour	Externalising behaviours are directed outwards (e.g. having a tantrum)
	Internalising behaviour	Internalising behaviours are directed inwards (e.g. being withdrawn or lonely)
Outcomes related to COM	Atelectasis	Retraction of the thin tympanic membrane with loss of the normal middle ear space
	Cholesteatoma	Structure made of keratin not usually found in the middle ear. Has tendency to enlarge and cause recurrent ear discharge and hearing loss
	COM	Fluid in the middle ear persisting for over 3 months
	Persistent tympanic membrane perforation	Hole in the tympanic membrane
	Persistent tympanic membrane retraction	Tympanic membrane pulled backwards due to negative pressure in the middle ear
	Tympanosclerosis	Damage to the tympanic membrane with resultant deposition of calcium within tympanic membrane
Outcomes related to development	Academic achievement	
	Cognitive development	
	Developmental progress	Progress in relation to developmental milestones
	Intelligence	
	Literacy	Reading
	Phonological memory	Ability to remember a sequence of unfamiliar sounds
	Psychosocial development	
Outcomes related to ear symptoms	Hearing loss	Hearing ability below the normal range for the population
	Otalgia	Earache
	Otorrhoea	Ear discharge
	Tinnitus	The perception of noises without an accompanying external signal
	Vertigo	Hallucination of movement
Outcomes related to middle ear status	Eustachian tube function	The eustachian tube is responsible for equalising the pressure in the middle ear with the outside world. In eustachian tube dysfunction this does not happen appropriately leading to negative pressure in the middle ear
	Stapedial reflex	This reflex occurs in response to loud noise and is thought to play a protective role, limiting the potential for noise-induced hearing loss. When a noise of certain loudness is heard the stapedial muscle contracts making the ossicular chain of bones in the middle ear stiffer and less energy is carried to the cochlea

continued

TABLE 19 Domains, outcomes and outcome tips used in the health professional Delphi (*continued*)

Domain	Outcome	Tip (where there is no tip the outcome was considered to be understandable by all clinical roles)
Outcomes related to nasal symptoms	Nasal obstruction	Blocked nose
	Rhinitis	Inflammation of the lining of the nose. Sometimes as a result of infection or allergy
Outcomes related to otitis media	AOM	Infection involving the middle ear
	OME	The presence of fluid within the middle ear
	Temporary tympanic membrane perforation	Hole in the tympanic membrane
Outcomes related to speech and language	Consonant production	How clearly consonants are pronounced
	Consonant production – cleft related speech patterns	Unusual consonant patterns that speech and language therapists attribute to CP/velopharyngeal dysfunction
	Expressive language skills	The child's ability to produce language, including vocabulary, grammar, use of language, and sentence length and structure (syntax)
	Parent's perspective of speech	Parents' views of their child's speech
	Receptive language skills	The child's ability to understand spoken language
	Speech development	The predictable pattern of speech sound development leading to the production of words
	Speech intelligibility	How easy it is to understand a child's speech
	Speech signs of velopharyngeal insufficiency	E.g. nasal tone of voice, nasal airflow accompanying speech, visible nasal grimace during speech, prevalence of consonants ('m' 'n' 'ng') and absent ' "f" "ssh" "t" "d" '
Outcomes related to VTs (grommets)	Early extrusion or blockage of VTs	Extrusion means the VT falls out of the tympanic membrane
	Necessity to remove VTs	
	Requirement for repeated VTs	
Parent and child stress	Child stress	
	Parental stress	
Parent/child satisfaction with treatment	Parental satisfaction with treatment	
Side effects of treatment	Side effects of treatment	
Upper respiratory tract infections	Upper respiratory tract infection	Infection involving the ears, nose or throat
Additional outcomes from SAG	Child's satisfaction with treatment	
	Child's perspective of speech	Ability to hear speech noises

AOM, acute otitis media; COM, chronic otitis media.

Identification of outcomes of importance to parents and children with cleft palate

Introduction

The opinions of parents and children about the treatment of OME for children with CP are important because this group will experience both the benefits and adverse effects of treatments, and be involved in the decision-making about which treatment to have, and should therefore have opportunities to contribute to identification of the most appropriate outcomes for use in future trials of OME.

Outcomes of importance to parents and children with CP were identified from two sources:

1. qualitative interviews with a purposive sample of parents and children
2. an online survey of children with CP and their parents.

The qualitative interviews with parents and resulting important outcomes are described in *Chapter 3* and *Table 13*. The results of the interviews, based on responses to specific questions about outcomes, were cross-checked against the outcomes list generated from systematic review of the literature and, as the qualitative interviews were in parallel to the first round of the health professionals Delphi, any additional outcomes from round 1 to assess if any new outcomes were identified. IAB and NH mapped each outcome from the interviews against the list of outcomes after round 1 with no new outcomes identified. (*Table 20*). Outcomes mentioned in the interviews that were related to HAs or service delivery were not considered. Although parental stress was not specifically referenced by parents when asked about outcomes, it was a topic that was talked about repeatedly in interviews and this represented an outcome already identified from review of the literature – 'parental stress'.[111]

Although the qualitative interviews completed with parents and children gave rich in-depth information on outcomes of importance, the sample (recruited from two cleft centres), was relatively small in comparison with the UK population of children with a CP. To gain a broader view of important outcomes an online survey similar to that completed by health professionals was developed. This was suggested by the Cleft Lip & Palate Association (CLAPA) Children and Young Persons Council (CYPC) after discussions about the mOMEnt study. The CYPC suggested ways in which the Delphi survey could be adapted for children based on their experiences and these suggestions were taken on board.

Methods

Children aged 7–16 years and their parents were invited to take part in an online survey. In addition, after advice from the CLAPA Adult Voices Group, the survey was also made available to adults with a CP who had experience of OME.

The survey included questions to ensure eligibility, namely confirmation of a CP or a child with CP and experience of OME. For the survey of parents and children the term 'glue ear' was used instead as this was considered to be a more appropriate terminology.

Advice was sought from the National Research Ethics Service who did not consider that ethical approval was required for an online survey of parental and child opinion.

TABLE 20 Relationship of outcomes identified from qualitative interviews with outcomes identified in the systematic review

Outcome mentioned in interviews	Outcome domain from outcomes list used in health professionals survey round 1	Associated outcome/s from outcomes list used in health professionals survey round 1	Number of parents interviewed	Number of parents who mentioned as an outcome	Percentage of parents who mentioned outcome	Number of parents who mentioned as the most important outcome	Percentage of parents who mentioned as the most important outcome
Improvement in hearing	Outcomes related to ear symptoms	Hearing	36	30	83	21	58
Speech/ communication	Outcomes related to speech and language	Consonant production	36	14	39	4	11
		Consonant production – cleft-related speech patterns					
		Expressive language skills					
		Parent's perspective of speech					
		Receptive language skills					
		Speech development					
		Speech intelligibility					
		Speech signs of velopharyngeal insufficiency					
		Parent's perspective of speech					
		Child's perspective of speech					
Child is less frustrated	Outcomes related to behaviour	Externalising behaviour	36	2	6	2	6
Make the child more alert	Additional outcomes identified in round 1	Listening skills	36	2	6	2	6

Outcome mentioned in interviews	Outcome domain from outcomes list used in health professionals survey round 1	Associated outcome/s from outcomes list used in health professionals survey round 1	Number of parents interviewed	Number of parents who mentioned as an outcome	Percentage of parents who mentioned outcome	Number of parents who mentioned as the most important outcome	Percentage of parents who mentioned as the most important outcome
Glue ear not to return. Remove fluid from the ears	Outcomes related to otitis media	OME	36	8	22	1	3
Reduce pain/not to cause pain	Outcomes related to ear symptoms	Otalgia	36	7	19	2	6
Reduced number of ear infections	Outcomes related to otitis media	AOM	36	9	25	1	3
Improvement in school work	Outcomes related to development	Academic achievement	36	6	17	0	0
		Cognitive development					
		Developmental progress					
		Intelligence					
		Literacy					
Improved social interactions	Outcomes related to development	Psychosocial development	36	10	28	2	6
		Psychosocial well-being					
	Additional outcomes identified in round 1						

AOM, acute otitis media.

Survey content

Parents and children were asked to score a list of outcomes which was based on the outcomes list generated from the systematic review. Each outcome was reviewed by NH and IAB, and a plain-language alternative suggested that was appropriate for a child aged 7–10 years. This description was tested for readability using the National Institute of Adult Continuing Education Simplified Measure of Gobbledygook calculator[111] and was further checked for understanding with the CLAPA CYPC and the local Happy Faces Group. Some outcomes which related to specific clinical observations were combined so that parents and children scored a total of 36 outcomes representing the 45 outcomes identified from the systematic review and SAG/SSC contribution.

The same outcome wording was used for all participants with the exception of minor changes such as 'your'/'your child's' to ensure appropriate context (*Table 21*).

TABLE 21 Outcomes included in the online survey for parents, children aged 7–16 years and adults

Original outcome	Outcome domain	Age 7–10 years	Age 11–16 years	Parents	Adults
Internalising behaviour	Things about behaviour/things about your child's behaviour/things about behaviour	How lonely you feel, feeling like an outsider	How lonely you feel, feeling like an outsider	How lonely your child feels, feeling like an outsider	How lonely you felt, feeling like an outsider
Externalising behaviour	Things about behaviour/things about your child's behaviour/things about behaviour	How angry you are towards others	How angry you are towards others	How angry your child is towards others	How angry you were towards others
Atelectasis Persistent tympanic membrane retraction Tympanosclerosis	Things about having problems with your ears for a long time/things about your child having problems with their ears for a long time	Not having problems inside your ear caused by having lots of ear infections over a long time (more than 3 months)	Not having problems inside your ear caused by having lots of ear infections over a long time (more than 3 months)	Your child not having problems inside their ear caused by having lots of ear infections over a long time (more than 3 months)	Not having problems inside your ear caused by having lots of ear infections over a long time (more than 3 months)
Cholesteatoma	Things about having problems with your ears for a long time/things about your child having problems with their ears for a long time	Not having problems inside your ear caused by bad skin growing behind your ear drum	Not having problems inside your ear caused by bad skin growing behind your ear drum	Your child not having problems inside their ear caused by bad skin growing behind your ear drum	Not having problems inside your ear caused by bad skin growing behind your ear drum
COM	Things about having problems with your ears for a long time/things about your child having problems with their ears for a long time	Not having problems inside your ear caused by having glue ear for a long time (more than 3 months)	Not having problems inside your ear caused by having glue ear for a long time (more than 3 months)	Your child not having problems inside their ear caused by having glue ear for a long time (more than 3 months)	Not having problems inside your ear caused by having glue ear for a long time (more than 3 months)
Original outcome	Outcome domain children aged 7–16 years and adults/parents	Age 7–10 years	Age 11–16 years	Parents	Adults

TABLE 21 Outcomes included in the online survey for parents, children aged 7–16 years and adults (*continued*)

Original outcome	Outcome domain	Age 7–10 years	Age 11–16 years	Parents	Adults
Persistent tympanic membrane perforation	Things about having problems with your ears for a long time/things about your child having problems with their ears for a long time	Not having problems inside your ear caused by having a hole in your ear drum for a long time (more than 3 months)	Not having problems inside your ear caused by having a hole in your ear drum for a long time (more than 3 months)	Your child not having problems inside their ear caused by having a hole in your ear drum for a long time (more than 3 months)	Not having problems inside your ear caused by having a hole in your ear drum for a long time (more than 3 months)
Academic achievement	Things about school and making friends	How well you are doing at school	How well you are doing at school or college	How well your child is doing at school or college	How well you did at school or college
Cognitive development					
Developmental progress					
Intelligence					
Literacy					
Phonological memory					
Psychosocial development	Things about school and making friends	How well you are learning to make friends and speak to new people	How well you are learning to make friends and speak to new people	How well your child is learning to make friends and speak to new people	How well you learnt to make friends and speak to new people
Hearing	Things about how your ear feels and works/things about how your child's ear feels and works	How well you can hear	How well you can hear	How well your child can hear	How well you could hear
Otalgia	Things about how your ear feels and works/things about how your child's ear feels and works	How painful your ear is	How painful your ear is	How painful your child's ear is	How painful your ear was
Otorrhoea	Things about how your ear feels and works/things about how your child's ear feels and works	Not having infected liquid leaking out of your ear	Not having pus (infected liquid) leaking out of your ear	Your child not having pus (infected liquid) leaking out of their ear	Not having pus (infected liquid) leaking out of your ear
Tinnitus	Things about how your ear feels and works/things about how your child's ear feels and works	How much you hear buzzing or ringing noises	How much you hear buzzing or ringing noises	How much your child hears buzzing or ringing noises	How much you heard buzzing or ringing noises

continued

TABLE 21 Outcomes included in the online survey for parents, children aged 7–16 years and adults (*continued*)

Original outcome	Outcome domain	Age 7–10 years	Age 11–16 years	Parents	Adults
Vertigo	Things about how your ear feels and works/things about how your child's ear feels and works	How dizzy you feel	How dizzy you feel	How dizzy your child feels	How dizzy you felt
Eustachian tube function	Things about how the middle part of your ear works/ things about how the middle part of your child's ear works	How well a special tube in your ear works. If this tube does not work properly you might hear popping and crackling noises	How well a special tube in your ear works. If this tube does not work properly you might hear popping and crackling noises	How well a special tube in your child's ear works. If this tube does not work properly your might hear popping and crackling noises	How well a special tube in your ear worked. If this tube did not work properly you might have heard popping and crackling noises
Stapedial reflex	Things about how the middle part of your ear works/ things about how the middle part of your child's ear works	How well your ear works when it hears a loud noise	How well your ear works when it hears a loud noise	How well your child's ear works when it hears a loud noise	How well your ear worked when it heard a loud noise
Nasal obstruction	Things about how your nose feels/ things about how your child's nose feels	How well you can breathe through your nose	How well you can breathe through your nose	How well your child can breathe through their nose	How well you could breathe through your nose
Rhinitis	Things about how your nose feels/ things about how your child's nose feels	How much your nose feels runny or stuffy	How much your nose feels runny or stuffy	How much your child's nose feels runny or stuffy	How much your nose felt runny or stuffy
AOM	Things about glue ear and ear infections	Not having ear infections	Not having ear infections	Your child not having ear infections	Not having ear infections
OME	Things about glue ear and ear infections	Not having glue ear and being able to hear better	Not having glue ear and being able to hear better	Your child not having glue ear	Not having glue ear and being able to hear better
Temporary tympanic membrane perforation	Things about glue ear and ear infections	Not having a hole in your eardrum that only lasts for a few weeks	Not having a hole in your eardrum that lasts for a few weeks	Your child not having a hole in their eardrum that lasts for a few weeks	Not having a hole in your eardrum that lasts for a few weeks
Consonant production Consonant production – cleft-related speech patterns Expressive language skills	Things about talking	Being able to say all your words clearly and grown-ups and children understanding what you say	Being able to say all your words clearly and grown-ups and children understanding what you say	Your child being able to say all their words clearly so that adults and other children can understand what they said	Being able to say all your words clearly so that adults and other children could understand what you said
Parent's perspective of speech	Things about talking	How much you talk like someone without a CP	How much you talk like someone without a CP	How much your child talks like someone without a CP	How much you talked like someone without a CP

TABLE 21 Outcomes included in the online survey for parents, children aged 7–16 years and adults (*continued*)

Original outcome	Outcome domain	Age 7–10 years	Age 11–16 years	Parents	Adults
Receptive language skills	Things about talking	How well your parents think you are speaking	How well your parents think you are speaking	How well you think your child is speaking	How well your parents thought you were speaking
Speech development	Things about talking	Being able to listen and understand what other people say	Being able to listen and understand what other people say	Your child being able to listen and understand what other people say	Being able to listen and understand what other people say
Speech intelligibility	Things about talking	Speaking as well as other children the same age as you	Speaking as well as other children the same age as you	Your child speaking as well as other children who are the same age	Speaking as well as other children who were the same age as you
Speech signs of velopharyngeal insufficiency	Things about talking	Your speech not sounding different to other children	Your speech not sounding different to other children	Your child's speech not sounding different to other children	Your speech not sounding different to other children
Early extrusion or blockage of VTs	Things about grommets	How often your grommets/VTs fall out or do not work	How often your grommets/VTs fall out or do not work	How often your child's grommets/VTs fall out or do not work	How often your grommets/VTs fell out or did not work
Necessity to remove VTs	Things about grommets	Not needing another operation to take grommets/VTs out	Not needing another operation to take grommets/VTs out	Your child not needing another operation to take grommets/VTs out	Not needing another operation to take grommets/VTs out
Requirement for repeated VTs	Things about grommets	Not needing another operation to have new grommets/VTs because the old ones fell out	Not needing another operation to have new grommets/VTs because the old ones fell out	Your child not needing another operation to have new grommets/VTs because the old ones fell out	Not needing another operation to have new grommets/VTs because the old ones fell out
Child stress	Things about how you or your parents feel/things about how you or your child feels	How often you feel upset or angry	How often you feel upset or angry	How often your child feels tense or upset	How often you felt upset or angry
Parental stress	Things about how you or your parents feel/things about how you or your child feels	How often your parents feel upset or angry	How often your parents feel upset or angry	How often you feel tense or upset	How often your parents felt upset or angry
Parental satisfaction with treatment	Things about how well your child's treatment has worked	How well your parents think that HAs or grommets have improved your hearing	How well your parents think that HAs or grommets have improved your hearing	How well you think that HAs or grommets have improved your child's hearing	How well your parents thought that HAs or grommets improved your hearing
Side effects of treatment	Things about problems caused by treatment/things about problems caused by your child's treatment	Not having problems, that can sometimes happen, that are caused by a treatment you have for glue ear	Not having problems, that can sometimes happen, that are caused by a treatment you have for glue ear	Your child not having problems, that can sometimes happen, that are caused by a treatment they have for glue ear	Not having problems, that can sometimes happen, that are caused by a treatment you had for glue ear

continued

TABLE 21 Outcomes included in the online survey for parents, children aged 7–16 years and adults (*continued*)

Original outcome	Outcome domain	Age 7–10 years	Age 11–16 years	Parents	Adults
Upper respiratory tract infection	Things about infections in the ear, nose or mouth	Not having infections in your ear, nose or mouth	Not having infections in your ear, nose or mouth	Your child not having infections in their ear, nose or throat	Not having infections in your ear, nose or mouth
Child's satisfaction with treatment	Other things	How much you think treatment has made you better	How much you think treatment has made you better	How much your child thinks that treatment has made them better	How much you think treatment made you better
Child's perspective of speech	Other things	How normal you think you sound when you are talking	How normal you think you sound when you are talking	How normal your child thinks they sound when they are talking	How normal you thought you sounded when you were talking

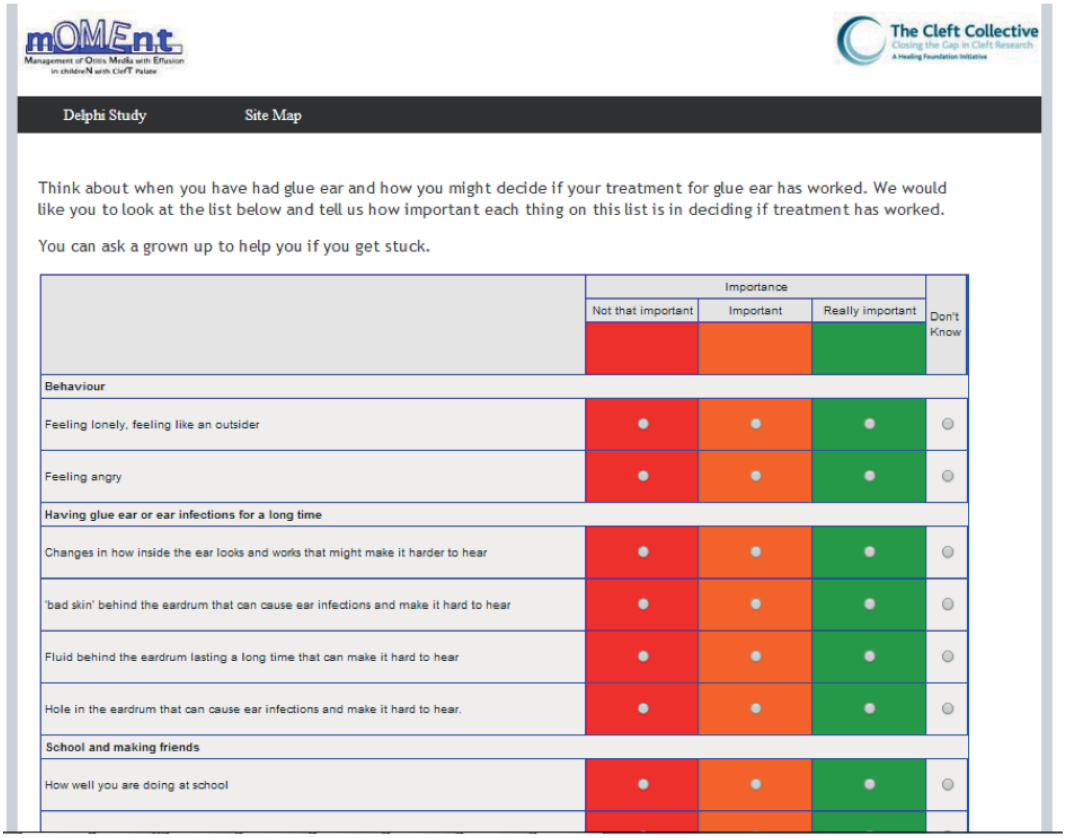
AOM, acute otitis media; COM, chronic otitis media.

Parents and adults were asked to consider the appropriate question described in *Table 22* and to then score each of the outcomes listed using the Grading of Recommendations, Assessment, Development and Evaluations (GRADE) scale of 1–9. In the online survey the scale was presented in the format 1–9 with 1–3 labelled as 'not that important', 4–6 labelled as 'important' and 7–9 labelled as 'really important'[112] (*Figure 9*). Parents were also provided with an option to add anything else that they considered relevant in a free-text box.

Children aged 7–16 years were shown the same list of outcomes as parents and were asked the question shown in *Table 22*. However, under the recommendation of the CYPC, scoring was completed using a traffic light system where scores of 1–3 were represented by a red box labelled as 'not that important', scores of 4–6 as an amber box labelled as 'important' and scores of 7–9 as a green box labelled 'really important' (see *Figure 9*). A free-text box was also provided so that participants could add anything else they considered relevant.

TABLE 22 Initial question asked prior to scoring outcomes for parents, adults and children with CP. The question asked of health professionals is included for comparison

Group	Initial question
Parents	Think about when your child has had glue ear and how you might decide if their treatment for glue ear has worked. We would like you to look at the list below and tell us how important each thing on this list is in deciding if treatment has worked
Adults	Think about when you have had glue ear, as a child, and how you might decide if your treatment for glue ear worked. We would like you to look at the list below and tell us how important each thing on this list is in deciding if treatment has worked
Children aged 7–16 years	Think about when you have had glue ear and how you might decide if your treatment for glue ear has worked. We would like you to look at the list below and tell us how important each thing on this list is in deciding if treatment has worked. You can ask a grown-up to help you if you get stuck
Health professionals	What outcomes influence your management of children with CP, with, or at high risk of, OME?



mOMEnt
Management of Otitis Media with Effusion in children with Cleft Palate

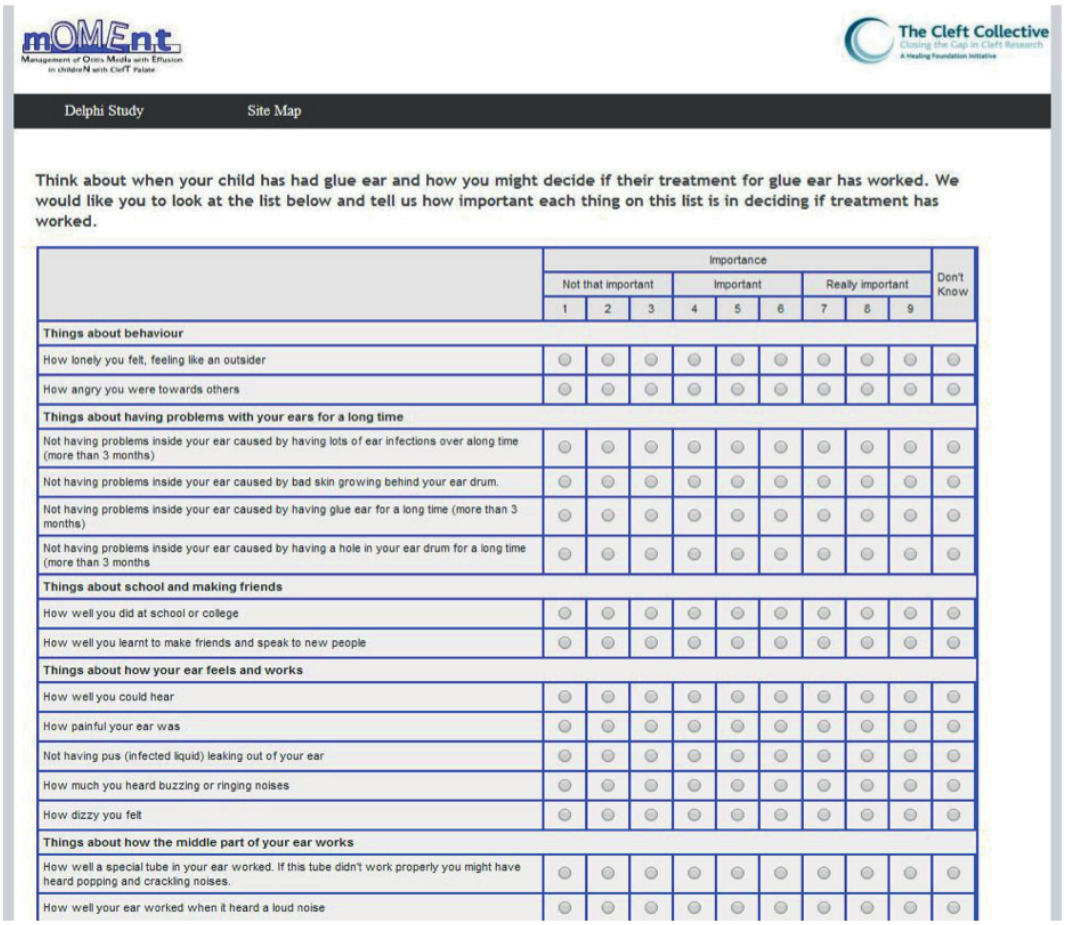
The Cleft Collective
Closing the Gap in Cleft Research
A Healing Foundation Initiative

Delphi Study Site Map

Think about when you have had glue ear and how you might decide if your treatment for glue ear has worked. We would like you to look at the list below and tell us how important each thing on this list is in deciding if treatment has worked.

You can ask a grown up to help you if you get stuck.

	Importance			
	Not that important	Important	Really important	Don't Know
Behaviour				
Feeling lonely, feeling like an outsider	○	○	○	○
Feeling angry	○	○	○	○
Having glue ear or ear infections for a long time				
Changes in how inside the ear looks and works that might make it harder to hear	○	○	○	○
'bad skin' behind the eardrum that can cause ear infections and make it hard to hear	○	○	○	○
Fluid behind the eardrum lasting a long time that can make it hard to hear	○	○	○	○
Hole in the eardrum that can cause ear infections and make it hard to hear.	○	○	○	○
School and making friends				
How well you are doing at school	○	○	○	○



mOMEnt
Management of Otitis Media with Effusion in children with Cleft Palate

The Cleft Collective
Closing the Gap in Cleft Research
A Healing Foundation Initiative

Delphi Study Site Map

Think about when your child has had glue ear and how you might decide if their treatment for glue ear has worked. We would like you to look at the list below and tell us how important each thing on this list is in deciding if treatment has worked.

	Importance									
	Not that important			Important			Really important			Don't Know
	1	2	3	4	5	6	7	8	9	
Things about behaviour										
How lonely you felt, feeling like an outsider	○	○	○	○	○	○	○	○	○	○
How angry you were towards others	○	○	○	○	○	○	○	○	○	○
Things about having problems with your ears for a long time										
Not having problems inside your ear caused by having lots of ear infections over along time (more than 3 months)	○	○	○	○	○	○	○	○	○	○
Not having problems inside your ear caused by bad skin growing behind your ear drum.	○	○	○	○	○	○	○	○	○	○
Not having problems inside your ear caused by having glue ear for a long time (more than 3 months)	○	○	○	○	○	○	○	○	○	○
Not having problems inside your ear caused by having a hole in your ear drum for a long time (more than 3 months	○	○	○	○	○	○	○	○	○	○
Things about school and making friends										
How well you did at school or college	○	○	○	○	○	○	○	○	○	○
How well you learnt to make friends and speak to new people	○	○	○	○	○	○	○	○	○	○
Things about how your ear feels and works										
How well you could hear	○	○	○	○	○	○	○	○	○	○
How painful your ear was	○	○	○	○	○	○	○	○	○	○
Not having pus (infected liquid) leaking out of your ear	○	○	○	○	○	○	○	○	○	○
How much you heard buzzing or ringing noises	○	○	○	○	○	○	○	○	○	○
How dizzy you felt	○	○	○	○	○	○	○	○	○	○
Things about how the middle part of your ear works										
How well a special tube in your ear worked. If this tube didn't work properly you might have heard popping and crackling noises.	○	○	○	○	○	○	○	○	○	○
How well your ear worked when it heard a loud noise	○	○	○	○	○	○	○	○	○	○

FIGURE 9 Screen shots of online survey for children aged 7–16 years and parents.

The study management team discussed whether or not parents, children and adults should see a free-text box first, before the list of outcomes, as there were concerns that participants might not feel that they had a voice once the list was shown. However, feedback received from parents and children was that it would be better to have the list first as that would help them to think about things and whether or not anything was missing.

Participants

Participants of the survey for parents and children were identified using the CLAPA mailing list and social media pages with a potential reach of 4710 and 9564 respectively. There is likely to be substantial overlap with membership of multiple Facebook pages (Facebook, Inc., Cambride, MA, USA) and groups, but it was not possible to assess this. An individual e-mail was sent to all those on the CLAPA mailing list together with a reminder in their e-newsletter. A link was posted on the Facebook page which included the researcher's name (NH) and a link together with a photograph. Examples of the newsletter and social media post are provided in *Appendix 5*.

Results

Although the survey was sent to a large number it was only accessed 293 times. Of this, 252 answered the initial question regarding eligibility and of the 235 eligible only 22% completed the survey. The 51 responses received comprised 35 parents, eight adults and eight children. Of the eight children four were in the 7- to 10-years age group and four were aged 11–16 years (*Figure 10*).

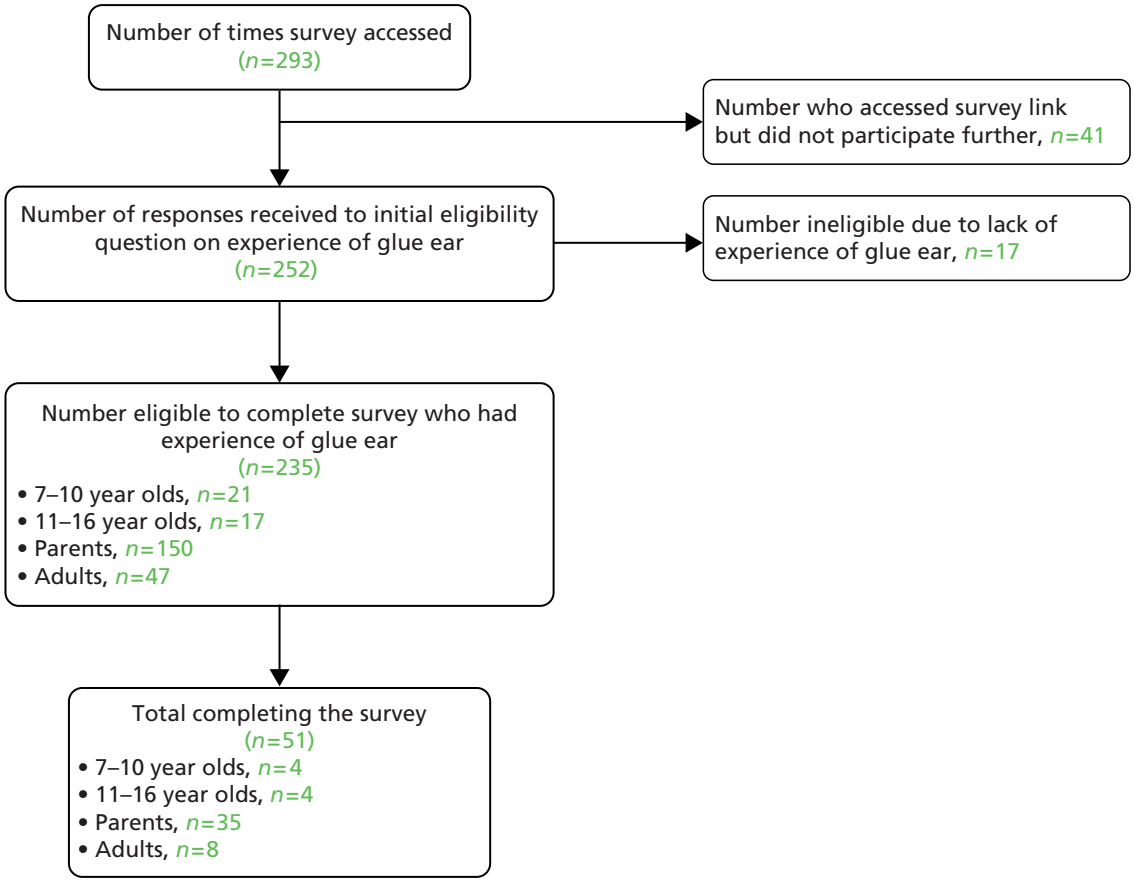


FIGURE 10 Response rate to parent and child survey.

Twenty-two free-text responses were received in addition to participant scores. Four responses were from adults who had completed the survey. The adult responses suggested that they might not have scored based on reflection of having OME as a child and instead based on current issues (*Table 23*). This, together with the small numbers of adults participating meant that adult scores were not considered further for the COS development.

TABLE 23 Online survey verbatim free-text responses: adults

Verbatim free-text response to 'Is there anything else you would like to tell us that is important to you?'	Outcomes mentioned	Notes
I had grommets because of my glue ear but it is only years later that I have found out that I have scars from them that I ***now have side effects and still hard of hearing and will need a hearing*** *aid in the near future.*	Tympanosclerosis; side effects of treatment; hearing	No new outcomes represented in free-text response
I'm not sure this makes sense do u want to know how important these things were to me or how much having glue ear effected these things? I would like to take part I'm 30 and still get clue ear. It effects my every day life especially though the winter. I'm miserable when I can't hear. ***I'm deaf now and don't want to go see my friends because I hate them having to repeat themselves constantly. It makes me so angry I want to punch someone or sometimes it makes me so sad I don't want to be around anymore***.	Externalising behaviour; internalising behaviour; AOM; upper respiratory tract infections; OME; chronic OME; psychosocial well-being	No new outcomes represented in free-text response. Free-text response suggests that this comment is based on current experience rather than when a child
It's the worst part of my life. ***When I get to many colds*** *and I can feel my ears going it's my worst feeling ever. I think at least deaf people have a constant deafness and can get used to poor hearing . . .*		
The only treatment I got when I was younger were grommets. They worked well but I loved the sea and couldn't swim. Later in childhood I those swimming over grommets and ear infections. The joke was ull go surfing all weekend then not again for a few months whilst I got ear infections and my ears recovered.		
Now I have hearing aids when my ears are bad but they r rubbish because my hearing fluctuates and the sounds I hear are muffled still. Hearing aids just make my hearing hat louder muffled noises.		
More research is need in this area and your test scale is very poor in terms of its wording a Likert scale would have been more appropriate for your results analysis.		No new outcomes represented in free-text response
I also feel that lots of people have been denied the appropriate care and attention when it comes to grommets/ glue ear etc and not enough has been done to amend this.		
This is the first time I have heard of this term today. I had tubes inserted as a child and removed. Now as a 50 year old adult I have major hearing loss.		No new outcomes represented in free-text response. Free-text response suggests that this comment is based on current experience rather than when a child

AOM, acute otitis media.
Items considered to represent an outcome are shown in bold text.

Free-text responses from children aged 7–10 years and 11–16 years did not mention any new outcomes but did reiterate some outcomes already in the outcomes list (*Table 24*). There were 15 free-text responses provided by parents and one of these represented a new outcome that was not included in the original outcomes list (*Table 25*). The new outcome 'hyperacusis', sensitivity to loud noises, was included in round 3 of the Delphi survey for health professionals.

TABLE 24 Online survey verbatim free-text responses: children aged 7–16 years

Child's age (years)	Verbatim free-text response to 'Is there anything else you would like to tell us that important to you?'	Outcomes mentioned	Notes
7–10	*If I keep getting ear infections or if I need to get another operation.*	AOM; requirement for repeated VT (could also be necessity to remove VTs as both require another operation); cholesteatoma	Cholesteatoma would also require operation therefore added. No new outcomes represented in free-text response
11–16	*Think that it should be know straight away whether an operation is needed for glue ear instead of having loads of operations, the grommets need to last longer and make sure they work so you don't need an operation every 6 months . . .*	Necessity to remove VTs; requirement for repeated VTs; early extrusion	No new outcomes represented in free-text response
11–16	*Unable to complete questionnaire as my glue ear of which i still suffer has been has been controlled with a hearing aid and is reliant on myself and my mum to decide if it needs to be worn and is also reliant on my mum chasing appointments which are few and far between. My mum says it is the one part of the cleft palate service that is lacking.*		No new outcomes represented in free-text response

AOM, acute otitis media.

TABLE 25 Online survey verbatim free-text responses: parents

Verbatim free-text response to 'Is there anything else you would like to tell us that important to you?'	Outcomes mentioned	Notes
*Glue ear significantly affects the **development of a child at crucial ages**. School teachers do not understand intermittent hearing loss and are usually not sympathetic or helpful to a child with glue ear (due to lack of understanding of the nature of the condition) and **often label a child as 'lazy' when they are inattentive**. Glue ear significantly affcts a **child's behaviour**.*	Internalising behaviour; externalising behaviour; listening skills; developmental progress	No new outcomes represented in free-text response
*Grommets being put in during the Cleft surgery meant the decision was a no brainer. It is hard to gauge how well they worked, but **not hard to remember how many ear infections he had**, whilst they were in. **Now they are out, and he still has glue ear**, our choices have been given on the basis of his hearing, not any other issues glue ear may cause (no dizziness, and his speech is very good) – as the decision is hearing versus ear infections, short term hearing aids would SEEM to be the best bet for us (it's about how **he hears and how that affects his making friends/learning,** not what that looks like. What I worry about, having completed this survey, is that we don't know the true COST of not having grommets (do they improve the condition or just the effects?)*	AOM; hearing; psychosocial well-being; academic achievement; cognitive development; intelligence; literacy; phonological memory; psychosocial development; OME	No new outcomes represented in free-text response

TABLE 25 Online survey verbatim free-text responses: parents (*continued*)

Verbatim free-text response to 'Is there anything else you would like to tell us that important to you?'	Outcomes mentioned	Notes
Having the treatment explained verbally AND visually to the child and parents in a way in which they will understand. Depending on the age of the child, have a folder available for them to take home with the details and/or photo's of key members of staff, the ward/medical setting where they will be staying, including pictures of the theatre and anaesthetic rooms where the ops take place, before they are admitted to hospital. And the chance to visit the ward during the pre-op assessment for those children who need it.	No outcomes noted	Information needs only. No new outcomes represented in free-text response
*I have had swim plugs made for my daughter who had long term grommets fitted after **short term grommets fell out**. If only we'd been made more aware of how crucial it is to prevent water from entering the ear canal after this kind of treatment we could have spared her **countless years of ear infections!** She uses them in the shower too. I had to go to a private hospital to have them made as she wasn't offered them routinely on the NHS. I forgot the plugs for one swimming lesson and she got a horrible bleeding infection that lasted 6 weeks which proved how essential the plugs are.*	AOM; early extrusion of VTs	No new outcomes represented in free-text response
*I have put everything as being very important, because to me nothing is more important than the health and happiness of my child, but perhaps the survey would gather better data by asking How Successful treatment has been and how often issues and arrise and the severity of these issues. My child has **constant glue ear** but in consultation with her doctors we decided **not to have further grommit surgery as the first surgery was unsuccessful and caused my child to become extremely unwell due to breathing problems and affects of anaesthetics**. There is no mention of bone conductor or other hearing aids. My daughter uses this attached to glasses and we are happy that it is having a clear beneficial affect.*	COM; OME; requirement for repeated VTs; hearing; side effects of treatment	No new outcomes represented in free-text response
*I think early treatment for glue ear is better my daughter did not have grommets fitted until she was 5. After having the grommets put in there was a very noticeable change to her **hearing and speech**.*	Hearing; consonant production – cleft-related speech patterns; expressive language skills; receptive language skills; speech development; speech intelligibility; speech signs of velopharyngeal insufficiency; parent's perspective of speech; child's perspective of speech	No new outcomes represented in free-text response
*I wish that he had been screened before he started school. His hearing probs not identified until he had started school and by that time he had **started to drift off in lessons because he could not hear properly, also had constant colds and stuffy noses**.*	Listening skills; hearing; upper respiratory tract infection; rhinitis	No new outcomes represented in free-text response

continued

TABLE 25 Online survey verbatim free-text responses: parents (*continued*)

Verbatim free-text response to 'Is there anything else you would like to tell us that important to you?'	Outcomes mentioned	Notes
*last time my son had glue ear, the volume, and consequently **tension levels in our house rose considerably**. My son struggles to sleep through the night when he has glue ear and **feels emotional as he is unable to fully participate in many social activities**. We opted for a 4th set of grommets and it was literally like flicking a switch – the atmosphere in our home changed almost immediately. We appreciate that should the glue ear re-occur, we will have to treat the subsequent hearing loss with hearing aids but appreciated as parents being given the option of either hearing aids or grommets last time around as this seems to help all aspects of the glue ear rather than just the hearing issue.*	Parental stress; child stress; psychosocial well-being; internalising/ externalising behaviour	No new outcomes represented in free-text response
*My child is 5 years old and started school last September. He has a cleft lip and pallet. Since having Grommits put in he has more **confidence and has come out off he's shell. In fact he never stop's talking**. We are pleased with results. Thank you*	Psychosocial well-being	No new outcomes represented in free-text response
My daughter has had hearing aids (short time) and gromits. I am astounded at the differance gromits have made to her. Her behaviour has totally changed, gone is my clingy girl. Now she runs off in playgroup by herself to play with others. And her speech has really come on. I am angry therefore that I had to really fight to get her gromits. Hearing aids were strongly pushed. I have seen for myself how great gromits have been for her so will gladly do battle again on her behalf to get more when the current ones come out. However my daughter has many medical issues and this is something that I would really prefer not to have to worry about.	Psychosocial well-being; internalising and externalising behaviour; consonant production – cleft-related speech patterns; expressive language skills; receptive language skills; speech development; speech intelligibility; speech signs of velopharyngeal insufficiency; parent's perspective of speech; child's perspective of speech	No new outcomes represented in free-text response
*My son had grommets fitted and by the time we went to a cleft clinic just over a week later one was out and sitting on the ear canal – the grommets made no discernible difference. We then decided to have hearing aids and they have transformed our lives. **I didn't realise how much my son could not hear until he had hearing aids and I used to get very angry at his seemingly bad behaviour – as a family we are happier and more content**. The audiologist and consultant had a 'wait and see' approach to my son's hearing, to see if the glue ear righted itself. In hindsight I think that condemned my son to 3 lost years where he couldn't hear properly when it was easily fixable. When we did ask the consultant for hearing aids we were left in no doubt that he was not happy at this choice, which I find strange.*	Early extrusion or blockage of VTs; externalising behaviour; parental stress; child stress; hearing	No new outcomes represented in free-text response

TABLE 25 Online survey verbatim free-text responses: parents (*continued*)

Verbatim free-text response to 'Is there anything else you would like to tell us that important to you?'	Outcomes mentioned	Notes
Professionals need to be aware that repeated grommet insertion is not always the best thing for a cleft child. The cause of the glue ear can be different than in a non-cleft child. My daughter was under the care of a normal ENT Surgeon and had ***one set of grommets, which fell out****. She was all set to have another set inserted, but was then moved to an ENT surgeon who specialised in cleft children. He was reluctant to carry out repeated grommet operations as each operation carries the* ***risks associated with a general anaesthetic/surgery, and the risk of the ear drum perforating****. His preference is to fit the child with hearing aids – they are non-invasive and should the child grow out of glue ear you simply stop using the hearing aid.*	Early extrusion of VTs; persistent/temporary tympanic membrane perforation; side effects of treatment.	No new outcomes represented in free-text response
The biggest measure of success for us as a family, was my daughter asking what the funny noise was in the supermarket, it was the 'beeping' of the checkout scanning items, she had never heard it before her grommets were inserted – ***clear indication of a great improvement in hearing****.*	Hearing	No new outcomes represented in free-text response
The length of time it has taken to get to the stage where the fitting of grommets was deemed required was 12 months which I find unacceptable. Then the surgeon hadn't even seen the last test results on the day of the fitting and refused to do one ear absolute joke, frustrating for myself and partner, distressing for my son who had already had a pre op at this point, some of the care we have received has been brilliant but on others has bordered on the negligent		No new outcomes represented in free-text response
There is also the problem of the ***variation in hearing levels*** *that children with glue ear can have – often on a daily basis, which means that hearing aids aren't always at the right level. Another issue is that children with glue ear are treated as deaf children rather than ones with fluctuating hearing and with other issues associated with their* ***hearing, such as earache, sensitivity to loud noise and subject to hearing popping sounds.*** *Also once hearing levels improve (if they do) to an extent where hearing aids are not needed, then no further treatment is considered, even though they still experience the pain, popping sounds, problems with loud noise and variation in hearing levels related to glue ear. There is no firm opinion as to when/if to have grommets fitted and yet once hearing rises just above the 20 dB level grommets are no longer an option even though glue ear is still causing a problem.*	AOM; eustachian tube function; hearing; otalgia	New outcome identified. Hyperacusis – sensitivity to loud noises

AOM, acute otitis media; COM, chronic otitis media.
Items considered to represent an outcome are shown in bold text.

Consensus matrix: parents and children

The results of the one-off survey of parents and children were reviewed against the definition of consensus agreed prior to the start of the study (see *Table 28*).[10] Using this definition parents and children had reached consensus in 26 and 9 outcomes respectively (*Table 26*).

TABLE 26 Outcome matrix for parent and child survey

Original outcome	Outcome scored by parents/children	Parents	Children
Internalising behaviour	How lonely you feel, feeling like an outsider		
Externalising behaviour	How angry you are towards others		
Atelectasis	Not having problems inside your ear caused by having lots of ear infections over a long time (more than 3 months)	✓	✓
Persistent tympanic membrane retraction			
Tympanosclerosis			
Cholesteatoma	Not having problems inside your ear caused by bad skin growing behind your ear drum		
COM	Not having problems inside your ear caused by having glue ear for a long time (more than 3 months)	✓	✓
Persistent tympanic membrane perforation	Not having problems inside your ear caused by having a hole in your ear drum for a long time (more than 3 months)		✓
Academic achievement	How well you are doing at school	✓	
Cognitive development			
Developmental progress			
Intelligence			
Literacy			
Phonological memory			
Psychosocial development	How well you are learning to make friends and speak to new people	✓	✓
Hearing	How well you can hear	✓	✓
Otalgia	How painful your ear is	✓	
Otorrhoea	Not having infected liquid leaking out of your ear		
Tinnitus	How much you hear buzzing or ringing noises	✓	
Vertigo	How dizzy you feel	✓	
Eustachian tube function	How well a special tube in your ear works. If this tube does not work properly you might hear popping and crackling noises	✓	
Stapedial reflex	How well your ear works when it hears a loud noise	✓	
Nasal obstruction	How well you can breathe through your nose		
Rhinitis	How much your nose feels runny or stuffy		

TABLE 26 Outcome matrix for parent and child survey (*continued*)

Original outcome	Outcome scored by parents/children	Parents	Children
AOM	Not having ear infections	✓	
OME	Not having glue ear and being able to hear better	✓	✓
Temporary tympanic membrane perforation	Not having a hole in your eardrum that only lasts for a few weeks		
Consonant production	Being able to say all your words clearly and grownups and children understanding what you say	✓	
Consonant production – cleft-related speech patterns			
Expressive language skills			
Parent's perspective of speech	How much you talk like someone without a CP	✓	
Receptive language skills	How well your parents think you are speaking	✓	✓
Speech development	Being able to listen and understand what other people say	✓	
Speech intelligibility	Speaking as well as other children the same age as you		
Speech signs of velopharyngeal insufficiency	Your speech not sounding different to other children	✓	
Early extrusion or blockage of VTs	How often your grommets/VTs fall out or do not work	✓	
Necessity to remove VTs	Not needing another operation to take grommets/VTs out	✓	✓
Requirement for repeated VTs	Not needing another operation to have new grommets/VTs because the old ones fell out	✓	✓
Child stress	How often you feel upset or angry	✓	
Parental stress	How often your parents feel upset or angry		
Parental satisfaction with treatment	How well your parents think that HAs or grommets have improved your hearing	✓	
Side effects of treatment	Not having problems, that can sometimes happen, that are caused by a treatment you have for glue ear	✓	
Upper respiratory tract infection	Not having infections in your ear, nose or mouth	✓	
Child's satisfaction with treatment	How much you think treatment has made you better	✓	
Child's perspective of speech	How normal you think you sound when you are talking	✓	

AOM, acute otitis media; COM, chronic otitis media.
Parents and children scored 36 outcomes; these are shown against the original 45 outcomes identified in the systematic review.

Identification of outcomes of importance to health professionals

Methods

To investigate outcomes of importance to clinicians a Delphi approach was adopted so that the anonymous opinions of the participant can be obtained in a way that gives equal influence to all who participate, and avoids an individual participant being overtly influenced by the opinions of any other participant.[113,114] An overview of the Delphi exercise is given in *Figure 11*. The method was prespecified at the start of the study and the protocol published.[10]

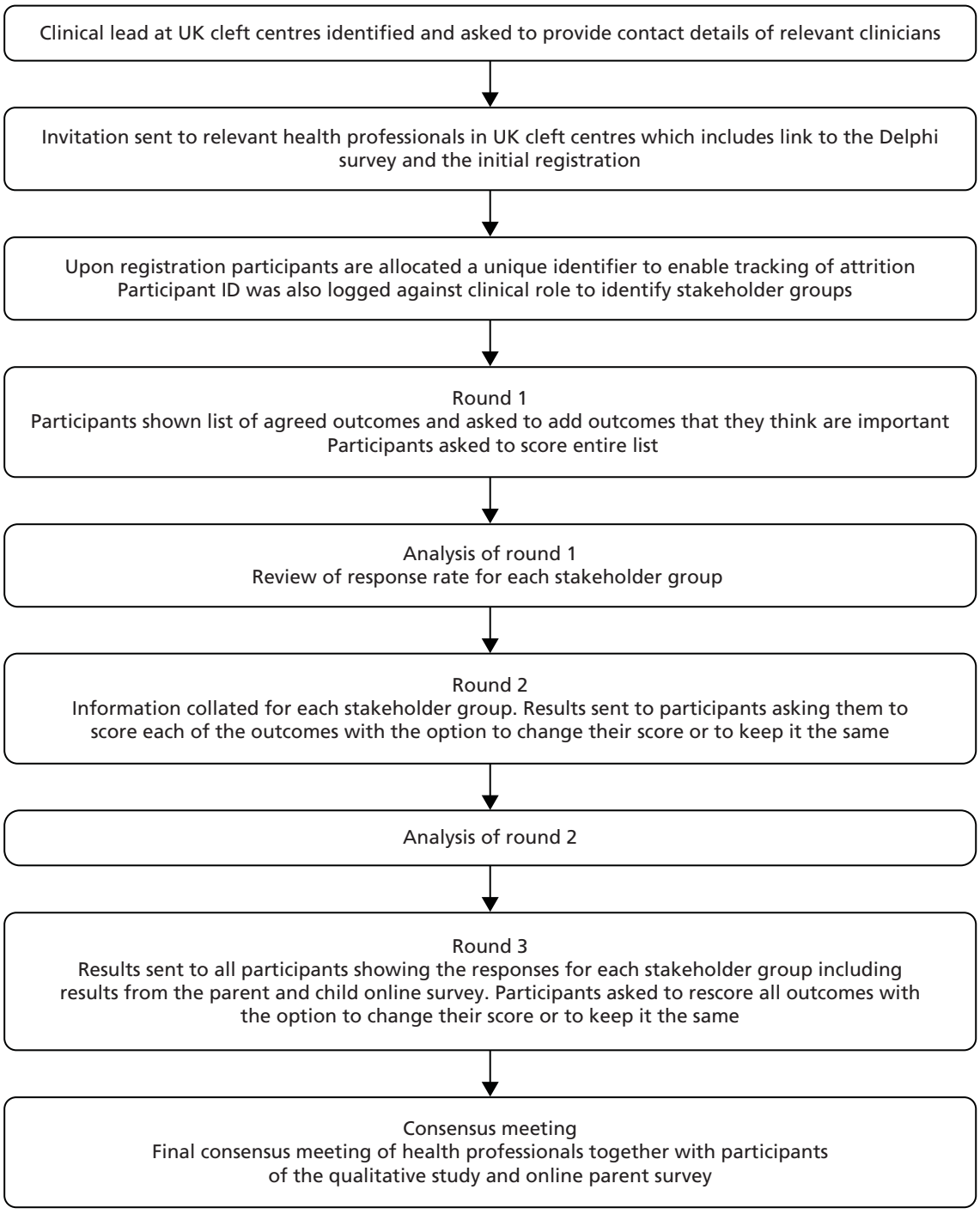


FIGURE 11 Overview of Delphi process. ID, identification.

Participants: health professionals

The Delphi study was conducted with the clinical teams from all UK cleft centres. Health professionals were selected from only UK centres due to time and cost constraints. The clinical lead at each of the UK cleft centres was asked to provide the names and roles of all members of the cleft team. The clinical roles considered key for completion of the Delphi were audiologists, ENT surgeons, speech and language therapists and specialist nurses. Other health professionals (e.g. paediatricians, clinical psychologists, clinical geneticists and cleft surgeons) were identified after consultation with the clinical director or cleft service co-ordinator at each cleft centre and were invited accordingly.

All health professionals, identified through correspondence with the clinical lead at UK sites, were contacted directly by e-mail with information about the mOMEnt study and instructions on how they could participate (see *Appendix 6*). A summary of invited health professionals is given in *Table 27*. All participants were asked to confirm their clinical role and cleft centre on registration for the mOMEnt Delphi survey.

At the beginning of the exercise participants were reminded of the importance of completing the entire Delphi process. Reminder e-mails were sent to aid completion of each round together with follow-up telephone calls to encourage completion. Participants who took part were given a unique identifier to allow tracking of attrition at each round. All data was stored against the unique identifier and participants were not able to identify other participants or individual responses.

Delphi survey: round 1

In the first round of the online questionnaire participants were asked to provide their name and e-mail address together with their clinical role and cleft centre. This information was stored in a separate database to the scores and used to provide the respondent with a unique identifier. The unique identifier was used to allow identification of individuals completing all rounds of the Delphi exercise and to track attrition between rounds.

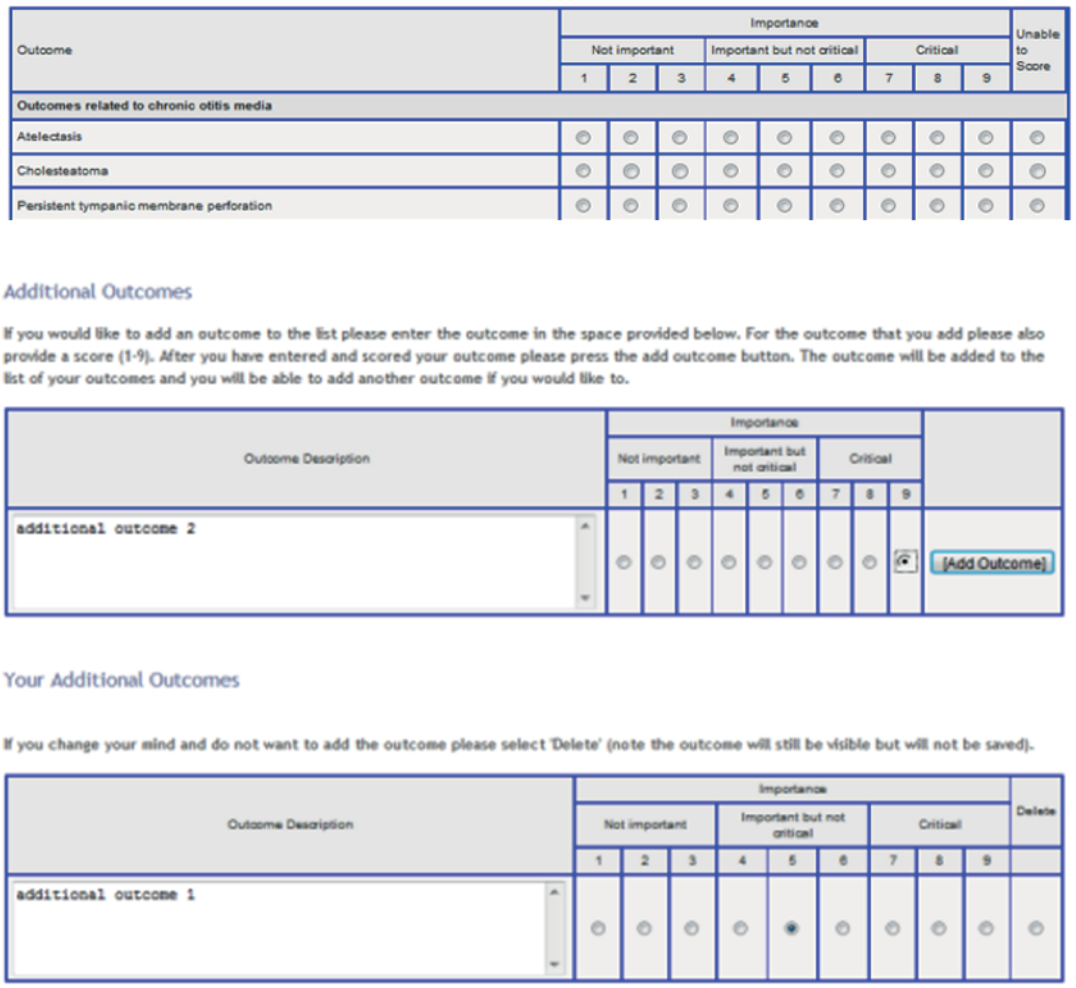
In round 1 participants scored the list of outcomes generated from the systematic review of the literature and through SAG and SSC contributions (see *Table 19*). Outcomes were ordered alphabetically, to avoid potential weighting of outcomes caused by the order in which they are displayed, and this was stated in the introductory text. Participants were asked to consider the question 'What outcomes influence your management of children with CP, with, or at high risk of, OME?' and to then score each of the outcomes listed using the GRADE scale of 1–9. In the Delphi exercise the scale was presented in the format 1–9 with 1–3 labelled as 'not important', 4–6 labelled as 'important but not critical' and 7–9 labelled as 'critical'. Participants were also provided with an option to add additional outcomes that they considered relevant together with a score for each outcome added (*Figure 12*).

Analysis of round 1

Additional outcomes listed by participants were reviewed and coded by two members of the study team (NH and IAB) to ensure they represent new outcomes. If there was uncertainty the SMG were consulted and the SAG if appropriate. For each outcome, the number of participants who scored the outcome and the distribution of scores (as percentage that have scored each outcome) were summarised by the stakeholder group. All outcomes were carried forward to round 2.

TABLE 27 Breakdown of health professionals invited to participate in the Delphi survey

Cleft centre number	Cleft surgeon, *n*	ENT surgeon, *n*	Geneticist, *n*	Specialist cleft nurse, *n*	Speech and language therapist, *n*	Paediatrician, *n*	Psychologist, *n*	Audiologist, *n*	Total at site, *N*
1	4	3	1	4	4	2	3	2	23
2	3	1	0	1	6	1	1	0	13
3	1	0	0	2	10	0	0	1	14
4	1	2	1	4	6	0	3	0	17
5	1	1	2	0	2	0	1	1	8
6	2	0	2	4	5	0	3	3	19
7	2	0	2	3	4	2	4	2	19
8	1	0	1	2	2	0	0	1	7
9	2	2	1	3	2	0	1	1	12
10	5	0	0	6	6	0	2	0	19
11	2	1	1	3	6	2	2	0	17
12	2	0	1	2	3	1	2	1	12
13	4	2	1	5	10	1	4	1	28
15	2	1	0	5	3	0	0	1	12
16	3	0	1	2	0	0	1	1	8
Total, *n*	35	13	14	46	69	9	27	15	228
Percentage representation	15	6	6	20	30	4	12	7	100



Outcome	Importance									Unable to Score
	Not important			Important but not critical			Critical			
	1	2	3	4	5	6	7	8	9	
Outcomes related to chronic otitis media										
Atelectasis	○	○	○	○	○	○	○	○	○	○
Cholesteatoma	○	○	○	○	○	○	○	○	○	○
Persistent tympanic membrane perforation	○	○	○	○	○	○	○	○	○	○

FIGURE 12 Example of online system for participants to score each outcome and to add additional outcomes they consider relevant.

Response rate in round 1

The number of participants in each stakeholder group who responded to round 1 were assessed following round 1 closure. Results will be presented as:

- total number of registrations
- breakdown of respondents who have completed the survey and their inclusion in the initial e-mail invitation
- total number of respondents who completed the round
- total number of respondents in each stakeholder group
- percentage of respondents compared with potential respondents as identified from the information provided by clinical leads
- percentage of respondents from other sources (not included in original e-mail invitation).

Continuation to round 2 was considered based on the response to round 1. If a low number of responders (< 10) was observed for one or more stakeholder groups, the Delphi protocol for future rounds was reviewed and revised. Where there is only one stakeholder group with a small number of respondents (potentially due to the sample available from clinical teams) then consideration was given to grouping with another stakeholder group. This was actioned in consultation with the SAG to ensure appropriateness of grouping.

Delphi survey: round 2

In round 2, each participant was presented with the number of respondents and distribution of scores for each outcome for their particular stakeholder group. Participants were shown their score from round 1, asked to consider responses from the other members of their stakeholder group and asked to rescore the outcome.

Any changes to scores in light of the stakeholder group or overall response were documented. Those who did not take part in round 1 and did not provide a score were not invited to participate in round 2 or participate further in the Delphi.

Analysis of round 2

The total number of participants invited to take part in round 2 was recorded. For each outcome, the number of participants who have scored the outcome and the distribution of scores was summarised by stakeholder group. All outcomes were carried forward to round 3.

Delphi round 3

In round 3, participants were shown the distribution of scores, for each outcome, for all stakeholder groups separately as well as a summary of the results from the survey of parents and children. Participants were asked to rescore all outcomes.

Analysis round 3

The total number of participants invited to take part in round 3 was recorded. For each outcome, the number of participants who scored the outcome and the distribution of scores was summarised together with the number of participants who scored the outcome in all rounds. Results of the stakeholder group response were compared with the whole group response and the percentage agreement used to determine the structure and focus of the final consensus meeting. Each outcome was classified as 'consensus in', 'consensus out' or 'no consensus' according to the classifications in *Table 28*.

Consensus meeting

The final phase of the study was a face-to-face consensus meeting with participants who had completed either the health professional Delphi, parent and child survey or the qualitative study and members of the SAG. Additional ethical approval was required to invite parents who participated in interviews to attend the consensus meeting. This was obtained from the Greater Manchester East Research Ethics Committee (reference 11/NW/0586).

The responses from round 3 of the health professionals Delphi and the parent and child survey were used to inform the structure and content of the consensus meeting.

Definition of consensus

The classification described in *Table 28* was used to determine if consensus was reached.

For consensus to have been reached that an outcome should be in the COS, agreement by the vast majority regarding the critical importance of the outcome, with only a small minority considering it to be not important at all, is required. Conversely, for consensus to have been reached that an outcome should not be in the COS, agreement by the vast majority regarding the lack of importance of the outcome, with only a small minority considering it to be critically important, is required.

Although the choice of thresholds is inevitably somewhat subjective, the specification of the definition of consensus upfront aimed to reduce the chance of consensus being defined post hoc in such a way as to bias the results towards the beliefs of the research team.

TABLE 28 Definition of consensus

Consensus classification	Description	Definition
Consensus in	Consensus that outcome should be included in the COS	≥ 70% participants scoring as 7–9 and < 15% participants scoring as 1–3
Consensus out	Consensus that outcome should not be included in the COS	≥ 70% participants scoring as 1–3 and < 15% of participants scoring as 7–9
No consensus	Uncertainty about importance of outcome	Anything else

Statistical considerations

Sample size

There is currently no standard method for sample size calculation in Delphi processes thus a pragmatic approach was taken. The number of participants in the present study is limited by the composition and number of UK cleft centres. Efforts were made to maximise the response rate across centres and stakeholder groups.

Results

Participation rates

The rate of completion was carefully monitored in each round against the number invited/eligible to participate. Regular reminder e-mails were sent which included information on the round together with the reason why a response was needed (see *Appendix 6*). In the third round a non-monetary incentive was used, e-mails were sent by a female member of staff and an image was included with the e-mail (as these methods have been previously reported to improve response rate).[115] In rounds 2 and 3 telephone calls were also made to participants and personalised e-mails were sent by the study co-ordinator in an effort to increase the response rate. The overall response rate per round is given in *Table 29*, the response rates by stakeholder group are discussed in the results of each round.

Round 1: health professionals

Of the 228 participants invited to round 1 a total of 104 completed the round with four of these participants providing partial scores only. Those who provided partial scores but scored > 50% of outcomes were invited to take part in round 2. The breakdown of participants by stakeholder group is shown in *Table 30*. After round 1 the free-text responses, from two of the three geneticists who had completed the round (*Table 31*), indicated that clinical geneticists were not directly involved in the care of children with OME and so were not invited to further rounds. In addition, of the three paediatricians who took part, all had a speciality in audiology, consequently for future rounds their scores were combined with audiologists into a new group 'audiologists and audiological physicians' as agreed by the SAG.

TABLE 29 Duration of each round of the Delphi survey and participation rates

Round	Date open	Date closed	Number of days round available (weekdays)	Number of participants eligible to complete	Number of participants completing round (% of total eligible)
1	22 November 2012	29 April 2013	158 (113)	228	104 (46)
2	13 June 2013	15 November 2013	155 (112)	99	85 (86)
3	9 December 2013	13 February 2014	66 (49)	81	73 (81)

TABLE 30 Round 1 response rates

Stakeholder group	Cleft surgeon	ENT surgeon	Geneticist	Specialist cleft nurse	Speech and language therapist	Paediatrician	Psychologist	Audiologist	Total
Number invited to participate	35	13	14	46	69	9	27	15	228
Number of respondents	15[a]	9[b]	3	18[c]	36[d]	3	13	7	104
Percentage completing round 1	43	69	21	39	52	33	48	46	46

a One participant scored only 44/45 outcomes.
b One participant scored only 29/45 outcomes.
c One participant scored only 35/45 outcomes.
d One participant scored only 30/45 outcomes.

TABLE 31 Verbatim comments provided in free-text response representing a general comment and not a potential outcome

Verbatim comment	Score	Stakeholder group
I do not think many of these outcomes are particularly relevant to a clinical geneticist	0	Clinical geneticist
I am fairly new to the role so still have so much to learn therefore my answers are based on my previous experience and what I have learnt so far therefore am not sure my responses will be reflective of specialist nurses as a whole.	0	Specialist cleft nurse
As a Clinical Psychologist, this study is really hard to respond to!	0	Psychologist
As a clinical geneticist my involvement is more in the diagnostic proccess rather than the decisions regarding surgery so I have been unable to comment on many of the survey questions.	0	Clinical geneticist
I found it difficult to complete this survey as it referred to 'your' management of children wth otitis media. There were many factors I gueesed were probably important but as an SLT it would not be my role to manage these. So I put unable to score for those	0	Speech and language therapist
Duplication of otological findings. Some of the questions used are too broad	0	ENT surgeon
found it difficult to fill in – unclear how to rate it.	0	Speech and language therapist
This is just a comment regarding signs of velopharyngeal incompetence/insufficiency – I do not think that this is an outcome that is indicative of the management of OME, but it may be correlated with and possibly contributory to the development of OME and therefore should be measured.	8	Cleft surgeon

Free-text responses provided in round 1

Eighteen free-text responses were provided by health professionals in round 1, all of which were reviewed by the SMG and SAG for classification. Eight responses represented a comment only with no associated score (see *Table 31*), two related to the use of HAs and were considered outside of the scope of the study (*Table 32*), the remaining eight described outcomes (*Table 33*). Of the eight potential outcomes two related to attention and behaviour, these outcomes are included in the broad domains of internalising and externalising behaviour and so were not carried forward. One participant listed both attention and listening, listening skills was considered to be a separate outcome and was taken forward to round 2. Two participants listed outcomes related to psychosocial well-being and this also represented a new outcome taken forward to round 2. Two responses were received about parental involvement and patient willingness to participate in speech activities, these were not considered to represent an outcome of treatment.

Following completion of round 1 the scores were compared with the definition of consensus to determine which stakeholder groups had reached the definition of 'consensus in'. The number of outcomes for which 'consensus in' was reached varied across stakeholder groups. ENT surgeons and audiologists/audiological physicians had reached consensus for the fewest outcomes ($n = 6$), whereas specialist cleft nurses considered 33 outcomes for inclusion in the COS.

The outcome with the most stakeholder groups (five out of six) reaching 'consensus in' after round 1 was 'hearing'.

Details of the stakeholder groups reaching consensus in for each outcome are shown in *Table 34*.

TABLE 32 Verbatim free-text responses related to HAs

Verbatim comment	Score	Stakeholder group	Resulting actions/decision
Use of hearing aid	6	Audiologist	SMG and SAG agreed that this was outside of the scope of the current study which was for surgical interventions
Outcomes related to hearing aids	9	Cleft surgeon	SMG and SAG agreed that this was outside of the scope of the current study which was for surgical interventions

TABLE 33 Round 1 verbatim free-text responses representing possible outcomes

Verbatim comment	Score	Stakeholder group	Resulting actions/decision
Attention/hyperactivity levels	8	Psychologist	Attention problems and ADHD are covered under the Child Behaviour Checklist as one of the domains categorised into higher domain of internalising/externalising activity
Attention and listening behaviour/skills	8	Speech and language therapist	Attention problems and ADHD are covered under the Child Behaviour Checklist as one of the domains categorised into higher domain of internalising/externalising activity Listening skills considered to represent a new outcome and added to the outcomes list for scoring in round 2
Self-esteem/self-concept/social confidence	9	Psychologist	After discussion with the SAG it was agreed to include an additional outcome in round 2 'psychosocial well-being'
Functional measure of how the patient is doing, e.g. making friends, chatting to peers, speaking on the phone etc.	9	Speech and language therapist	After discussion with the SAG it was agreed to include an additional outcome in round 2 'psychosocial well-being'
Prevention of otis media – affects of breast feeding/early palate surgery/nasal regurgitation when eating and drinking	7	Specialist cleft nurse	After discussion with the SAG this was not considered to represent an outcome related to treatment of OME
Auditory responsiveness to speech stimuli in classroom/at home/in speech therapy	9	Speech and language therapist	Auditory responsiveness was considered to be part of 'listening skills' which is considered to represent a new outcome and was added to the outcomes list for scoring in round 2
Willingness to participate in speech activities	7	Speech and language therapist	After discussion with the SAG this was not considered to represent an outcome related to treatment of OME
Parental involvement (doing activities/ exercises at home)	8	Speech and language therapist	After discussion with the SAG this was not considered to represent an outcome related to treatment of OME

ADHD, attention deficit hyperactivity disorder.

TABLE 34 Consensus matrix for round 1 of the health professionals survey

Outcome	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist/ audiological physician
Internalising behaviour			✓		✓	
Externalising behaviour			✓		✓	
Atelectasis						
Cholesteatoma	✓	✓				✓
COM	✓		✓	✓		
Persistent tympanic membrane perforation	✓		✓			
Persistent tympanic membrane retraction	✓		✓			
Tympanosclerosis	✓		✓			
Academic achievement			✓			
Cognitive development			✓			
Developmental progress			✓	✓	✓	
Intelligence						
Literacy			✓			
Phonological memory						
Psychosocial development			✓			
Hearing	✓	✓	✓	✓		✓
Otalgia						
Otorrhoea	✓					
Tinnitus						
Vertigo			✓			
Eustachian tube function	✓		✓			
Stapedial reflex			✓			
Nasal obstruction						
Rhinitis						
AOM	✓	✓	✓			
OME	✓	✓	✓	✓		
Temporary tympanic membrane perforation	✓		✓			
Consonant production			✓	✓		
Consonant production – cleft-related speech patterns	✓			✓		
Expressive language skills			✓	✓		

continued

TABLE 34 Consensus matrix for round 1 of the health professionals survey (*continued*)

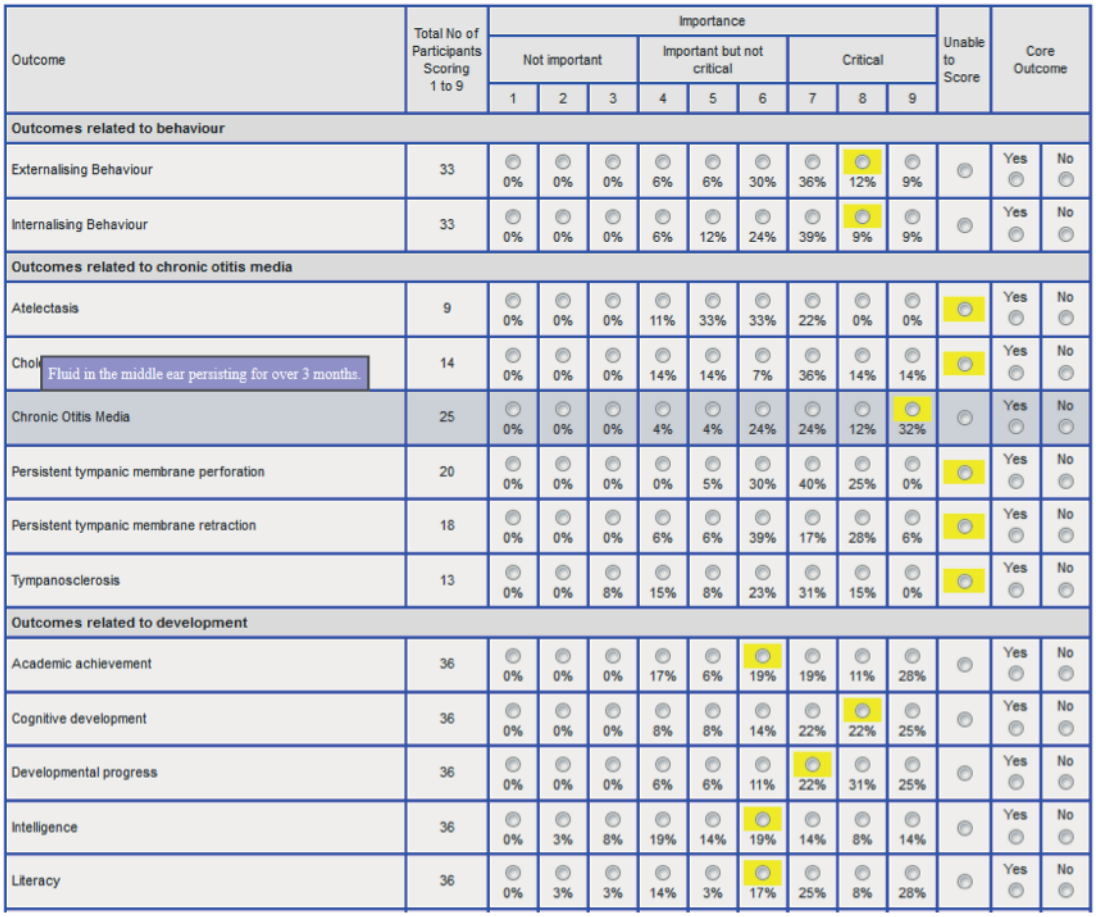
Outcome	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist/ audiological physician
Parent's perspective of speech			✓	✓		
Receptive language skills			✓	✓		
Speech development	✓		✓	✓		
Speech intelligibility	✓		✓	✓		
Speech signs of velopharyngeal insufficiency			✓	✓		
Early extrusion or blockage of VTs			✓			✓
Necessity to remove VTs	✓					✓
Requirement for repeated VTs	✓		✓			✓
Child stress			✓		✓	
Parental stress			✓		✓	
Parental satisfaction with treatment		✓	✓			
Side effects of treatment		✓	✓			✓
Upper respiratory tract infection						
Child's satisfaction with treatment			✓		✓	
Child's perspective of speech			✓	✓	✓	

AOM, acute otitis media; COM, chronic otitis media.
A ✓ indicates where the definition of 'consensus in' was achieved.

Round 2: health professionals

In round 2 a total of 47 outcomes, representing the original list of 45 plus the addition of two outcomes identified by health professionals' free-text responses in round 1, were scored by participants. Participants were shown their own score in round 1 together with the percentage of participants giving each score for their stakeholder group (*Figure 13*). A total of 85 responses were received in round 2 (86% of those completing round 1). The breakdown of participants by stakeholder group is shown in *Table 35*. After round 1 two participants, both speech and language therapists, went on maternity leave and so did not take part in future rounds of the study.

Three of the 85 participants completing the round provided partial scores only. The percentage of outcomes scored ranged from 21% to 62%. In this round all participants who provided scores were carried forward to round 3 irrespective of the percentage of outcomes scored.



Outcome	Total No of Participants Scoring 1 to 9	Importance									Unable to Score	Core Outcome	
		Not important			Important but not critical			Critical					
		1	2	3	4	5	6	7	8	9			
Outcomes related to behaviour													
Externalising Behaviour	33	0%	0%	0%	6%	6%	30%	36%	12%	9%		Yes	No
Internalising Behaviour	33	0%	0%	0%	6%	12%	24%	39%	9%	9%		Yes	No
Outcomes related to chronic otitis media													
Atelectasis	9	0%	0%	0%	11%	33%	33%	22%	0%	0%		Yes	No
Chol	14	0%	0%	0%	14%	14%	7%	36%	14%	14%		Yes	No
Chronic Otitis Media	25	0%	0%	0%	4%	4%	24%	24%	12%	32%		Yes	No
Persistent tympanic membrane perforation	20	0%	0%	0%	0%	5%	30%	40%	25%	0%		Yes	No
Persistent tympanic membrane retraction	18	0%	0%	0%	6%	6%	39%	17%	28%	6%		Yes	No
Tympanosclerosis	13	0%	0%	8%	15%	8%	23%	31%	15%	0%		Yes	No
Outcomes related to development													
Academic achievement	36	0%	0%	0%	17%	6%	19%	19%	11%	28%		Yes	No
Cognitive development	36	0%	0%	0%	8%	8%	14%	22%	22%	25%		Yes	No
Developmental progress	36	0%	0%	0%	6%	6%	11%	22%	31%	25%		Yes	No
Intelligence	36	0%	3%	8%	19%	14%	19%	14%	8%	14%		Yes	No
Literacy	36	0%	3%	3%	14%	3%	17%	25%	8%	28%		Yes	No

Fluid in the middle ear persisting for over 3 months.

FIGURE 13 Example of presentation of round 1 stakeholder group and individual scores.

TABLE 35 Round 2 response rates

Stakeholder group	Cleft surgeon	ENT surgeon	Geneticist	Specialist cleft nurse	Speech and language therapist	Paediatrician	Psychologist	Audiologist/ audiological physician	Total
Number invited to participate	15	9	0[a]	17[b]	34[c]	0[d]	13	10	98
Number of respondents	13[e]	9	n/a	14	28[f]	n/a	12	9[g]	85
Percentage completing round 1	87	100	n/a	82	82	n/a	92	90	86

n/a, not applicable.
a Geneticists not invited to participate in further rounds.
b Eighteen completed round 1 but one was on maternity leave at the time of round 2.
c Thirty-six completed round 1 but two were on maternity leave at the time of round 2.
d Paediatricians combined with audiologists into group 'audiologists/audiological physicians'.
e One participant with partial scores (29/47 outcomes).
f One participant with partial scores (40/47 outcomes).
g One participant with partial scored (10/47 outcomes).

Attrition bias in round 2

To identify whether or not attrition in round 2 would introduce bias, the scores from round 1 were compared with those completing both rounds ($n = 85$) versus those completing round 1 only ($n = 14$). *Figure 14* shows the distribution of the average score across all 47 outcomes by stakeholder group. The results of those who did not complete round 2 did not represent extreme views suggesting that bias had not been introduced through attrition between round 1 and 2.

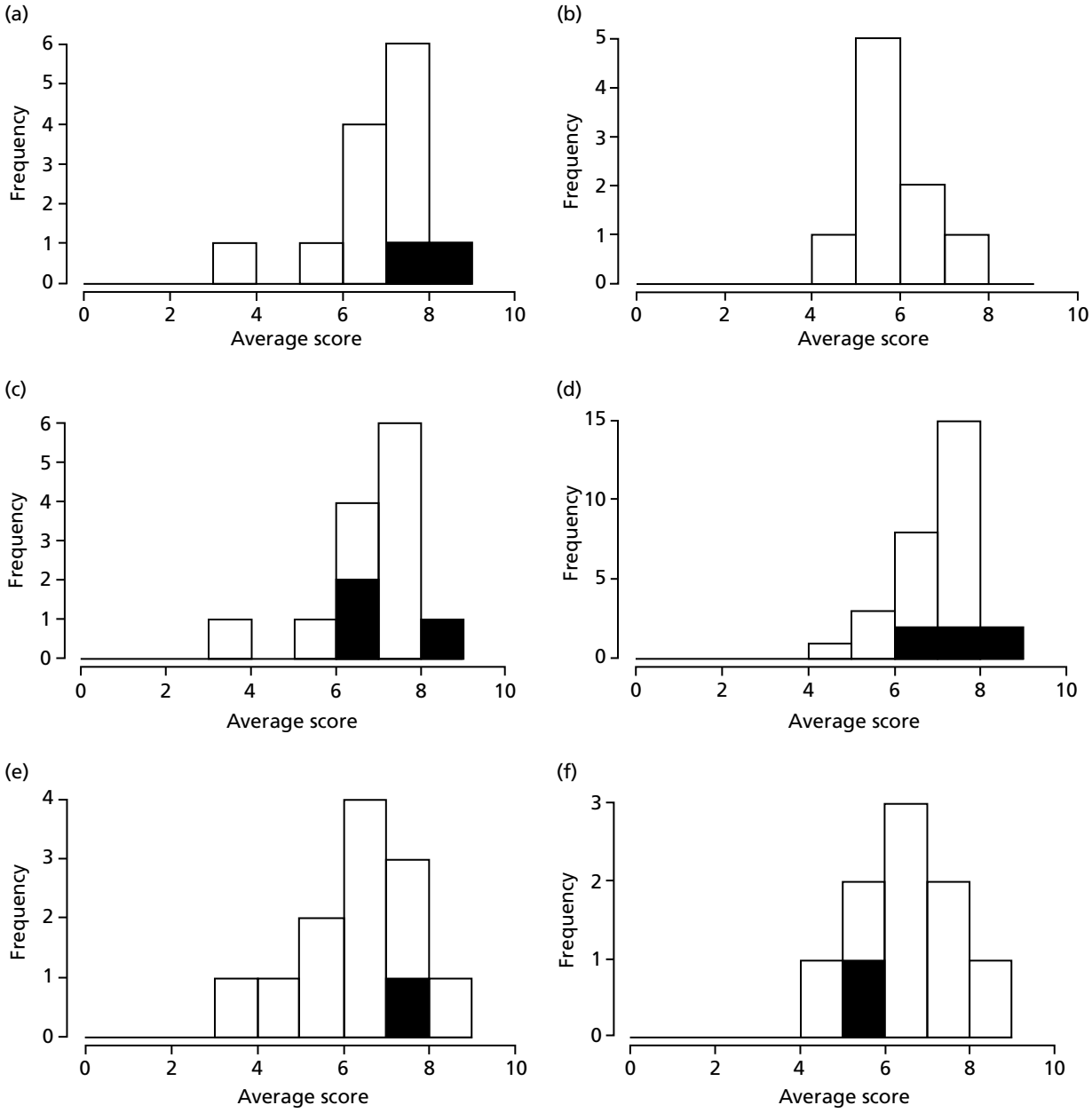


FIGURE 14 Round 1 average scores across all outcomes by stakeholder group. Shaded bars represent those who provided scores in round 1 only, open bars represent those scoring in both rounds 1 and 2. (a) Cleft surgeon; (b) ENT surgeon; (c) specialist care nurse; (d) speech and language therapist; (e) psychologist; and (f) audiologist.

Changes in score between rounds 1 and 2

In round 2 participants were shown the results for their stakeholder group and asked to rescore each outcome. Participants were informed that they could change their score or keep it the same as their score in round 1. Six participants (7%) did not change any scores between rounds 1 and 2, whereas two participants (2%) changed between 80% and 100% of their scores (*Figure 15*).

Consensus matrix

The scores in round 2 were again compared against the definition of consensus to determine which stakeholder groups had reached the definition of 'consensus in' (*Table 36*).

The percentage of outcomes scored as consensus in was again lowest in the ENT surgeon group and highest in the specialist cleft nurse group. However, between rounds the most important outcomes had changed. For example, in round 1 ENT surgeons had reached 'consensus in' for the outcomes 'acute otitis media' (AOM) and 'side effects of treatment', but in round 2 these were no longer included and had been replaced by 'speech intelligibility' and 'speech signs of velopharyngeal insufficiency'. In the audiologist/audiological physicians group the number of outcomes where 'consensus in' had been reached increased from 6 to 19.

As in round 1 five of the six stakeholder groups had reached 'consensus in' for the outcome 'hearing'. However, in round 2 five of the six groups also reached consensus that the outcomes 'OME', 'psychosocial development' and 'speech intelligibility' should be included in the COS.

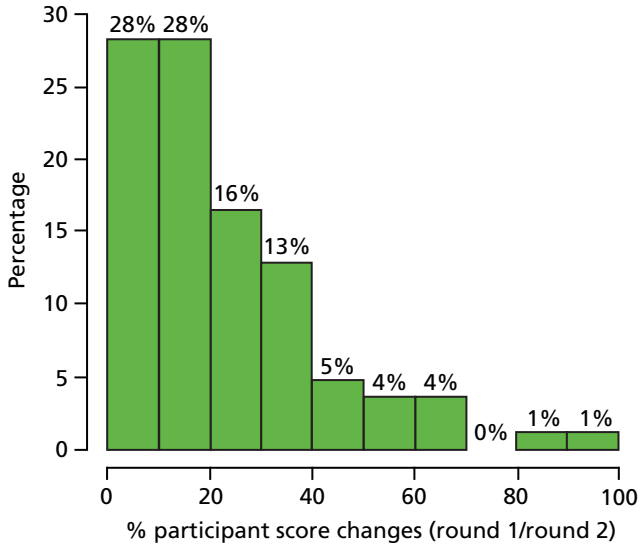


FIGURE 15 Percentage of scores changed between rounds 1 and 2 after viewing the results by stakeholder group.

TABLE 36 Consensus matrix for round 2 of the health professional's Delphi survey

Outcome	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist/ audiological physician
Internalising behaviour			✓		✓	
Externalising behaviour			✓		✓	
Atelectasis	✓		✓			
Cholesteatoma	✓	✓	✓	✓		✓
COM	✓		✓	✓		✓
Persistent tympanic membrane perforation	✓		✓	✓		✓
Persistent tympanic membrane retraction	✓		✓			
Tympanosclerosis	✓		✓			
Academic achievement			✓			
Cognitive development	✓		✓	✓	✓	
Developmental progress	✓		✓	✓	✓	✓
Intelligence						
Literacy			✓	✓		
Phonological memory				✓		
Psychosocial development			✓	✓	✓	
Hearing	✓	✓	✓	✓		✓
Otalgia	✓					
Otorrhoea	✓					✓
Tinnitus	✓					
Vertigo	✓		✓			
Eustachian tube function	✓		✓			
Stapedial reflex	✓		✓			
Nasal obstruction						
Rhinitis						
AOM	✓		✓	✓		
OME	✓	✓	✓	✓		✓
Temporary tympanic membrane perforation			✓			
Consonant production			✓	✓		✓
Consonant production – cleft-related speech patterns	✓			✓		✓
Expressive language skills			✓	✓		
Parent's perspective of speech				✓		
Receptive language skills	✓		✓	✓		✓
Speech development	✓		✓	✓		✓
Speech intelligibility	✓	✓	✓	✓		✓

continued

TABLE 36 Consensus matrix for round 2 of the health professional's Delphi survey (*continued*)

Outcome	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist/ audiological physician
Speech signs of velopharyngeal insufficiency	✓	✓	✓	✓		
Early extrusion or blockage of VTs			✓			✓
Necessity to remove VTs	✓					✓
Requirement for repeated VTs	✓		✓			✓
Child stress			✓		✓	
Parental stress			✓		✓	
Parental satisfaction with treatment		✓	✓		✓	
Side effects of treatment	✓		✓			✓
Upper respiratory tract infection						
Child's satisfaction with treatment			✓	✓	✓	✓
Child's perspective of speech			✓	✓	✓	✓
Psychological well-being			✓	✓	✓	
Listening skills	✓		✓	✓		✓

AOM, acute otitis media; COM, chronic otitis media.
A ✓ indicates where the definition of 'consensus in' was achieved.

Round 3: health professionals

Forty-nine outcomes were scored in round 2. These represented the 47 outcomes scored in round 2 together with the outcome 'hyperacusis' identified from the free-text response to the parent/child survey and 'psychosocial well-being'. An error in the entry of outcomes onto the online system in round 2 meant that 'psychosocial well-being' was listed as 'psychological well-being' which is considered to be a different outcome. Therefore in round 3 this was clarified and participants asked to score 'psychosocial well-being' as well as rescore 'psychological well-being'. Participants were shown their own score in round 2 together with the scores for each of the stakeholder groups including parents and children. A total of 73 responses were received in round 2 (86% of those completing round 1). The breakdown of participants by stakeholder group is shown in *Table 37*. After round 2, four participants left the cleft service and so were no longer eligible to participate in round 3.

Attrition bias in round 3

To identify whether or not attrition in round 3 would introduce bias, the scores from round 1 were compared for those completing both rounds ($n = 73$) with those completing round 1 only ($n = 8$). *Figure 16* shows the distribution of the average score across all 47 outcomes by stakeholder group. The results of those who did not complete round 3 did not represent extreme views suggesting that bias had not been introduced through attrition between rounds 2 and 3.

TABLE 37 Round 3 response rates

Stakeholder group	Cleft surgeon	ENT surgeon	Geneticist	Specialist cleft nurse	Speech and language therapist	Paediatrician	Psychologist	Audiologist/ audiological physician	Total
Number invited to participate	12[a]	8[b]	0[c]	13[d]	27[e]	0[f]	12	9	81
Number of respondents	11	7[g]	n/a	13[h]	24[i]	n/a	11	7	73
Percentage completing round 1	92	88	n/a	100	89	n/a	92	92	90

n/a, not applicable.
a One participant had left the cleft service and retired since completing round 2 (11 of 12 completing round 2 invited).
b One participant had left the cleft service since completing round 2 (8 of 9 completing round 2 invited).
c Geneticists not invited to participate in further rounds.
d One participant had left the cleft service since completing round 2 (13 of 14 completing round 2 invited).
e One participant had left the cleft service since completing round 2 (27 of 28 completing round 2 invited).
f Paediatricians combined with audiologists into group 'audiologists/audiological physicians'.
g One participant with partial scores (4 of 49 outcomes).
h One participant with partial scores (13 of 49 outcomes).
i Two participants with partial scores (6 and 4 of 49 outcomes).

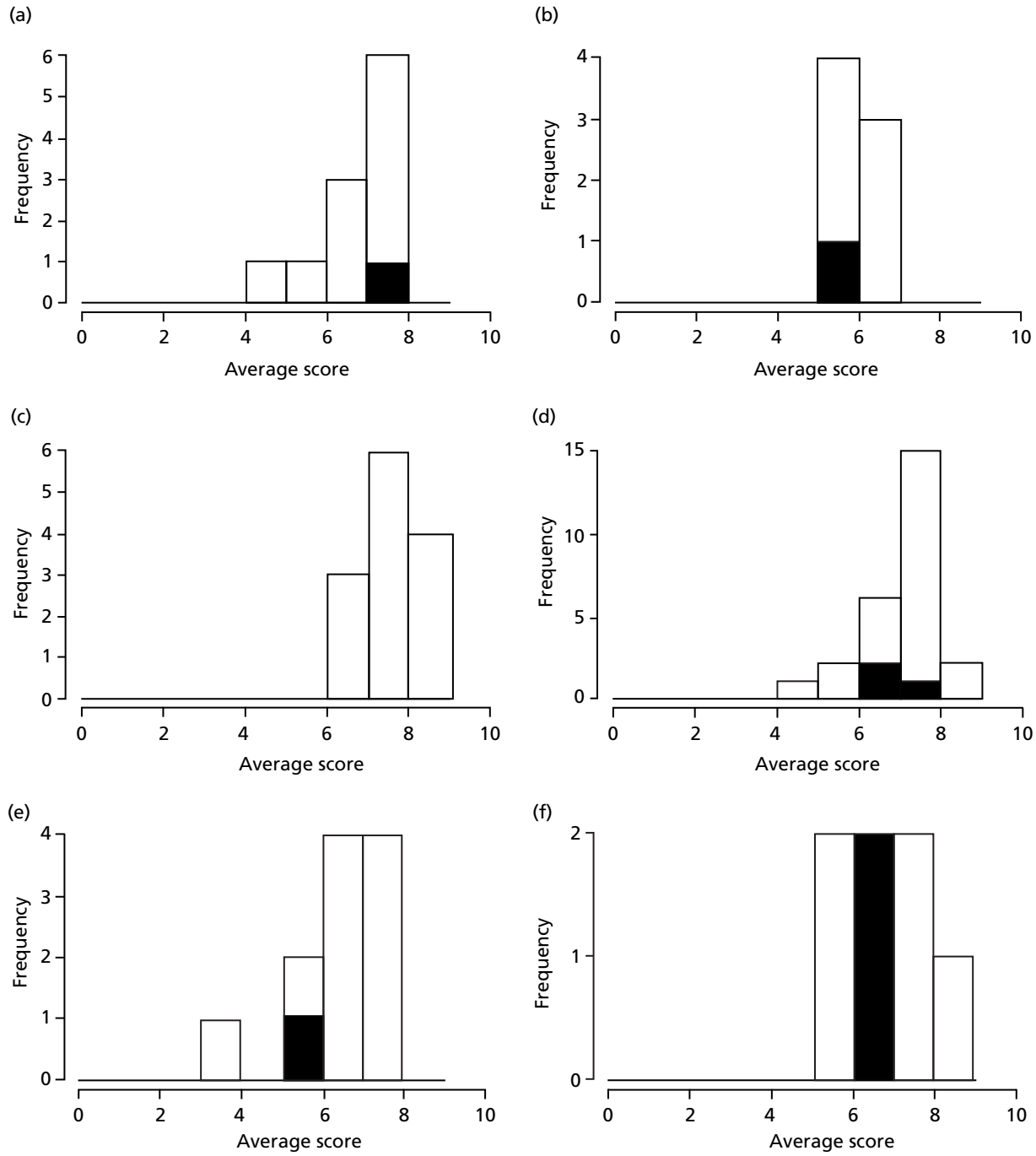


FIGURE 16 Average scores in round 2 across all outcomes by stakeholder group. Shaded bars represent those who provided scores in round 2 only, open bars represent those scoring in both round 2 and 3. (a) Cleft surgeon; (b) ENT surgeon; (c) specialist care nurse; (d) speech and language therapist; (e) psychologist; and (f) audiologist.

Changes in score between rounds 2 and 3

In round 3 participants were shown the results for all stakeholder groups, which also included the results of the parent and child survey, and were asked to rescore each outcome. Participants were informed that they could change their score or keep it the same as their score in round 2. Three participants (4%) did not change any scores between rounds 2 and 3. A larger proportion of participants changed between 20% and 60% of their scores in round 3 (46%) compared with round 2 (38%) (*Figure 17*).

Consensus matrix

The scores in round 3 were again compared against the definition of consensus to determine which stakeholder groups had reached the definition of 'consensus in'.

After round 3 all eight of the stakeholder groups (health professionals plus parents and children) had reached 'consensus in' for the outcome 'hearing'. Seven of the stakeholder groups had reached consensus in for the outcomes 'OME' and 'COM'. Details of the stakeholder groups reaching consensus in for each outcome are shown in *Table 38*.

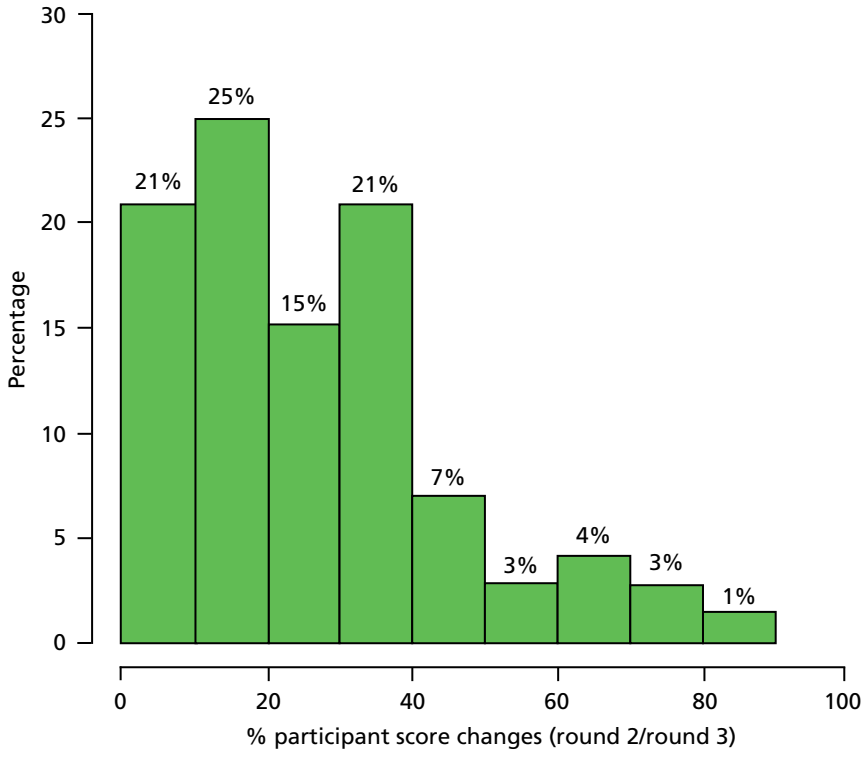


FIGURE 17 Percentage of score changed between rounds 2 and 3 after viewing the results by stakeholder group including the results of the survey of parents and children.

TABLE 38 Consensus matrix for round 3 of the Delphi survey of health professionals

Outcome	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist/ audiological physician
Internalising behaviour			✓		✓	
Externalising behaviour			✓		✓	
Atelectasis	✓		✓			✓
Cholesteatoma	✓	✓	✓	✓		✓
COM	✓	✓	✓	✓		✓
Persistent tympanic membrane perforation	✓		✓			
Persistent tympanic membrane retraction	✓		✓			
Tympanosclerosis			✓			
Academic achievement			✓			
Cognitive development			✓	✓	✓	
Developmental progress			✓	✓	✓	
Intelligence						
Literacy			✓			
Phonological memory				✓		
Psychosocial development			✓	✓	✓	
Hearing	✓	✓	✓	✓	✓	✓
Otalgia	✓		✓			
Otorrhoea	✓	✓				*
Tinnitus			✓			
Vertigo			✓			
Eustachian tube function	✓		✓			
Stapedial reflex			✓			
Nasal obstruction						
Rhinitis						
AOM	✓		✓	✓		✓
OME	✓	✓	✓	✓		✓
Temporary tympanic membrane perforation			✓			
Consonant production	✓		✓	✓		✓
Consonant production – cleft-related speech patterns	✓		✓	✓		
Expressive language skills			✓	✓		
Parent's perspective of speech				✓		
Receptive language skills	✓		✓	✓		✓
Speech development	✓	✓	✓	✓		✓
Speech intelligibility	✓	✓	✓	✓		✓
Speech signs of velopharyngeal insufficiency			✓	✓		
Early extrusion or blockage of VTs			✓			✓

TABLE 38 Consensus matrix for round 3 of the Delphi survey of health professionals (*continued*)

Outcome	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist/ audiological physician
Necessity to remove VTs	✓		✓			✓
Requirement for repeated VTs	✓		✓			✓
Child stress			✓		✓	
Parental stress			✓		✓	
Parental satisfaction with treatment		✓	✓		✓	✓
Side effects of treatment		✓	✓			✓
Upper respiratory tract infection						
Child's satisfaction with treatment			✓	✓	✓	✓
Child's perspective of speech			✓	✓	✓	✓
Psychological well-being			✓	✓	✓	
Listening skills	✓		✓	✓		✓
Psychosocial well-being	✓		✓	✓	✓	
Hyperacusis						

COM, chronic otitis media.
A ✓ indicates where the definition of 'consensus in' was achieved.

Variability in outcomes achieving consensus between rounds

Between rounds there was variability in the number of outcomes achieving consensus. Fewer outcomes achieved consensus in all rounds compared with those achieving consensus in round 3 only (*Table 39*). This was consistent across all stakeholder groups.

In round 3 participants were shown the results of all stakeholder groups, including parents and children with CP. In round 3 more outcomes were considered important, by the individual health professional stakeholder groups, and achieved consensus compared with round 2 (suggesting that the availability of the scores from all stakeholder groups influenced responses). The individual outcomes reaching consensus across all three rounds for each health professional stakeholder group are shown in *Tables 40–45*.

TABLE 39 Variability in outcomes achieving consensus between rounds

Health professional stakeholder group	Number of outcomes reaching consensus in all 3 rounds	Number reaching consensus in round 2 and staying in consensus in round 3	Number only reaching consensus in round 3	Additional outcomes achieving consensus in round 3 compared with round 2
Cleft surgeon	14	18	20	2
ENT surgeon	4	5	9	4
Specialist cleft nurse	32	36	41	5
Speech and language therapist	13	21	22	1
Psychologist	7	11	13	2
Audiologist	6	12	19	7

TABLE 40 Changes in outcomes reaching consensus between rounds for cleft surgeons

Outcome	R1 consensus	R2 consensus	R3 consensus
Atelectasis		✓	✓
Cholesteatoma	✓	✓	✓
COM	✓	✓	✓
Persistent tympanic membrane perforation	✓	✓	✓
Persistent tympanic membrane retraction	✓	✓	✓
Tympanosclerosis	✓	✓	
Cognitive development		✓	
Developmental progress		✓	
Hearing	✓	✓	✓
Otalgia		✓	✓
Otorrhoea	✓	✓	✓
Tinnitus		✓	
Vertigo		✓	
Eustachian tube function	✓	✓	✓
Stapedial reflex		✓	
AOM	✓	✓	✓
OME	✓	✓	✓
Consonant production			✓
Temporary tympanic membrane perforation	✓		
Consonant production – cleft-related speech patterns	✓	✓	✓
Receptive language skills		✓	✓
Speech development	✓	✓	✓
Speech intelligibility	✓	✓	✓
Speech signs of velopharyngeal insufficiency		✓	
Necessity to remove VTs	✓	✓	✓
Requirement for repeated VTs	✓	✓	✓
Side effects of treatment		✓	
Listening skills		✓	✓
Psychosocial well-being			✓

COM, chronic otitis media.

TABLE 41 Changes in outcomes reaching consensus between rounds for ENT surgeons

Outcome	R1 consensus	R2 consensus	R3 consensus
Cholesteatoma	✓	✓	✓
COM			✓
Hearing	✓	✓	✓
OM	✓		
Otorrhoea			✓
OME	✓	✓	✓
Speech development			✓
Speech intelligibility		✓	✓
Speech signs of velopharyngeal insufficiency		✓	
Parental satisfaction with treatment	✓	✓	✓
Side effects of treatment	✓		✓

COM, chronic otitis media; OM, otitis media.

TABLE 42 Changes in outcomes reaching consensus between rounds for specialist cleft nurses

Outcome	R1 consensus	R2 consensus	R3 consensus
Internalising behaviour	✓	✓	✓
Externalising behaviour	✓	✓	✓
Atelectasis		✓	✓
Cholesteatoma		✓	✓
COM	✓	✓	✓
Persistent tympanic membrane perforation	✓	✓	✓
Persistent tympanic membrane retraction	✓	✓	✓
Tympanosclerosis	✓	✓	✓
Academic achievement	✓	✓	✓
Cognitive development	✓	✓	✓
Developmental progress	✓	✓	✓
Literacy	✓	✓	✓
Psychosocial development	✓	✓	✓
Hearing	✓	✓	✓
Otalgia			✓
Tinnitus			✓
Vertigo	✓	✓	✓
Eustachian tube function	✓	✓	✓
Stapedial reflex	✓	✓	✓

continued

TABLE 42 Changes in outcomes reaching consensus between rounds for specialist cleft nurses (*continued*)

Outcome	R1 consensus	R2 consensus	R3 consensus
AOM	✓	✓	✓
OME	✓	✓	✓
Temporary tympanic membrane perforation	✓	✓	✓
Consonant production	✓	✓	✓
Consonant production – cleft-related speech patterns			✓
Expressive language skills	✓	✓	✓
Parent's perspective of speech	✓		
Receptive language skills	✓	✓	✓
Speech development	✓	✓	✓
Speech intelligibility	✓	✓	✓
Speech signs of velopharyngeal insufficiency	✓	✓	✓
Early extrusion or blockage of VTs	✓	✓	✓
Necessity to remove VTs			✓
Requirement for repeated VTs	✓	✓	✓
Child stress	✓	✓	✓
Parental stress	✓	✓	✓
Parental satisfaction with treatment	✓	✓	✓
Side effects of treatment	✓	✓	✓
Child's satisfaction with treatment	✓	✓	✓
Child's perspective of speech	✓	✓	✓
Psychological well-being		✓	✓
Listening skills		✓	✓
Psychosocial well-being			✓

COM, chronic otitis media.

TABLE 43 Changes in outcomes reaching consensus between rounds for speech and language therapists

Outcome	R1 consensus	R2 consensus	R3 consensus
Cholesteatoma		✓	✓
COM	✓	✓	✓
Persistent tympanic membrane perforation		✓	
Cognitive development		✓	✓
Developmental progress	✓	✓	✓
Literacy		✓	
Phonological memory		✓	✓
Psychosocial development		✓	✓
Hearing	✓	✓	✓
AOM		✓	✓
OME	✓	✓	✓
Consonant production	✓	✓	✓
Consonant production – cleft-related speech patterns	✓	✓	✓
Expressive language skills	✓	✓	✓
Parent's perspective of speech	✓	✓	✓
Receptive language skills	✓	✓	✓
Speech development	✓	✓	✓
Speech intelligibility	✓	✓	✓
Speech signs of velopharyngeal insufficiency	✓	✓	✓
Child's satisfaction with treatment		✓	✓
Child's perspective of speech	✓	✓	✓
Psychological well-being		✓	✓
Listening skills		✓	✓
Psychosocial well-being			✓

COM, chronic otitis media.

TABLE 44 Changes in outcomes reaching consensus between rounds for clinical psychologists

Outcome	R1 consensus	R2 consensus	R3 consensus
Internalising behaviour	✓	✓	✓
Externalising behaviour	✓	✓	✓
Cognitive development		✓	✓
Developmental progress	✓	✓	✓
Psychosocial development		✓	✓
Hearing			✓
Child stress	✓	✓	✓
Parental stress	✓	✓	✓
Parental satisfaction with treatment		✓	✓
Child's satisfaction with treatment	✓	✓	✓
Child's perspective of speech	✓	✓	✓
Psychological well-being		✓	✓
Psychosocial well-being			✓

TABLE 45 Changes in outcomes reaching consensus between rounds for audiologists

Outcome	R1 consensus	R2 consensus	R3 consensus
Atelectasis			✓
Cholesteatoma	✓	✓	✓
COM		✓	✓
Persistent tympanic membrane perforation		✓	
Developmental progress		✓	
Hearing	✓	✓	✓
Otorrhoea		✓	✓
AOM			✓
OME		✓	✓
Consonant production		✓	✓
Consonant production – cleft-related speech patterns		✓	
Receptive language skills		✓	✓
Speech development		✓	✓
Speech intelligibility		✓	✓
Early extrusion or blockage of VTs	✓	✓	✓
Necessity to remove VTs	✓	✓	✓
Requirement for repeated VTs	✓	✓	✓
Parental satisfaction with treatment			✓
Side effects of treatment	✓	✓	✓
Child's satisfaction with treatment		✓	✓
Child's perspective of speech		✓	✓
Listening skills		✓	✓

COM, chronic otitis media.

Consensus meeting

A meeting was held on the 6 March 2014 to review the results of the mOMEnt study COS development. The meeting was attended by health professionals, parents, parent representatives and observers. Each outcome from the list scored by parents and health professionals, in the online survey and Delphi survey, respectively, was reviewed at the meeting with some discussed further and rescored for importance.

The meeting resulted in a preliminary COS together with a set of outcomes that would need further discussion with parents. In addition, a set of outcomes related to speech that possibly represent 'how' a particular outcome would be measured were discussed with the SAG.

Pre meeting

An invitation to attend the COS consensus meeting was sent to:

- health professionals who had completed all rounds of the online Delphi survey and expressed an interest in attending future meetings
- all parents who had completed an online survey, expressed an interest and provided contact details to be informed about future meetings
- parents who had taken part in a qualitative interview whose contact details were still valid
- CLAPA members in the north west based on CLAPA mailing list.

All those who confirmed attendance received an e-mail with information on what they should expect at the meeting and also three documents related to the meeting content: the Core Outcome Measures in Effectiveness Trials (COMET) plain language summary, the meeting agenda and a meeting overview document (see *Appendix 7*).

A separate session was scheduled immediately before the main consensus meeting for 30 minutes to allow HB and ST to meet with parents. This meeting allowed parents to meet one another and for any questions to be answered about the structure of the day, expectations and for additional information to be given on COS development.

Twenty-five participants attended the consensus meeting of whom 14 were eligible to vote (*Table 46*). All stakeholder groups with the exception of clinical psychologists were represented (*Table 47*). Two parents were in attendance (another two were due to attend, but shortly before the meeting notified NH that they were unable to do so due to child illness for one and jury duty for the other). On the day, three health professionals (one audiologist, one cleft surgeon and one speech and language therapist) were unable to attend due to illness or the need to cover a colleague's clinic.

Meeting agenda

The meeting was structured according to the moMEnt consensus meeting agenda version 1.0 (see *Appendix 7*) with the exception of the meeting summary which, due to the time taken for discussion of each outcome, was sent to participants after the meeting.

The day began with an informal session where participants were asked to sit next to someone they did not know and to find out a little bit about them including one of their favourite things or places. This encouraged good interaction and all participants were energetic and engaged. PW stressed at the beginning of the meeting that all should feel free to ask questions and that no question was trivial.

After the introduction IAB gave an overview of the mOMEnt study and of OME. This included some technical terms.

PW then went on to describe what was meant by an outcome and what a COS represents. After that the plan for the day was summarised reiterating that everyone should feel free to ask questions and to share their opinions or experiences.

TABLE 46 Participants of the consensus meeting

Initials	Meeting role	Stakeholder group	Membership
PW	Meeting facilitator	n/a	SMG
IAB	Presenter – introduction to mOMEnt	ENT surgeon[a]	SMG
NH	Presenter – methods	n/a	SMG
ST	Presenter – qualitative results	n/a	SMG
PC	Presenter – qualitative results	n/a	SMG
HB	COMET PPI co-ordinator	n/a	COMET
AHB	Participant	Speech and language therapist[a]	SAG
RC	Participant	Audiologist/audiological physician[a]	SAG
PH	Participant	Cleft surgeon[a]	SAG
AH	Participant	Audiologist/audiological physician[a]	Health professionals Delphi
SD	Participant	ENT surgeon[a]	Health professionals Delphi
NHu	Participant	Cleft nurse specialist[a]	Health professionals Delphi
FJ	Participant	Speech and language therapist[a]	Health professionals Delphi
TB	Participant	Cleft nurse specialist[a]	Health professionals Delphi
CH	Participant	Speech and language therapist[a]	Health professionals Delphi
AC	Participant	Speech and language therapist[a]	Health professionals Delphi
JH	Participant	Parent[a]	Parent online survey
LH	Participant	Parent[a]	Parent interviews
RP	Participant	Chief executive of CLAPA[a]	CLAPA
CB	Meeting organiser	n/a	SMG
KOB	Observer	n/a	SMG
BS	Observer	n/a	SMG
KW	Observer	n/a	University of Liverpool
AW	Observer	n/a	University of Liverpool
BE	Observer	n/a	The Healing Foundation
JT	Observer	n/a	The Healing Foundation

n/a, not applicable; PPI, patient and public involvement.
a Voting member.

TABLE 47 Stakeholder representation at consensus meeting

Stakeholder group	Number of voting members attending consensus meeting	Percentage representation at consensus meeting
ENT surgeon	2	14
Cleft nurse specialist	2	14
Speech and language therapist	4	29
Audiologist/audiological physician	2	14
Cleft surgeon	1	7
Clinical psychologist	0	0
Parent/parent representative	3	21

NH presented the methods used in mOMEnt for both health professionals and parents and children. TB asked how we had anticipated people would respond when they saw other health professionals' scores, as when she received them in round 2 of the survey she was unclear on how to react and spent time thinking about this. PW responded that this was exactly what we wanted participants to do; we wanted them to think about their own score and how it fitted with the scores of others. Those at the meeting were reminded that health professionals in the Delphi were advised that they could change their score or keep it the same.

CH said that in round 3 it was helpful to have the terminology used for parents to help understand each outcome.

JH asked how the health professionals were identified and why there were smaller numbers of audiologists and ENT surgeons. NH responded to say that clinical leads at each of the UK cleft centres were contacted and asked to provide the names, clinical roles and contact details of their teams. Not all cleft centres in the UK have a dedicated ENT surgeon or audiologist which is reflected in the numbers invited and has also been supported by responses to the clinician survey. There was general agreement from health professionals in the room to this response.

NH went on to describe the survey for parents and children including the number of parents and children who had completed. NH also presented the definition of consensus that was agreed at the start of this study and published in the trial protocol. NH concluded by summarising that the meeting would aim to bring the information from all of the sources together but that we would really like to do more work to get further input from parents and children.

ST and PC then presented the results of the qualitative interviews. PC gave a clear and lay explanation of what qualitative research is and how the data was analysed. ST summarised what children and parents said about physical and psychological outcomes and how the research team perceived they were interconnected.

PW presented the results of the Delphi survey, in terms of the numbers responding, attrition bias and the effect of each round on changes to individual's scores. The responses by stakeholder group are provided in *Appendix 8*.

The summary of round 3 results based on which of the eight stakeholder groups (cleft surgeons, ENT surgeons, specialist cleft nurses, speech and language therapists, psychologists, audiologists, parents and children) had reached the definition of consensus for each outcome were tabled (*Table 48*).

After lunch PW continued to present the results. There was an initial test of the voting buttons. Participants who were eligible to vote are detailed in *Table 46*.

TABLE 48 Summary of all groups reaching consensus for individual outcomes

Outcome	Round 3 and survey of parents and children with CP							
	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist	Parent	Child
Internalising behaviour			✓		✓			
Externalising behaviour			✓		✓			
Atelectasis	✓		✓			✓	✓	✓
Cholesteatoma	✓	✓	✓	✓		✓		
COM	✓	✓	✓	✓		✓	✓	✓
Persistent tympanic membrane perforation	✓		✓					✓
Persistent tympanic membrane retraction	✓		✓				✓	✓
Tympanosclerosis			✓				✓	✓
Academic achievement			✓				✓	
Cognitive development			✓	✓	✓		✓	
Developmental progress			✓	✓	✓		✓	
Intelligence							✓	
Literacy			✓				✓	
Phonological memory				✓			✓	
Psychosocial development			✓	✓	✓		✓	✓
Hearing	✓	✓	✓	✓	✓	✓	✓	✓
Otalgia	✓		✓				✓	
Otorrhoea	✓	✓				✓		
Tinnitus			✓				✓	
Vertigo			✓				✓	
Eustachian tube function	✓		✓				✓	
Stapedial reflex			✓				✓	
Nasal obstruction								
Rhinitis								
AOM	✓		✓	✓		✓	✓	
OME	✓	✓	✓	✓		✓	✓	✓
Temporary tympanic membrane perforation			✓					
Consonant production	✓		✓	✓		✓	✓	

TABLE 48 Summary of all groups reaching consensus for individual outcomes (*continued*)

Outcome	Round 3 and survey of parents and children with CP							
	Cleft surgeon	ENT surgeon	Specialist cleft nurse	Speech and language therapist	Psychologist	Audiologist	Parent	Child
Consonant production – cleft-related speech patterns	✓		✓	✓			✓	
Expressive language skills			✓	✓			✓	
Parent's perspective of speech				✓			✓	
Receptive language skills	✓		✓	✓		✓	✓	✓
Speech development	✓	✓	✓	✓		✓	✓	
Speech intelligibility	✓	✓	✓	✓		✓		
Speech signs of velopharyngeal insufficiency			✓	✓			✓	
Early extrusion or blockage of VTs			✓			✓	✓	
Necessity to remove VTs	✓		✓			✓	✓	✓
Requirement for repeated VTs	✓		✓			✓	✓	✓
Child stress			✓		✓		✓	
Parental stress			✓		✓			
Parental satisfaction with treatment		✓	✓		✓	✓	✓	
Side effects of treatment		✓	✓			✓	✓	
Upper respiratory tract infection							✓	
Child's satisfaction with treatment			✓	✓	✓	✓	✓	
Child's perspective of speech			✓	✓	✓	✓	✓	
Psychological well-being			✓	✓	✓			
Listening skills	✓		✓	✓		✓		
Psychosocial well-being	✓		✓	✓	✓			
Hyperacusis								

COM, chronic otitis media.

Results of voting and review of outcomes

Each outcome was considered in turn. Those presented first represented those on which the most stakeholder groups had reached consensus. For each outcome, participants decided if they wanted to discuss it, to simply revote or to not discuss it further. A summary of the result for each outcome is given in *Table 49*. Detailed notes of the discussion and a breakdown of scores are given in *Appendix 9*.

Each outcome has been categorised based on the following:

1. discussed and voted
2. discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured
3. discussed and agreed that further discussion with parents is needed
4. agreed not to discuss further or vote – not in the COS.

TABLE 49 Summary of outcomes discussed

Outcome	Number of the eight stakeholder groups achieving consensus prior to meeting	Percentage of meeting participants scoring 7–9	Percentage of meeting participants scoring 1–3	Category of meeting conclusion	Description of category of meeting conclusion
Hearing	8	100	0	1	Discussed and voted
COM	7	100	0	1	Discussed and voted
OME	7	93	7	1	Discussed and voted
Speech intelligibility	6	85	0	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Also agreed by SAG post meeting to include as 'how' of speech development
Receptive language skills	6	100	0	1	Discussed and voted
Speech development	6	93	7	1	Discussed and voted
Atelectasis	5	46	9	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Atelectasis to be combined with 'COM'
Cholesteatoma	5	84	0	3	Discussed and agreed that further discussion with parents is needed
Psychosocial development	5	71	7	1	Discussed and voted
AOM	5	78	7	1	Discussed and voted

TABLE 49 Summary of outcomes discussed (*continued*)

Outcome	Number of the eight stakeholder groups achieving consensus prior to meeting	Percentage of meeting participants scoring 7–9	Percentage of meeting participants scoring 1–3	Category of meeting conclusion	Description of category of meeting conclusion
Consonant production	5	76	8	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Also agreed by SAG post meeting to include as 'how' of speech development
Necessity to remove VTs	5	0	67	4	Agreed not to discuss further or vote – not in the COS as this relates to a specific treatment
Requirement for repeated VTs	5	44	27	4	Agreed not to discuss further or vote – not in the COS as this relates to a specific treatment
Parental satisfaction with treatment	5	69	8	1	Discussed and voted
Child's satisfaction with treatment	5	61	0	1	
Child's perspective of speech	5	69	0	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Also agreed by SAG post meeting to include as 'how' of speech development
Persistent tympanic membrane retraction	4	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Persistent tympanic membrane retraction to be combined with 'COM'
Cognitive development	4	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured To be combined with 'how well you are doing at school'

continued

TABLE 49 Summary of outcomes discussed (*continued*)

Outcome	Number of the eight stakeholder groups achieving consensus prior to meeting	Percentage of meeting participants scoring 7–9	Percentage of meeting participants scoring 1–3	Category of meeting conclusion	Description of category of meeting conclusion
Developmental progress	4	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Developmental progress to be combined with 'how well you are doing at school'
Consonant production – cleft-related speech patterns	4	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Consonant production cleft-related speech patterns to be combined with 'consonant production' Also agreed by SAG post meeting to include as 'how' of speech development
Side effects of treatment	4	100	0	1	Discussed and voted
Listening skills[a]	4	84	0	1	Discussed and voted
Psychosocial well-being[a]	4	69	0	1	Discussed and voted
Tympanosclerosis	3	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Tympanosclerosis to be combined with 'COM'
Persistent tympanic membrane perforation	3	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Persistent tympanic membrane perforation to be combined with 'COM'
Otalgia	3	67	24	1	Discussed and voted
Otorrhoea	3	50	8	1	Discussed and voted
Eustachian tube function	3	27	0	3	Discussed and agreed that further discussion with parents is needed

TABLE 49 Summary of outcomes discussed (*continued*)

Outcome	Number of the eight stakeholder groups achieving consensus prior to meeting	Percentage of meeting participants scoring 7–9	Percentage of meeting participants scoring 1–3	Category of meeting conclusion	Description of category of meeting conclusion
Expressive language skills	3	n/a	n/a	3	At the consensus meeting this was considered as part of the 'how' speech development is measured and it was agreed not to vote. However, post-meeting discussion with the SAG identified that the grouping of this outcome for parents might not have been appropriate and so this outcome should be discussed further
Speech signs of velopharyngeal insufficiency	3	n/a	n/a	3	Wording of lay description should be revisited and discussed with parents
Early extrusion or blockage of VTs	3	n/a	n/a	4	Agreed not to discuss further or vote – not in the COS as this relates to a specific treatment
Child stress	3	51	26	1	Discussed and voted
Psychological well-being[a]	3	n/a	n/a	3	Discussed and agreed that further discussion with parents is needed
Internalising behaviour	2	n/a	n/a	4	Agreed not to discuss further or vote – not in the COS
Externalising behaviour	2	n/a	n/a	4	Agreed not to discuss further or vote – not in the COS
Academic achievement	2	66	8	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Academic achievement to be combined with 'how well you are doing at school'
Literacy	2	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Literacy to be combined with 'how well you are doing at school'

continued

TABLE 49 Summary of outcomes discussed (*continued*)

Outcome	Number of the eight stakeholder groups achieving consensus prior to meeting	Percentage of meeting participants scoring 7–9	Percentage of meeting participants scoring 1–3	Category of meeting conclusion	Description of category of meeting conclusion
Phonological memory	2	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured To be combined with 'how well you are doing at school'
Tinnitus	2	25	50	3	Discussed and agreed that further discussion with parents is needed
Vertigo	2	67	0	1	Discussed and voted
Stapedial reflex	2	0	50	1	Discussed and voted
Parent's perspective of speech	2	n/a	n/a	2	Discussed and agreed to be considered as part of the 'how' an outcome is measured relating to 'speech development'. Also agreed by SAG post meeting to include as 'how' of speech development
Parental stress	2	43	14	1	Discussed and voted
Intelligence	1	n/a	n/a	2	Discussed and agreed to combine with another outcome and to be considered as part of the 'how' an outcome is measured Intelligence to be combined with 'how well you are doing at school'
Temporary tympanic membrane perforation	1	n/a	n/a	4	Agreed not to discuss further or vote – not in the COS
Upper respiratory tract infection	1	0	43	3	Discussed and agreed that further discussion with parents is needed
Nasal obstruction	0	n/a	n/a	4	Agreed not to discuss further or vote – not in the COS
Rhinitis	0	n/a	n/a	4	Agreed not to discuss further or vote – not in the COS
Hyperacusis[a]	0	n/a	n/a	3	Discussed and agreed that further discussion with parents is needed

COM, chronic otitis media; n/a, not applicable.
a Not scored by parents and children therefore total number of stakeholder groups that could reach consensus = 7.

The results of the health professionals Delphi, the parent and child Delphi and discussion at the consensus meeting contributed to a preliminary COS (*Table 50*). This was agreed at a follow-up meeting with the SAG.

The consensus meeting also identified outcomes where ambiguity may have been introduced by the wording used and which all agreed would benefit from further discussion with parents. After a post-meeting discussion with the SAG about the outcomes which represented the 'how' of speech development, a further outcome, 'expressive language', was identified as needing further exploration with parents as the grouping with other outcomes for the parent survey might have been misleading. The outcomes requiring further discussion with parents due to potential issues around the wording used to describe the outcome are described in *Table 51*.

For some outcomes which were seen less frequently among children with CP and OME concerns were raised that the sample of respondents and those present at the meeting may not have had experience of this outcome which, in turn, might have affected its relative importance. All agreed that these outcomes, described in *Table 52*, should be discussed further with a larger group of parent.

Three of the outcomes discussed at the meeting were scored as 'consensus in' by parents and also scored highly at the meeting yet did not meet the definition of consensus in. In addition, participants of the meeting felt that one outcome 'psychological well-being' needed development of a description that could then be explored further with parents as this was not included in the parent survey. The four outcomes that will be discussed further with parents are detailed in *Table 53*.

Meeting feedback

The results of the meeting were summarised in a report which was circulated to all meeting participants. This report described outcomes that had been identified for inclusion in the COS together with outcomes that need further discussion. The resulting preliminary COS was confirmed by the SAG. The outcomes detailed in *Tables 51–53* needed to be discussed further with parents to determine inclusion in a COS representing outcomes that should be measured in all future studies of OME in children with CP. If consensus was reached, the outcome was added to the COS.

The engagement of parents in the next stages of the mOMEnt study will be essential and the study team will work with CLAPA to identify how best to engage their members.

TABLE 50 Outcomes agreed for inclusion in the preliminary COS

Outcome	Number of stakeholder groups scoring as 'consensus in'	Percentage scoring 7–9 at meeting	Percentage scoring 1–3 at meeting
Hearing	8	100	0
COM	7	100	0
OME	7	93	7
Receptive language skills	6	100	0
Speech development	6	93	7
Psychosocial development	5	71	7
AOM	5	78	7
Side effects of treatment	4	100	0
Listening skills	4	84	0

COM, chronic otitis media.

TABLE 51 Outcomes requiring further discussion with parents due to ambiguity of wording

Outcome	Number of stakeholder groups scoring as 'consensus in'	Percentage scoring 7–9 at meeting	Percentage scoring 1–3 at meeting	Notes
Speech signs of velopharyngeal insufficiency	3	n/a	n/a	Further discussion with parents and further consideration of wording used to describe the outcome needed
Upper respiratory tract infection	1	0	43	Further discussion with parents and further consideration of wording used to describe the outcome needed
Eustachian tube function	3	27	0	Further discussion with parents and further consideration of wording used to describe the outcome needed

n/a, not applicable.

TABLE 52 Outcomes requiring further discussion with parents due to potentially only a small number of parents who completed the survey having had experience of the outcome

Outcome	Number of stakeholder groups scoring as 'consensus in'	Percentage scoring 7–9 at meeting	Percentage scoring 1–3 at meeting	Notes
Cholesteatoma	5	84	0	Further discussion with parents
Tinnitus	2	25	50	Further discussion with parents
Hyperacusis	0	n/a	n/a	Further discussion with parents

n/a, not applicable.

TABLE 53 Outcomes requiring further discussion with parents due to under-representation of parents and/or relevant health professionals

Outcome	Number of stakeholder groups scoring as 'consensus in'	Percentage scoring 7–9 at meeting	Percentage scoring 1–3 at meeting	Notes
Psychological well-being	3	n/a	n/a	Further discussion with parents needed with input from clinical psychologists. Clinical psychology was not represented at the consensus meeting to provide support to the interpretation of this outcome
Academic achievement grouped into 'how well your child is doing at school'	2	66	8	Parents in the online survey and three health professional groups in Delphi reached consensus on outcomes included in parent outcome 'how well your child is doing at school'. No consensus at face-to-face meeting and so this will be discussed further with parents
Otalgia	3	67	24	Parents in the online survey and two health professional groups in Delphi reached consensus. No consensus at face-to-face meeting and so this will be discussed further with parents
Child stress	3	51	26	Parents in the online survey and two health professional groups in Delphi reached consensus. No consensus at face-to-face meeting and so this will be discussed further with parents

n/a, not applicable.

Discussion

There is currently no published COS for effectiveness trials of interventions for OME in children with CP, and indeed the outcomes measured in previous studies are variable. The development of a COS in this clinical area aims to improve the interpretation and comparison of future studies, and reduce the risk of ORB and heterogeneity across studies.

The preliminary COS includes nine outcomes related to the management of OME in children with CP (see *Table 50*). The next steps will involve consideration of how each of these outcomes should be defined and how each outcome should be measured. The definition of each outcome will need to take into consideration the meaning of terms to both health professionals and parents as each may have a different understanding of the outcome and potential definitions. Each outcome will also be assessed for potential outcome measurement instruments, whether or not a validated tool already exists and what methods have been used to measure this outcome in previous studies as described in the systematic review.

For the outcomes included in the preliminary COS this will include:

- Consideration of methods of assessing hearing that might be influenced by the intervention (e.g. differing methods depending on VT or HA use).
- Agreeing a definition of COM and methods of measurement.
- Determining which aspects of speech development should be measured and identifying if methods of measurement are already available.
- Reviewing methods for assessment of receptive language, psychosocial development, AOM and listening skills.
- Establishing the most appropriate way to measure side effects of treatment. The question asked in the Delphi survey of health professionals and the survey of parents and children did not specify outcomes in relation to surgical management of OME and so on further reflection, and discussion at the consensus meeting, outcomes related to VT tubes were excluded from the COS in the same way as outcomes specific to HAs. The outcome 'side effects of treatment' included in the COS may be dependent on the interventions/treatments that are being compared. There should also be consideration of potential crossover of AOM as both an individual outcome and a potential side effect of VT insertion.

Guidelines on the selection on outcome measurement instruments to be included in a COS developed by the Core Outcome Measurement Instrument Selection project will be consulted when available. Furthermore, the UK Cleft Audit means that for some outcomes there are potentially methods of measurement that have already been agreed by health professionals providing cleft care in the UK. For example, measures of psychosocial outcomes are included in the audit process at 5, 10, 15 and 20 years. These include both generic measures (to facilitate comparisons with population norms) and questions tailored to those affected by cleft. Speech is also measured using a validated tool which has been tested for its reliability, validity and applicability.[116] Additionally, outcome measures which include multiple domains related to OME, such as the OM8-30 and OMQ-14, warrant further investigation as a method of assessing outcomes.

Internationally, the need for a harmonised approach to outcome assessment in the general management of CP has been identified with recommendations made for standardised data sets.[117,118]

The mOMEnt study has involved multiple key stakeholder groups to ensure that a COS is suitable and well accepted in future research. However, although a preliminary COS has been developed, further engagement is needed with parents to ensure that all outcomes relevant to this group have been adequately considered and that the wording of each outcome was interpreted as the study team had intended. The number of parents attending the consensus meeting resulted in low representation of parents particularly in comparisons with the number of health professionals present (of the voting

participants two parents and 12 health professionals). The number of health professionals in attendance was also lower than expected based on the response rate to the health professional Delphi, with clinical psychologists not being represented at all. In the mOMEnt study the consensus meeting was held for both health professionals and parents to allow integration of views.

Previous studies included in the systematic review of the literature identified papers from a range of countries. The mOMEnt COS has been developed with input from health professionals, parents and children with CP who are based in the UK, but to promote good uptake of the COS into future studies international consensus is needed. Cleft organisations exist in both Europe (the European Cleft Organisation) and the USA (American Cleft Palate-Craniofacial Association), and may represent an opportunity to engage international health professionals through their membership.

Although OME is prevalent in children with CP, affecting around 75%, it is also a common childhood condition for children without cleft, with estimates that approximately 19% of non-cleft children are affected by the condition.[2] The preliminary COS described in the current study includes outcomes that have been identified from previous studies in both cleft and non-cleft populations suggesting that they may also be of relevance to studies of OME in children without CP. Future work will involve contacting a wider group, such as the British Academy of Audiology, so that the relevance of the COS to non-cleft children can be assessed.

Lessons learnt

There are aspects of the mOMEnt study which have identified lessons learnt or future considerations for COS development.

The mOMEnt study aimed to bring together the views of health professionals and parents in a final face-to-face meeting. However, the number of participants in each stakeholder group who were able to attend was lower than expected based on the numbers invited. Health professionals and parents may each have different preferences for the timing of meeting attendance or specific needs, such as child care, which should be taken into consideration for future studies. In the present study both child care and travel costs were reimbursed and travel booked on behalf of participants, but child care was organised by participants themselves which might be challenging if parents do not regularly use child care services. Further discussion with parents will explore the optimum day/timing of a similar type of meeting to inform future COS development of relevance to children with CP.

The low response rate of parents completing the online survey might reflect the transient nature of OME or that this is in fact a lower priority compared with the other needs of a child with CP only, unless perhaps their child is experiencing pain/discomfort or noted issues with hearing. The online system used was not optimised for use with a smartphone or tablet which may too have contributed to the small numbers responding, particularly in the 11–16 years age group where it is estimated that nearly half of UK teenagers own a smartphone.[119] The health professional Delphi survey benefited from SAG input into the design of the online system. For the survey of parents and children with CP all 36 outcomes were shown on one page which might have been potentially off-putting, warranting a future approach which involves system testing with parents and children with CP to explore accessibility and preferences.

The COS development for the mOMEnt study has involved multiple stakeholders with different clinical backgrounds. For health professionals this means that the role each stakeholder group plays in the management of OME and the knowledge of the condition across stakeholder groups is variable. At the consensus meeting a health professional noted that the parent/child descriptors of each outcome were helpful, illustrating the need to use plain language to ensure that the process of COS development is accessible to all. The varying roles of stakeholders may have also influenced the level of engagement with a COS for OME in children with CP. This could, in part, be alleviated by carefully considering the initial invitation, ensuring that the e-mail subject is relevant to all and giving consideration to providing a list of outcomes to be scored with the initial invite so that assumptions about relevance are not made.

Substantially longer was taken to respond to the online survey than had been originally anticipated and multiple reminder e-mails and telephone calls were needed to improve the response rate. Clinical workload is also likely to have contributed to the time taken to respond.

The impact of the COS cannot be assessed in the present study but details will be included in the COMET database so that the COS is readily accessible to future studies of OME in children with CP allowing a future comparison of uptake.

Chapter 5 Economic analysis

Introduction

A key objective of the mOMEnt feasibility study was to perform a decision-analytic model-based economic evaluation to assess the potential impact of the surgical insertion of VTs for the management of persistent bilateral OME in children with CP. The primary aim of the economic analysis was to provide information on the potential key drivers of cost-effectiveness given the current evidence base for each of the potential strategies for managing children with CP and OME. The objectives of the economic analysis involved:

i. conducting a systematic review and critical appraisal of published economic models that aimed to evaluate various treatment options for the management of persistent bilateral OME in children with CP
ii. structuring and populating a de novo economic model to estimate the incremental costs and quality-adjusted life-years (QALYs) of four potential strategies for managing children with CP and OME
iii. performing VOI analyses to quantify the potential value of future research.

Methods

Systematic review of existing model-based economic evidence

A systematic search of the literature was conducted to identify published decision-analytic model-based economic evaluations of treatment options for the management of OME in children with CP. The search strategy (see *Appendix 11*) was designed to retrieve relevant studies from the following databases: MEDLINE, EMBASE and the American Economic Association's electronic bibliography (EconLit). These databases were searched from the date of their inception to January 2014. The systematic search did not identify any published economic studies. One published study[120] was found which gave a qualitative summary of guidelines on the surgical management of children with OME (CG60) produced by NICE.

A systematic review completed to inform CG60[11] identified three economic evaluations[121–123] that explored the cost-effectiveness of treatment options for the management of OME, but these were not model-based evaluations. A de novo economic model was developed to inform the recommendations for treatment options as part of CG60.[11] A decision tree was constructed that compared the deterministic incremental costs and benefits of four strategies [HAs; VTs; VTs plus adenoidectomy; and do nothing (DN)] for the management of children with persistent bilateral OME. The decision problem addressed in CG60 was not directly relevant to the management of OME in children with CP. Furthermore, the analysis made additional assumptions that limited its generalisability: (i) the time horizon for the analysis was 12 months; (ii) three surgical insertions of VTs were permitted to take place within a 12-month time frame to a proportion of children that is unlikely to reflect actual clinical practice; (iii) no gain in QALYs were assumed for children in the HAs and DN strategies; (iv) no probabilistic sensitivity analysis (PSA) was conducted. Neither the recommendations from the NICE CG60,[11] nor the results of the previously published economic evaluations, can be generalised to a population of children with CP. Therefore a de novo economic model was developed to evaluate the incremental cost-effectiveness of relevant treatment options for the management of OME in children with CP.

Economic model

A decision-analytic model was structured to estimate the costs and QALYs associated with four strategies for the management of persistent bilateral OME in children with CP: (i) VTs; (ii) HAs; (iii) HAs plus VTs; and (iv) DN.

The model structure

The model followed a hypothetical cohort of 10,000 children with CP and persistent OME under the age of 12 years. This age range for the relevant study population was used as there is evidence to indicate OME-related ear problems settle in children with CP after the age of 12 years.[124] Furthermore, the

incidence[65] and consequences[125] of OME decline as a child grows older. The model structure was informed by a previously published model that had been used to develop national guidance (CG60) on the management of children with OME.[11]

The model used the UK NHS perspective for costs. Hearing gain was the primary clinical outcome which was transformed, using published utility values, to generate an estimate of the impact on health-related quality of life (HRQoL). A decision tree was used to represent care pathways for a 24-month time horizon. The time frame for the analysis was based on the advice from clinical experts ($n = 3$) in the mOMEnt SMG and the literature which indicated two reasons: (i) a period of 24 months is a reasonable follow-up period to detect key outcomes following an intervention for OME;[11,78,80] and (ii) the total number of potential multiple VTs insertions and retractions could reasonably be performed within a 24-month period. All costs and benefits incurred beyond 12 months were discounted at an annual rate of 3.5%, as recommended by the NICE reference guide for the methods of technology appraisal.[126]

The schematic for the decision tree and care pathways represented is shown in *Figure 18*. Following the criteria for selecting an appropriate modelling approach set out by Barton *et al.*,[127] a decision tree structure was considered to be the most appropriate modelling approach for this decision problem. As the children within the model are assumed to be independent of each other, the care pathways outlined following

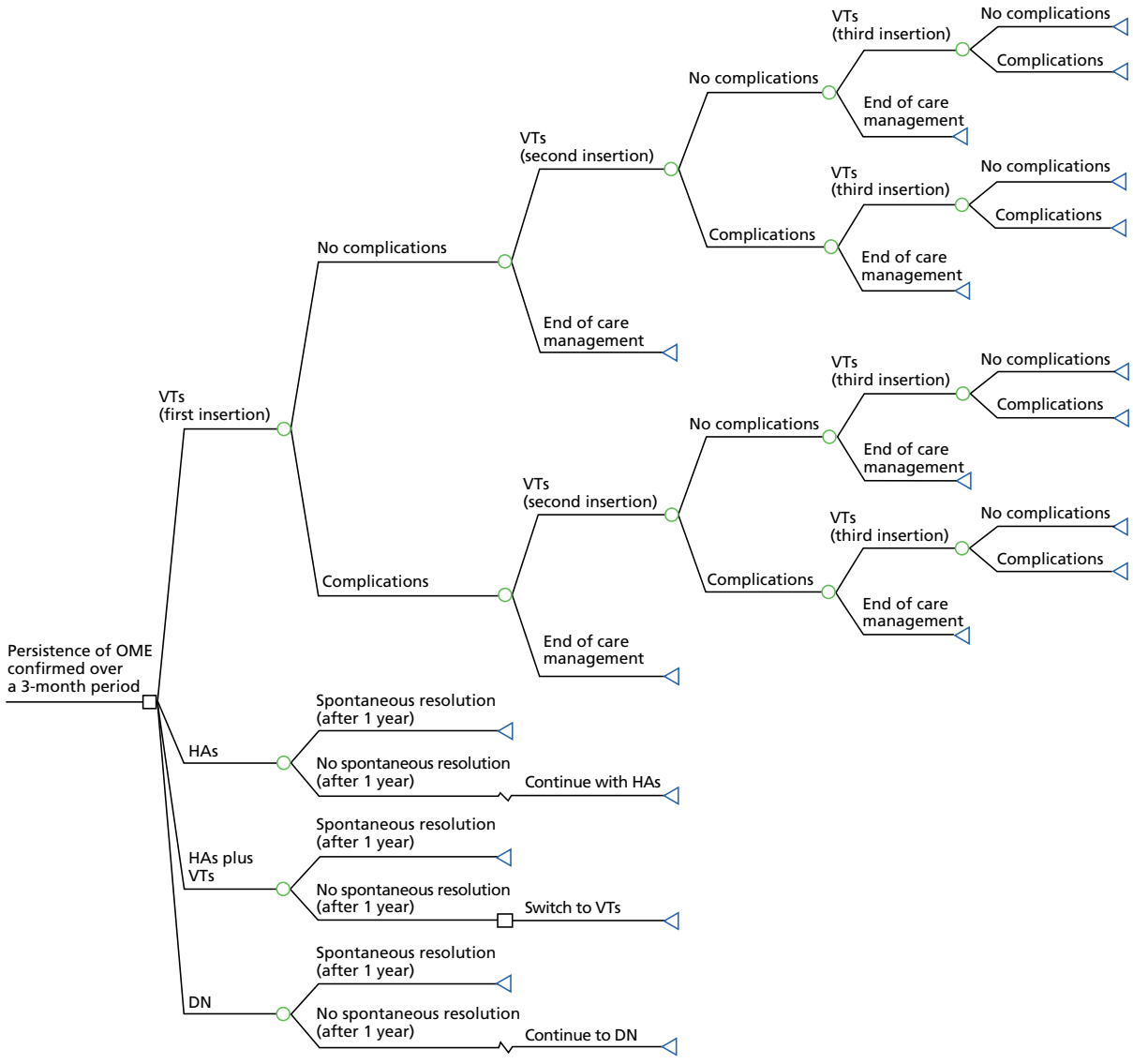


FIGURE 18 The decision tree diagram.

consultation with clinical experts can adequately be represented by a probability tree and a short follow-up time horizon was considered. The model was built in Microsoft Excel® 2010 (Microsoft Corporation, Redmond, WA, USA) and programmed using Visual Basic for Applications® to estimate the expected costs and benefits for each strategy.

The treatment strategies

The four strategies included in the economic analysis and the key assumptions associated with these strategies are now described.

Ventilation tubes strategy

The strategy involving the surgical insertion of VTs assumes that children with CP are recommended for the intervention once the persistence of OME has been confirmed over a 3-month period. The model starts at the point when the first surgical insertion of VTs takes place. A proportion of children are assumed to have a second insertion of VTs and a smaller proportion of children will require a third insertion because of persistent or relapsing OME,[11,124] or early extrusion of the VTs from the eardrum.[1,11] To avoid the high frequency of insertion in a single year, the second surgical insertion of VTs is assumed to take place within the first year of management follow-up, while the third surgical insertion is assumed to take place within the second year of management follow-up. Based on the literature,[11,124,128] the maximum number of VT insertions per child was limited to three within the assumed 24-month time horizon because children who undergo several VT insertions increase the risk of conductive hearing loss in the long run[70] and also to prevent scarring of the eardrum from repeated operations. The end point of the VT strategy was defined as 'end of care management' to represent when children do not require a subsequent insertion of VTs within the assumed 24-month time horizon for the care pathway.

The surgical insertion of VTs is commonly accepted as a safe operation, but there are some potential minor postoperative complications. None of the potential complications related to the VTs insertion operation are life-threatening. Following each surgical insertion, there is a probability of complications such as otorrhoea, granulation tissue formation and eardrum perforation. These complications were assumed to occur as reflected in the estimates extracted from the identified published literature (see *Table 54*). The risk of eardrum perforation is usually higher after repeated VTs insertion;[80,129] thus, a higher risk was expected for eardrum perforations in subsequent surgeries. Cholesteatoma (the abnormal collection of skin cells) formation was not included as a complication of OME because previous investigators among many others suggested that VT insertion can avert sequelae of OME such as cholesteatoma formation.[11,128,130,131] Although there may be a chance of calcium deposition within the eardrum with subsequent increased eardrum rigidity (tympanosclerosis) that could be either due to AOM,[132] OME itself[133] or VT insertion.[134] Therefore no incidence of tympanosclerosis formation was predicted as part of any of the strategies included in this analysis given that the actual cause of tympanosclerosis is not fully understood.[135]

No serious injury or surgical death was assumed to occur in this strategy since it is extremely unlikely for children to suffer serious injury or death from an insertion of VTs under modern anaesthesia.[136,137] Occasionally there may be a need to have the VTs removed,[11,80] and the impact of this has been explored in the model. It was estimated that children will have their first ENT review within 6 weeks of an operation and subsequent ENT reviews every 26 weeks thereafter until the mean 'extrusion time' (i.e. the time which the VTs should naturally fall out by) of 39 weeks.[11] It was also estimated that children will require one or two audiological review(s) after each surgery based on the advice published in current guidelines (CG60)[11] that hearing levels of children who underwent the insertion of VTs for OME should be reassessed postoperatively. The model assumed that a proportion of children who suffer from otorrhoea and/or granulation tissue formation will need a visit to a general practitioner (GP) for a course of antibiotics or eardrops.

Hearing aids strategy

The HAs strategy assumed that children with CP are offered the intervention once the persistence of OME has been confirmed over a 3-month period. It was estimated that in some children OME will resolve spontaneously by the end of 12 months.[4,138,139] However, the model assumed that children in whom OME had not naturally

resolved by 12 months will continue with using their HAs in the hope of spontaneous resolution without surgery. The initial costs of this strategy include the HAs, batteries for HAs, ear moulds (to help fit the HAs into a child's ear and enable the amplified sound to enter the ear canal), HAs care kit and HAs fitting in an audiology department. Batteries for HAs are estimated to need replacing every 4 weeks.[11] In addition, ear moulds are estimated to need replacing every 13 weeks[11] because ear moulds repeatedly turn yellow and inflexible with time and, hence, require replacement on a regular basis. For a proportion of children some of these costs are expected to be incurred again due to breakage or loss of HAs.[11]

Acute otitis media is a common sequelae in children who suffer from OME, which if left untreated will generally lead to episodes of AOM that require active intervention with a course of appropriate antibiotics.[139–141] AOM is the most common reason for children to take antibiotics.[139] The antibiotic, amoxicillin, is the treatment of choice to cure the AOM infection.[141,142] The HAs strategy included the use of antibiotics, and based on the meta-analysis conducted by Rosenfeld and Kay,[140] an average 2.8 episodes [95% confidence interval (CI) 2.2 to 3.4 episodes] of AOM were predicted to occur each year. AOM is one of the foremost causes of doctors' consultations[139] and prescribing antibiotics for AOM is known to encourage GP visits for subsequent episodes.[141] Thus, the children with untreated OME in this strategy were assumed to make 2.8 GP visits (on average) every year due to AOM episodes.[139–141] It was estimated that children will make one or two ENT visit(s) every year.[138,142] Furthermore, it was assumed that children will have their first audiological review after 13 weeks and subsequent audiological reviews every 26 weeks thereafter.[11] Previous work has suggested that adherence to wearing HAs is a problem,[4,138] because children frequently take the HAs device out. To reflect the impact of acceptability of wearing the HAs, the model also included an estimate of the level of adherence to wearing the HAs and associated impact on QALYs.

Hearing aids plus ventilation tubes strategy

The combined strategy of HAs plus VTs assumed that children with persistent OME confirmed over a 3-month period will initially be fitted with HAs. Children who do not experience spontaneous resolution of OME by the end of the first 12 months were then assumed to switch to the VTs strategy for the remainder of the follow-up period. Therefore, in effect, the pathways of this strategy resembled that of the first 12 months of the HAs strategy followed by the first 12 months of the VTs strategy.

Do nothing strategy

Do nothing is defined as an 'extended period of watchful waiting'. The DN strategy in the model therefore reflected extending the initial watchful waiting period of 3 months by a further 24-month period. This strategy assumed that children with CP have no planned intervention,[4,98,138] but they will be offered an appropriate course of antibiotics to treat any emerging instances of AOM.[139–141] Similar to the HAs strategy, children in this strategy were expected to experience 2.8 episodes (95% CI 2.2 to 3.4 episodes) of AOM every year.[140] As such, every year, the children with untreated OME were assumed to make 2.8 annual visits to the GP (on average) due to AOM episodes.[139–141] Furthermore, the children were assumed to require ongoing contact with health-care services including one or two audiological review(s)[4,138,142] and one or two ENT visit(s)[138,142] every year. The model assumed that, apart from the direct costs related to HAs devices and the need for any subsequent audiological reviews, the resource consequences of this strategy will effectively be similar to the HAs strategy.

Model input parameters

The data used to populate the model were derived from a variety of sources including systematic reviews of clinical effectiveness and existing economic evaluation literature, and rapid reviews of resource use and utility literature. Pondhuri *et al*.[1] also conducted a systematic review to identify all studies that reported on the association between early insertion of VTs and subsequent outcome in children with CP. Most of the studies identified from their systematic review of the relevant clinical literature were judged to be of low quality. The main challenges in terms of study quality were that identified studies were small, without sample size calculations and generally had poor reporting of data. The results of the systematic review of clinical effectiveness literature (see *Chapter 4*) confirmed that it was not possible to run a meta-analysis to estimate an overall measure of clinical effect because of study heterogeneity; thus, it was necessary to purposively select the papers deemed to have most direct relevance to the study population of interest.

The model inputs in terms of probabilities, clinical effectiveness, utility values, resource use and unit costs are now described.

Probabilities

The probabilities identified for each aspect of the care pathway associated with the VTs strategy (and sources of data) are shown in *Table 54* (for VT-related complications) and in *Table 55* (for VT insertion).

For the HAs strategy, a probability for breakage or loss over a 12-month period of 16.44% was used to populate the model. This value was calculated from estimates that 25% of children break or lose their HAs over a period of 21 months, presented by NICE,[11] under the assumption of a constant hazard data. Rosenfeld and Kay[140] reported a meta-analysis that generated the value for spontaneous resolution of

TABLE 54 Probability data for the insertion of VT-related complications

Complication	Probability	Distribution for PSA	Source	Notes
Otorrhoea	0.25	Beta~(3,9)	Maheshwar *et al.*[4]	In view of incidence of otorrhoea, a more conservative value was used. The value of 0.25 reported (for cleft population) by Maheshwar *et al.*[4] is in line with the meta-analysed value of 0.26 reported (for non-cleft population) by Kay *et al.*[129] Maheshwar *et al.*[4] conducted their retrospective study in the UK. Russell *et al.*[143] claimed a slightly lower risk of otorrhoea for the cleft population than the non-cleft population
Granulation tissue formation	0.042	Beta~(37,850)	Kay *et al.*[129]	Granulation tissue formation is a post-operative complication cited by Kay *et al.*[129] and CG60[11] among others
Eardrum perforation (first procedure)	0.024	Beta~(2,81)	Phua *et al.*[80]	A retrospective New Zealand-based study. Only one study was identified relevant to children with CP from a published systematic review that reported a subsequent higher risk of perforations of the eardrum due to repeated VTs insertion
Eardrum perforation two or more procedures)	0.098	Beta~(4,37)	Phua *et al.*[80]	

TABLE 55 Probability data for the insertion of VTs strategy

Parameter	Probability	Distribution for PSA	Source	Notes
Removal of VTs (first procedure)	0.072	Beta~(6,77)	Phua *et al.*[80]	This was the only identified study of children with CP from a published systematic review that reported different risk of retraction following subsequent VTs insertion procedures
Removal of VTs (two or more procedures)	0.171	Beta~(7,34)	Phua *et al.*[80]	
Reinsertion of second VT	0.38	Beta~(68,110)	Sheahan *et al.*[124]	This was a questionnaire-based study of cleft children. This was the only identified study that reported a reinsertion rate for two or more VTs. A previous model used a reinsertion rate of 0.25 for general children[11]
Reinsertion of third VT	0.38	Beta~(68,110)	Sheahan *et al.*[124]	
Time to extrusion	VTs fall out by 39 weeks	Normal~(39,2.93)[a]	CG60[11]	The 'time to extrusion' is defined as the time by when the VTs naturally should fall out

a This is estimated based on the information that VTs fall out between 26 and 52 weeks.[11]

chronic OME documented for 3 months or longer, and in line with this, the model predicted a spontaneous resolution rate of 30.8% by the end of the first 12 months for children in the non-surgical strategies. The resolution of OME was assumed to have a constant rate over 12 months, following advice from clinical experts and the evidence base. In calculating the QALY gain associated with the HAs strategy, the model assumed that 90.9% of the cohort of children will adhere to wearing their HAs, based on a published non-adherence rate of 9.1%.[138] The 9.1% of children who do not adhere to wearing their HAs were then assumed to have gain in QALY equivalent to that used in the DN strategy.

Health gain and utilities

Hearing level measured in decibels (dBHL) over two pre-defined time periods of 12 and 24 months was used to value the impact of each strategy on children with CP and OME. The study conducted by Maw and Bawden[144] was identified as the primary source to provide estimates for the quantity of hearing gain associated with each strategy, and in the baseline analysis the assumed hearing gains were (i) 13.06 dBHL after 12 months and 12.24 dBHL after 24 months for the VTs strategy; and (ii) 4.88 dBHL after 12 months and 7.57 dBHL after 24 months for the non-surgical strategies (HAs and DN). QALYs were then calculated to value the 'quality' of the observed gain in hearing, as a change in HRQoL, which was assumed to be a linear function of potential improvement in hearing. To calculate the QALYs, a utility value per unit increase in hearing gain was attached to the identified dBHLs for each strategy.

A systematic search strategy (see *Appendix 11*) was designed to identify relevant utility data suitable for informing estimates of QALYs for the economic model. The search was carried out in MEDLINE, EMBASE and EconLit. These databases were searched from the date of their inception to January 2014. Studies were considered as being eligible for inclusion if the studies (i) were published in peer-reviewed journals as full papers; (ii) reported HRQoL data based on utility values for 'hearing' of OME-affected children with no other comorbidity; and (iii) reported utilities that are appropriate for estimating QALYs. Eighteen references were identified from the electronic search strategy; none of which met the inclusion criteria.

Published expert opinion was used to apply a value for the assumed utility gain associated with per unit increase in dBHL. This estimate of a utility gain per unit increase in dBHL of 0.00874 (95% CI 0.00500 to 0.01200)[11] was based on the interpretation of an unpublished study by Kubba (2004)[145] that collected individual-patient data on the Health Utility Index Mark III for children with a median age of 5 years. The use of VTs can improve the level of a child's hearing by approximately 50.5% (95% CI 47.0% to 54.5%) when compared with no intervention.[140] To estimate the impact of HRQoL the utility gain per unit increase in dBHL of 0.00874 was reduced by 50.5% for the children in the DN strategy.

Resource use and costs

Table 56 summarises the point estimates of resource use and unit costs used for each strategy and pathway for the model together with the assumed ranges and distributions used in the PSA. All prices are presented in UK£ for the year 2010–11.

Probabilistic sensitivity analysis

Probabilistic sensitivity analysis was used to quantify the joint uncertainty in the model by assigning a range and specific distribution to each of the input parameters. The PSA was run using 10,000 iterations. Gamma distributions were used to represent the uncertainty in the cost parameters (see *Table 56*), because these values are constrained to be zero or positive. The gamma distribution is parameterised by two parameters (shape and scale), which are expressed as functions of the expectation and variance of the distribution. Beta distributions were used to represent the uncertainty in the probability parameters (see *Tables 54* and *55*) as these values are defined on the interval with a minimum (0) and maximum (1) value. The beta distribution is parameterised by two parameters (alpha and beta); alpha corresponds to the 'number of events' observed and beta corresponds to the 'number of non-events' observed. Normal distributions were used to represent the uncertainty in the hearing gain parameters to reflect the likelihood of an increase or decrease unit in dBHL during the recovery period: Normal~(13.06,9.49) after 12 months and Normal~(12.24,9.1) after 24 months for the VTs strategy; and Normal~(4.88,11.11)

TABLE 56 Resource use and unit cost data

	Resource use		Cost			
Parameter	Mean description (per child)	Distribution for PSA	Mean unit cost (£)	Distribution for PSA	Source	Notes
Insertion of VTs	One procedure	Fixed	891	Gamma~(130.45,6.83)	NSRC1[146] 2010–11 (HRG[146] code CZ08T; day case)	Minor ear procedures for children aged ≤ 18 years through tympanic membrane. One procedure is equivalent to inserting two VTs for each child due to bilateral OME
Tympanoplasty	One procedure	Fixed	1831	Gamma~(100.11,18.29)	NSRC1[146] 2010–11 (HRG[146] code CZ10U; day case)	Major ear procedures for children aged ≤ 18 years due to perforation of eardrum
Removal of VTs	One procedure	Fixed	891	Gamma~(130.45,6.83)	NSRC1[146] 2010–11 (HRG[146] code CZ08T; day case)	Minor ear procedures for children aged ≤ 18 years through tympanic membrane. One procedure is equivalent to removing one or two VT(s) for each child due to bilateral OME
HA	Two units	Fixed	80	Fixed	GDG[11] estimate has been inflated based on the HCHS[147] index 2010–11 (PSSRU[147])	Two HAs are required for each child due to bilateral OME
Ear mould	Eight units per year	Fixed	17	Fixed	GDG[11] estimate has been inflated based on the HCHS[147] index 2010–11 (PSSRU[147])	Two ear moulds are required for each child every 13 weeks[11]
HA care kit	One unit	Fixed	20.88	Fixed	Connevans[148]	This cost incurred once due to the maintenance of the HA(s)
HA battery	Twenty-six units per year	Fixed	0.49	Fixed	HAB[149]	Batteries are required to be replaced every 4 weeks[11]
HA fitting	One procedure	Fixed	76	Gamma~(71.03,1.07)	NHS reference costs (2005–6)[11] (service code AS1FA) has been inflated based on the HCHS[147] index 2010–11 (PSSRU[147])	HAs fitting in an audiology department. One procedure is equivalent to fit two HAs for each child due to bilateral OME

continued

TABLE 56 Resource use and unit cost data (*continued*)

	Resource use		Cost			
Parameter	Mean description (per child)	Distribution for PSA	Mean unit cost (£)	Distribution for PSA	Source	Notes
GP visit/antibiotic medication	VTs: 0.25 (otorrhoea) + 0.042 (granulation) time	See *Table 54*	GP visit: 41 Antibiotic medication: 11	Fixed	GP visit: PSSRU[147] (unit costs of health and social care 2010–11) Antibiotic medication: GDG[11] estimate has been inflated based on the HCHS[147] index 2010–11 (PSSRU[147])	GP visit and cost for a course of antibiotics or eardrops due to otorrhoea, granulation and/or AOM
	HAs: 2.8 times	Normal~(2.8,0.3)				
	DN: 2.8 times	Normal~(2.8,0.3)				
Audiological review	VTs: 1.5 visits following each surgery	Uniform~(1,2)	48	Gamma~(64,0.75)	NHS reference costs (2005–6)[11] (service code AS1FU) has been inflated based on the HCHS[147] index 2010–11 (PSSRU[147])	One or two visit(s) following each surgery for VTs.[11] One visit within 13 weeks and subsequent visits every 26 weeks thereafter for HAs.[11] One or two visit(s) every year for DN[4,11,138,142]
	HAs: 1 visit initially + 1.5 visits per year thereafter	Fixed				
	DN: 1.5 visits per year	Uniform~(1,2)				
ENT specialist visit	VTs: 2.5 visits following each surgery	Fixed	91.72	Gamma~(84.14,1.09)	NSRC1[146] 2010–11 (service code 120)	One visit within 6 weeks of each surgery and subsequent visits every 26 weeks thereafter until the time of extrusion.[11] One or two visit(s) every year[138,142] for both HAs and DN
	HAs: 1.5 visits per year	Uniform~(1,2)				
	DN: 1.5 visits per year	Uniform~(1,2)				

GDG, Guideline Development Group; HAB, Hearing Aid Batteries; HCHS, Hospital and Community Health Service; HRG, Healthcare Resource Group; PSSRU, Personal Social Services Research Unit.

after 12 months and Normal~(7.57,12.76) after 24 months for the non-surgical strategies.[144] The utility gain per unit increase in dBHL was parameterised by a Gamma distribution [Gamma~(24.38, 0.0004)] with the shape (24.38) and scale (0.0004) determined from the mean and variance reported by Kubba.[145]

The probability of spontaneous resolution was sampled from the distribution labelled as Beta~(61,137).[140] The probability of breakage or loss of HAs was re-estimated within the PSA based on uncertainty surrounding the original 21-month data that was represented by a Beta~(6,18).[138] Adherence to HAs was sampled from Beta~(20,2),[138] whereas expected episodes of AOM were sampled from Normal~(2.8,0.3).[140] Based on the statement described earlier that VTs can improve a child's quality of hearing by approximately 50.5% (95% CI 47.0% to 54.5%)[150] when compared with DN, the QALY gain associated with the DN strategy was adjusted according to a normal distribution [Normal~(0.505,0.02)].

Value of information analysis

Using the decision tree structure and subsequent PSA, an expected value of perfect information (EVPI) analysis was conducted to estimate the potential value of future research. *Equation 1* shows that EVPI estimates the difference between the expected value of a decision made with perfect information and the expected value of a decision made on the basis of the current evidence base:

$$EVPI = E_{\theta} \max_j NB(j,\theta) - \max_j E_{\theta} NB(j,\theta), \quad (1)$$

where $E_{\theta}\max_j NB(j,\theta)$ represents the expected value of the decision with perfect information and $\max_j E_{\theta} NB(j,\theta)$ represents the expected value without perfect information. Using *Equation 2*, the net benefit (NB) associated with each treatment strategy was calculated by combining the respective health gain and expected cost consequences:

$$NB = \lambda . E - C, \quad (2)$$

where λ represents the willingness-to-pay (WTP) threshold, E represents the QALY gained and C represents the expected cost.

In its simplest form, EVPI represents the maximum amount that a decision-maker would be willing to pay to gain access to perfect information. However, the societal value of research should ideally be estimated across the population of future patients for whom the decision is pertinent since the information provided by research is a public good. *Equation 3* shows the calculation of the population-level expected value of perfect information (pEVPI):

$$pEVPI = EVPI \cdot \sum_{t=1}^{T} \frac{1_t}{(1+r)^t}, \quad (3)$$

where T = effective lifetime of a technology; I_t = incidence of the condition relevant to the health technology in period T; and r = discount rate. pEVPI represents an upper bound of the expected benefit of conducting further research. If pEVPI is greater than the expected cost for conducting further research, then it should potentially be considered worthwhile to conduct the further research. Here the estimate of the population was based on an assumption that every year 720 children will be eligible for VTs in the UK. Data from the CRANE database showed there were 800 children born with CP in England, Wales and Northern Ireland in 2012.[151] Of which, 720 (90% of 800[113,124,152,153]) were assumed to suffer from OME. The lifetime for the technology was assumed to reflect that the decision would be relevant for 10 years (T). Armstrong[154] first described the use of VTs in 1954, and, since then, use of VTs to restore hearing to normal has been increased. Given the historical longevity of the technology revealed in the literature, it seemed reasonable to assume that use of VTs will last for at least another 10 years before a new technology comes along and replaces it. A discount rate of 3.5% (r) was used to be in line with the recommendations in the NICE reference guide for methods of technology appraisal.[155]

The basic method for estimating EVPI was then extended to identify the type of evidence which will be most important by identifying the parameter(s) for which more precise estimates would be most valuable.[156,157] The expected value of partial perfect information (EVPPI) can be estimated using *Equation 4*:

$$EVPPI_{\vartheta} = E_{\vartheta}\max_j E_{\varphi/\vartheta}NB(j,\varphi,\vartheta) - \max_j E_{\theta}NB(j,\theta). \quad (4)$$

Here, ϑ represents parameter(s) of interest, φ represents other uncertainties, θ represents all parameters, $E_{\vartheta}\max_j E_{\varphi/\vartheta}NB(j,\varphi,\vartheta)$ corresponds to the expected value with perfect information for parameter ϑ, and $\max_j E_{\theta}NB(j,\theta)$ corresponds to the expected value of current information for all parameters θ. The EVPPI analysis was conducted on four parameters including (1) unit cost of surgical procedure (in isolation); (2) dBHL (in isolation); (3) unit measurement of dBHL (in isolation); and (4) dBHL plus utility gain per unit increase in dBHL (in group). These parameters were identified a priori by clinical members of the mOMEnt study team ($n = 3$) as parameters most likely to impact on the relative expected costs and QALY gains of each of the four management strategies. Following the recommendations made in by Brennan *et al.*,[156] a total of 100,000 simulations (100 simulations in the outer loop and 1000 simulations in the inner loop) were used to estimate the maximum possible EVPPI values.

Results

The results of the baseline (deterministic) and probabilistic analyses are shown in *Table 57*. All results were initially generated for a hypothetical cohort of 10,000 children but the final results are presented in terms of costs and QALYs gained per child. The expected values from the PSA showed that the use of HAs plus VTs strategy was the most expensive option at £2663 per child. The insertion of VTs was the second most costly strategy with a cost of £2086 per child compared with £1237 per child for the HAs strategy and £593 per child for the DN strategy. The associated gains in QALY were 0.218 for the VTs strategy; 0.136 for HAs plus VTs; 0.102 for HAs; and 0.053 for DN.

The incremental cost-effectiveness ratios (ICERs) per QALY gained based on the expected values from the PSA were (i) £13,143 per QALY gained for the HAs strategy compared with the DN strategy; and (ii) £7338 per QALY gained for the VTs strategy compared with the HAs strategy. The HAs plus VTs strategy was dominated by the VTs strategy because it was shown to be less effective and more costly. Applying the weak dominance principle shows that the HAs strategy was extended dominated by the VTs strategy because HAs compared with DN has an ICER (£13,143 per QALY gained) greater than that of VTs compared with HAs (£7338 per QALY gained). This means that the VTs strategy should be compared with the DN strategy, giving an ICER of £9065 per QALY gained. All four strategies were included in the subsequent PSA and VOI analyses as there could be some possible realisations of the uncertainty where the order of costs and QALYs gained would change, which would affect the relevant comparators for the incremental analyses.

TABLE 57 Expected cost and QALY gain for each strategy from the deterministic and probabilistic analyses

	Deterministic		Probabilistic		ICER per QALY gained	
Strategy	Cost (£)	QALY	Cost (£)	QALY	Deterministic	Probabilistic
DN	592	0.0528	593	0.0529	–	–
HAs	1235	0.1017	1237	0.1019	Extended dominated by VTs	Extended dominated by VTs
VTs	2083	0.2175	2086	0.2176	£9053 (VTs vs. DN)	£9065 (VTs vs. DN)
HAs plus VTs	2661	0.1357	2663	0.1358	Dominated by VTs	Dominated by VTs

ICER, incremental cost-effectiveness ratio.

Figure 19 shows the scatter plot of expected incremental costs and effects (gain in QALYs) for each of the 10,000 iterations run in the PSA. The DN strategy was assumed to be the status quo and hence has been anchored at the origin. Comparing the values for positive expected incremental costs and gain in QALYs, some 77% of the simulated values for HAs versus DN, 90% for VTs versus DN and 85% for HAs plus VTs versus DN, fell in the north-east quadrant of the cost-effectiveness plane. The north-east quadrant represents values in which an intervention would be more costly and more effective compared with its comparators, and it then becomes necessary to make a decision about the threshold value for WTP for an additional QALY. The PSA revealed that the HAs strategy was extended dominated by the VTs strategy for some 61% of the simulated realisations. *Figure 19* indicates that some of the expected costs and gain in QALYs would result in negative ICERs. For this reason, it was not appropriate to calculate pseudo-CIs around the mean estimates of ICERs from the PSA.

Figure 20 presents cost-effectiveness acceptability curves for each of the strategies based on the results of the PSA. The probability that the VTs strategy was cost-effective is 0.49 at the WTP threshold of £10,000 per QALY and 0.63 at the WTP threshold of £20,000 per QALY.

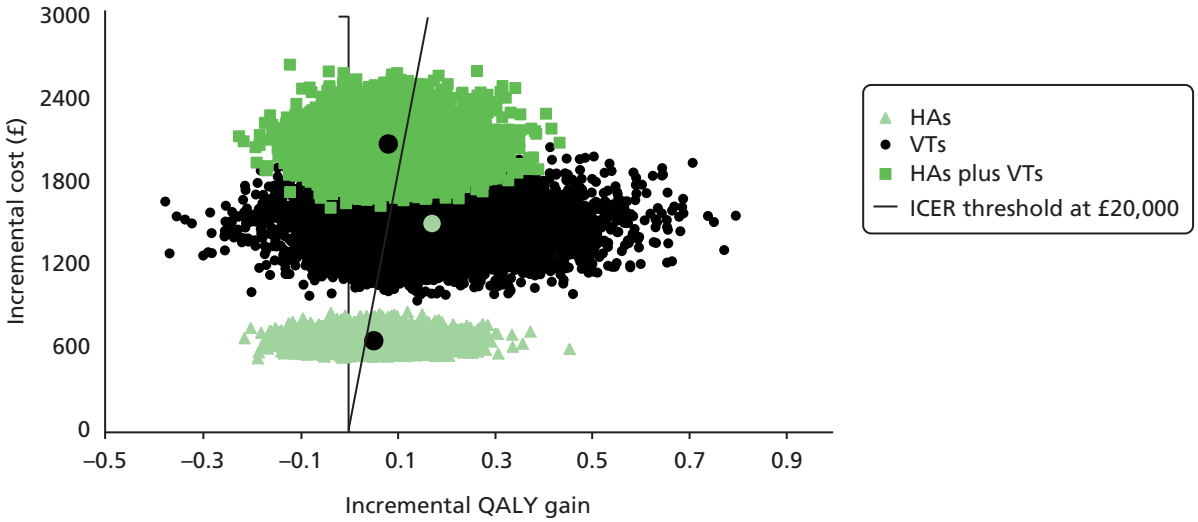


FIGURE 19 Scatterplot of incremental expected costs and QALYs obtained from PSA. The circles represent the expected value from the PSA. The DN strategy was anchored at the origin.

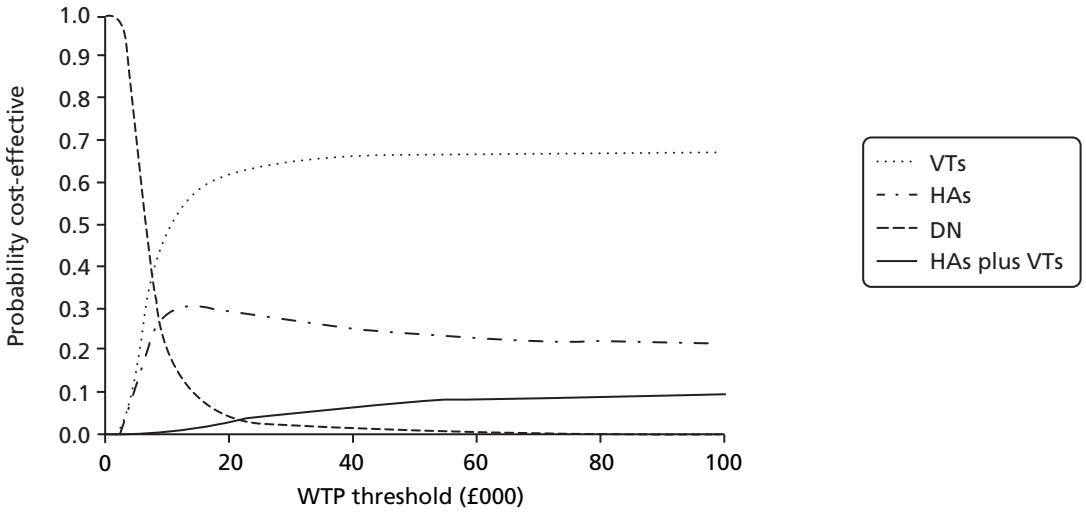


FIGURE 20 Cost-effectiveness acceptability curve for four strategies.

The EVPI values at both individual and population levels for a range of different values of the WTP threshold are presented in *Table 58*. At the population level, the maximum potential value of approximately £5.24M at an assumed WTP threshold of £20,000 suggests that further research work in assessing the impact of the surgical insertion of VTs for the management of persistent bilateral OME in children with CP could potentially be worthwhile, provided that the total cost of undertaking the further research remains under this estimated EVPI value.

Figure 21 shows the relationship between the pEVPI values and different values of WTP per QALY gained. *Figure 21* indicates that the value of further research exceeds £4M for all values of the WTP threshold beyond £10,000 per QALY gained. This value is likely to exceed the total cost of future research. At a WTP threshold of £30,000 per QALY gained, the value of further research exceeds £7M. However, should the WTP threshold be < £1500, the pEVPI is zero indicating that there is no value in additional information from future research.

The EVPI analysis was important in deciding whether or not the value from undertaking further research could be worthwhile. The EVPPI analysis extended this analysis to provide the breakdown values of further research for key parameters. The EVPPI analysis, using an assumed WTP threshold of £20,000 per QALY, suggested that further research on dBHL with or without utility values could be potentially worthwhile. *Figure 22* shows the maximum possible population EVPPI values associated with a number of key uncertain parameters at the WTP threshold of £20,000. For instance, improving the estimate of dBHL parameters would accrue the maximum possible return of approximately £3.5M at an assumed WTP threshold of £20,000.

TABLE 58 Individual and population EVPI values at different WTP thresholds

WTP threshold (£)	Individual EVPI (£)	pEVPI over a 10-year decision horizon (£)	pEVPI over a 5-year decision horizon (£)
5000	102	632,148	343,191
10,000	641	3,972,619	2,156,720
15,000	721	4,468,422	2,425,889
20,000	845	5,236,916	2,843,101
25,000	988	6,123,164	3,324,242
30,000	1136	7,040,399	3,822,205

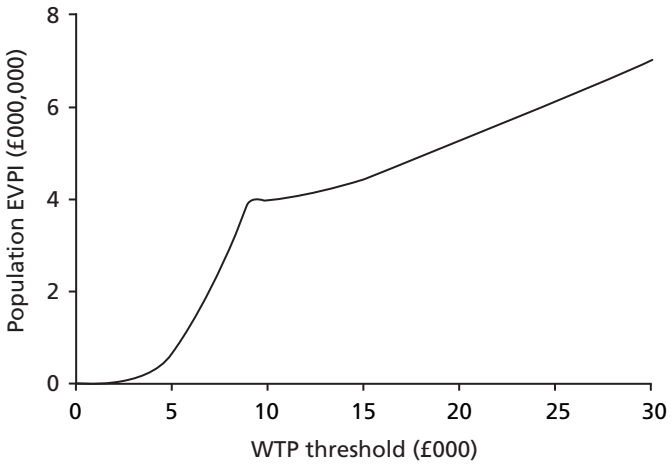


FIGURE 21 Expected population value of perfect information at various WTP thresholds.

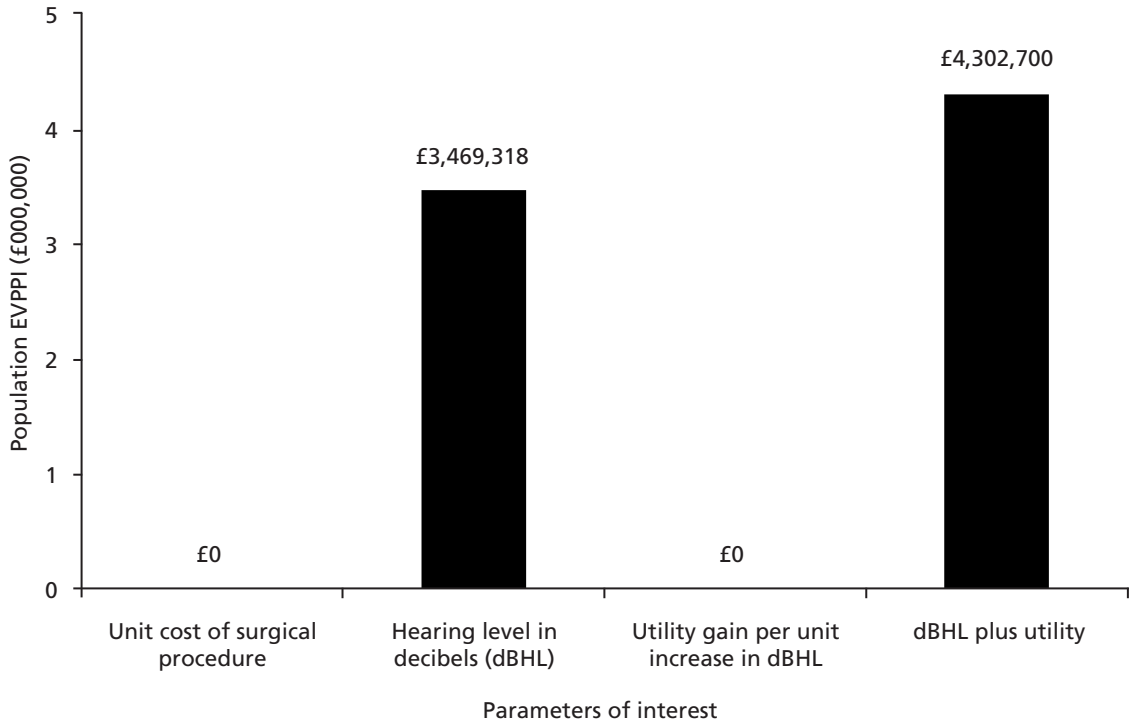


FIGURE 22 Population EVPPI values for key uncertain parameters at the WTP threshold of £20,000.

Discussion

The association between children with CP and hearing loss that results from OME is well documented.[64,158] The surgical insertion of VTs is one of the most common surgical procedures in childhood today.[134,136] Although the disagreement regarding the relative benefits and risks for the insertion of VTs in children with CP is unresolved within the surgical community, affected children still desire to function better and parents want their children to be in a position to participate fully in education. Despite a large body of evidence on incidence and prevalence rates of OME, there is still a paucity of research on the potential impact of the surgical insertion of VTs in children with CP. Therefore it was essential to assess whether or not the surgical insertion of VTs can have a positive impact on expected health benefits. A systematic search of the published literature was conducted to identify decision-analytic model-based economic evaluations of surgical insertion of VTs in the management of persistent bilateral OME in children with CP. No economic evaluations were identified that were relevant to children who are born with CP. Hence a de novo model-based economic analysis was carried out to assess the impact of surgical insertion of VTs compared with three alternatives in the management of bilateral OME persisting after the watchful waiting for 3 months in children with CP.

The insertion of VTs strategy was found to be the optimal strategy with the expected value of the ICER from the PSA of £9065 per QALY gained compared with the DN strategy. The HAs and VTs strategy was dominated by the VTs strategy. The HAs strategy was extended dominated by the VTs strategy. The ICER for VTs was well below the WTP threshold of £20,000 per QALY, which is commonly taken by NICE to be a reasonable threshold for WTP for an additional QALY.[159] The gain in QALY resulting from improvement in dBHL associated with the VTs strategy to manage OME came at a higher cost, which was mainly driven by the resource use attributable to the surgical process. The results of the PSA indicated the existence of considerable uncertainty surrounding the existence and extent of the incremental QALY gain associated with the VTs strategy compared with the DN strategy. In addition, there was some uncertainty surrounding the extent of the incremental cost associated with the VTs strategy compared with the DN strategy. This observed uncertainty perhaps explains why there remains disparity in the medical community regarding the use of VTs in individual children with CP for OME.

The results from this early economic analysis should not be used to inform any current changes in clinical practice; it was carried out to understand whether or not there is a need for further research regarding the utilisation of VTs in children with CP and persistent OME. A key strength of this analysis was that it extended the analysis beyond PSA and calculated the EVPI and EVPPI values to provide a measurable insight of whether or not further research in this area is potentially worthwhile. The feasibility and implications of using the EVPI and EVPPI methods for informing the future research prioritisation process have previously been well described and their use is recommended in the context of commissioned HTAs.[160] The maximum potential EVPI values of approximately £845 for every child and £5.24M for a population of children in England, Wales and Northern Ireland, assuming the WTP threshold of £20,000 per QALY and a decision horizon of 10 years, suggest that further research work in this area is potentially worthwhile.

This model was an early economic evaluation that used all available data from multiple sources. However, there were some limitations in terms of the availability and relevance of the data used for parameter inputs, which should be revised and reassessed once more relevant clinical effectiveness, resource use and utility data become available. The calculation of QALYs was driven by a combination of utility values and effectiveness data, specified in terms of dBHL. The key uncertainty in the model inputs indicated the need for further research to obtain more reliable and relevant values for the dBHL parameters. This analysis used a utility value taken from a published source and the same value has been used previously in an appraisal completed by the Guideline Development Group.[11] A further area of research is to explore whether or not gain in QALYs, that focus on measuring improvements in health status, are the only relevant outcome to assess an intervention aimed at young children. Other non-health gains such as improvements in educational attainment and ability to play with their peers may also be important outcomes for children and their parents.

The eligible patient population was also an area of uncertainty suggested to be important by clinical experts. This economic evaluation focused on a cleft population of children under the age of 12 years. This focus was necessary because of a paucity of epidemiological data for other age groups. This age group was selected for this analysis because clinical experts considered this group to represent children in which the condition is most prevalent. However, further work is needed, informed by robust epidemiological data, to understand the relative cost-effectiveness of the insertion of VTs in different age groups and also the most appropriate age for the surgical procedure in a child with CP. This analysis used the only available source for estimates of health-care resource use. These data were not directly relevant to a population of cleft children, but provided the best evidence in absence of directly relevant data. Using these data was likely to be a conservative approach as OME is relatively more common in cleft children compared with non-cleft children and has an extended recovery period.[124]

This analysis posed two technical challenges. Interpretation of this analysis should be undertaken with caution as, with no definitive guidelines identified for the treatment of OME in children, the clinical pathway used to structure the economic evaluation was developed using assumptions based on available published evidence. Therefore the clinical pathways used to structure the economic model were developed using assumptions based on an existing economic model, used in a previous appraisal conducted by NICE,[11] and adapted for a population of children with CP using advice from clinical experts. Furthermore, the limited number of studies meant that it was difficult to generate ranges based on empirical data around some parameters included in the PSA.

This is the first model-based economic evaluation to identify and quantify the costs and benefits of different management options of persistent bilateral OME in children with CP. The model has demonstrated the potential for resources to be released from other health-care interventions when VTs insertion is applied for managing OME. The total cost of the VTs strategy is relatively high, but this intervention appears to provide good value for money, based on the current evidence base, if used after the initial 3-month period of watchful waiting as a means to correct significant hearing impairment and prevent complications of untreated OME. The early management of OME-related complications should generate expected NBs that might compensate the additional expenditure incurred because of repeated

clinic visits[141] and prompt rapid hearing gain that is also important for childhood speech development and associated educational attainment.[161,162] Schönweiler *et al.*[163] showed that language development depends more on hearing ability than severity or surgical repair of CP. Paradise and Bluestone,[64] some 40 years ago, advocated a policy of early VTs and replacements when necessary in order to decrease the long-term otological complications and minimise the effects on speech and language development. Furthermore, the use of VTs could release clinicians from the pressure to prescribe antibiotics to manage multiple instances of infections, which could impact on antibiotic resistance.[114,164] However, this analysis has shown limitations in the current evidence base and identified that it is potentially worthwhile undertaking further research in this area. Examples of the additional evidence that is needed include the link between hearing gain and utility gain; the actual use of health-care resources; and clinical effectiveness data to inform the appropriate age for the insertion of VTs in children with OME. Furthermore, another issue that could not be identified by using EVPPI, but is clearly relevant given the nature of the intervention and target patient population, is whether or not using hearing gain alone as an outcome is appropriate.

Conclusion

Based on the assumptions used in this analysis, the surgical insertion of VTs for the management of persistent bilateral OME in children with CP is most likely to be a cost-effective strategy, but the need for acquiring further information from future study is evident to inform this treatment choice. The probability that the VTs strategy is likely to be more cost-effective than its comparators was about 63% at the WTP threshold of £20,000 per QALY. The EVPI analysis has shown that undertaking further research in this area is potentially worthwhile. The EVPPI analysis indicated that the main uncertainty centres around the estimate of dBHL parameters. Consequently, if future research is to be undertaken it should then aim to improve the estimate of dBHL parameters using a RCT design. Future research may also focus on improving the estimate of utility values to the observed change in dBHL from a cohort study or substudy within a trial. The results presented here should not be considered as an option for all age groups; thus, further research is required to identify subgroups of cleft children likely to benefit most from the surgical insertion of VTs.

Chapter 6 Summary discussion and conclusions

This section of the report is concerned with a discussion of our findings that were presented to the SAG in order for them to make recommendations on potential study designs. It then concludes with our final recommendations on further studies.

Clinician survey

This part of the study suggested that some of the UK cleft centres were in a position to take part in a future study. These were centres that could nominate a lead ENT/audiologist to act as a local principal investigator and they were able to perform age-appropriate hearing testing from 1 year old to adolescence. These centres also exhibited adherence to the NICE Guideline Development Group guidelines and prescribed both HAs and VTs.

One of the most relevant findings of the survey was concerned with the method of delivery of care. It was clear that most of the networks operated a 'hub and spoke' infrastructure with the centre operating a monitoring service and recommending that care be provided locally to the patient in hospitals or smaller clinics. This has implications for potential study design. For example, it would be difficult to engage peripheral clinicians with the random allocation of care, as they may not be in equipoise and the probability of protocol deviations would be high. Furthermore, obtaining trust R&D for multiple sites with potentially low caseloads would be problematic and inefficient. Finally, there will be the additional problem of standardising both audiological assessment and treatment away from the 'hub' clinic. As a result, it is important that if a trial were to be carried out consideration should be given to an evaluation of whether or not the patients would need to receive their treatment in the hub or centre. This will result in a reduction in eligibility of patients in that they will have to either live in the catchment area of the centre or be willing to travel to the centre for their treatment.

Another important finding was that despite our best efforts in engaging the clinicians and encouraging them to participate in the survey, the co-operation level was not high. In effect, the clinicians were either unwilling or unable to provide us with all the information that we required, although the condensed information was good. This suggests that there is a lack of engagement of the wider clinical community with this study and this should be considered in identifying the design of a further study.

Finally, the yearly caseload figures of children with non-syndromic CP for each of the centres showed some uniformity in caseload with most centres seeing between 35 and 60 new referrals per year, three centres received between 90 and 130 referrals per year, while four had < 35 referrals per year. Although these figures need to be interpreted with a degree of caution as the CRANE database tends to slightly under-record patients (because not all parents consent for their child's information to be included), it is generally felt to provide a good approximation of annual caseload. It is clear that for the effective running of a potential study that the centres with the higher caseloads are included. *Table 59* includes data on yearly caseload and an estimate of numbers who would be recruited into a trial. This estimate is based on the number of patients who are likely to have OME (90%) and then taking a conservative estimate of those who would meet trial eligibility criteria (50%), and finally factoring in the predicted consent rate estimated from the qualitative research (25%).

TABLE 59 The yearly caseload and estimate of potential number of patients who could be recruited into a trial

Centre	Number of new referrals (non-syndromic)	Estimate of potential numbers recruited into a trial
Newcastle	65 (49)	6
Leeds	65 (49)	6
Liverpool	64 (48)	6
Manchester	69 (52)	7
Nottingham	93 (70)	8
Birmingham	121 (91)	10
Cambridge	87 (65)	8
North Thames	173 (130)	14
Oxford	45 (34)	4
Salisbury	53 (40)	5
Swansea	51 (38)	5
Bristol	65 (49)	6
South Thames	145 (109)	12
Belfast	31 (23)	3
Edinburgh	29 (22)	3
Glasgow	46 (35)	4

Qualitative component

This part of the study provided us with useful information on outcomes that were important to parents and patients, and their willingness to take part in a potential study. All the aims of this component were achieved.

It was clear that parents and children held strong opinions about treatment and participation in a future trial. Importantly, only 25% of those in our sample would be happy to enter their children into a trial. This reflected the fact that most parents were not in equipoise. We also felt that parents required comprehensive and detailed information about HAs and VTs. In addition to information on safety procedures in a trial, based on interview data, the following appear to be important: a clear explanation of clinical equipoise; a need for the investigators to understand patients/parents previous experience of treatment (bearing in mind that the burden of care for a child with a cleft is very high); ensuring that the study is introduced by clinicians with whom the parent and child are familiar and trust; and finally emphasising how the study will enhance knowledge and help others in the future. Addressing these issues may optimise trial recruitment.

When we evaluated outcomes that were important to interviewees, they stressed the significance of speech and language development, educational outcomes and establishing social networks. It was also clear that some of their concerns were not solely related to hearing difficulties but were associated with having a cleft. As a result, although hearing was the key outcome, this was largely because of its consequences on social and educational development and psychological well-being. The outcomes from this part of the project fed into the component of the study concerned with the development of a COS.

Core outcome set

This was the first time that a COS had been developed for research in children with cleft lip and palate. It is important to point out that we have only identified 'what' should be measured; we have not made any recommendations on 'how' the outcomes should be measured. This requires further development that is outside of the scope of this feasibility study. Although the current COS has had input from clinical stakeholders, parents and children with CP, the proportion of parents and children contributing was lower than we would have liked. We will build on our current work to ensure that the opinion of parents included in the current COS is representative.

Economic analysis

This work represents the first model-based economic evaluation study to identify and quantify the costs and benefits of different management options for children with clefts. The objective of this part of the feasibility study was to perform a decision-analytic model-based economic analysis to assess the potential impact of the surgical insertion of grommets for the management of persistent bilateral OME in children with CP.

The aim of this model was to provide information on the potential key drivers of cost-effectiveness given the current evidence base for each of the potential strategies for managing children with CP and OME. The focus of the health economics work involved:

1. completing a systematic review and critical appraisal of published models that aim to evaluate the incremental costs and benefits of the insertion of grommets in children
2. structuring and populating a decision-analytic model to determine the ICERs of 'VTs' strategy compared with 'HAs', 'HAs plus VTs' and 'DN' for the management of persistent bilateral OME in children with CP
3. performing the VOI analyses to demonstrate the value for money from the surgical insertion of VTs.

This provided highly relevant and useful information to the study. First, it emphasised the limitations of the current evidence base for the management of OME in children with CP. It also revealed that the surgical insertion of VTs is likely to be the most cost-effective option. Nevertheless, the need for additional information from a future study is required to inform this treatment choice. Importantly, the EVPI was approximately £5.24M for a population of children with CP in England, Wales and Northern Ireland, assuming the WTP of £20,000 per QALY and a decision horizon of 10 years, this suggests that further research work in this area is potentially worthwhile.

The EVPPI analysis also illustrates that if future research is to be commissioned, it should then prioritise improving the estimates of parameters ('utility' and 'hearing gain in decibels') used to calculate the QALY gain associated with each strategy because further information on these parameters could have a significant impact on decision uncertainty. However, interpretation of this analysis should be undertaken with caution, as with no definitive guidelines identified for the treatment of OME in children, the clinical pathway used to structure the economic evaluation was developed using assumptions based on available published evidence.

The effect of other research projects on potential co-operation of the centres

There are currently two major studies involving the cleft centres. These are a study into the timing of primary surgery (Timing of Primary Surgery for Cleft Palate; TOPS) and the Bristol Gene Bank and Birth Cohort. Although both these studies are involving younger children, it is important that we consider that there is risk of the staff in the cleft centres becoming 'research fatigued' and this will influence potential co-operation capability of some centres.

Meeting of the Study Advisory Group to decide on potential study design

The SAG met with members of the study team and evaluated the information that was derived from the components of the study. They made the following recommendations:

1. The primary outcome of a potential trial would be hearing. They generally supported the concept that a difference in hearing loss of 15 dB between two interventions would be a worthwhile clinical difference to detect.
2. The SAG suggested that we should explore the use of the OMQ-14 questionnaire to collect some information on hearing and other outcomes that may be included in the COS.
3. They suggested that the 'ideal' study design could be a cohort with a nested trial. All children aged 2 years old with OME, diagnosed by otoscopy and tympanometry, and CP would be entered into the cohort, with the study information including description of randomisation in the event that an intervention was needed for OME. Prior to commencement of the study, the audiological assessment protocol for centres enrolled in the study should, where necessary, be adapted and standardised. This would ensure conformity of assessment throughout the study and minimise impact on existing practice at individual centres. Additional information about the impact of OME could be collected between audiological assessment appointments using an appropriate validated questionnaire. At, or following, recruitment should a child meet the criteria for active treatment of OME (NICE CG60[11]) they would be asked to participate in the randomised trial of treatment involving HAs or VTs. All the children would be followed until they were aged 5 years. This is a standard data collection point for all children with clefts in the centres.[165]

Nevertheless, they felt that this would be a highly ambitious study and we should make this recommendation with caution because of difficulties associated with the 'hub and spoke' model of care, the potentially low participation rate of patients and concerns with the engagement of clinicians.

Recommendations on future research

Before we make our final recommendations, it is useful to consider the HTA commissioning brief. This stated that the study should be directed at

> *What is the most appropriate way to manage OME in children with CP.*

and

> *To carry out a feasibility and VOI study to assess the possibility of a randomised controlled trial or multicenter prospective cohort study. The primary outcome of the feasibility study is to identify the feasibility of a definitive study.*

With this in mind, we have not attempted to design a trial but have aimed to provide information to the HTA programme which will help it to consider whether or not to commission further research and decide on options for the potential design of that research.

If we address the first point on whether or not a multicentre cohort is currently feasible, we are clear that a cohort study should not be explored for the following reasons:

1. The model of delivery of care would lead to major difficulties with monitoring the care provided and the uniformity of measurement of outcomes such as hearing, unless the cohort receives treatment in the centre and not in the spokes of the network.
2. There is a high risk of non-engagement of the clinical community.

3. There will be major issues with minimising bias.
4. If the cost of a cohort study would be similar to that of a trial, the trial would be the preferred option because the VOI, in terms of providing information that would reduce uncertainty, would be much greater with a trial.

Potential future trial

We decided that the primary outcome for a potential study would be hearing loss, based on the information from the economic analysis, the qualitative research, the COS and the opinion of the SAG. We asked the SAG to define a clinically meaningful difference for this between two interventions and they recommended that this was 10 dB. We could not find data of direct relevance to this study for children with CP, as all the trials identified have excluded children with CP. Nevertheless, we identified a trial of VTs versus 'no treatment' in children aged 19 months without CP, with a follow-up of 12 months. This is similar to the study we propose (ignoring the absence of CP patients) with our HA group not receiving a medical or surgical treatment which could effect the presence of OME (i.e. they will receive 'no treatment').[166]

A sample size calculation based on data from this study shows that a study with a two-sided significance of 5% and a power of 90% would require 40 patients per intervention group to detect a difference of 10 dB with a standard deviation of 13.8. If we factor in potential loss to follow up of 10% we should aim for a total sample of 90 patients. If we then assume a 2-month run in for each recruiting site, the recruitment of 90 children in eight centres would take 20 months.

However, this sample size is likely to be an underestimate because the limited length of follow-up in the Rovers *et al.* study[166] is likely to underestimate the variability in hearing loss.

Taking these factors into account, the issues that need to be considered in moving forward to a trial are:

1. there is limited data available for an accurate sample size calculation
2. the recruitment rate for any study may be low.

We suggest that additional data, which might strengthen the sample size calculation, could be obtained from a note review of hospital records to extract information for hearing levels in CP children at 2 and 5 years of age.

Concerns about the recruitment rate could be addressed by having an internal pilot in the trial, in order to identify likely recruitment rates and, through a qualitative research component, to identify barriers to recruitment and optimise methods of recruitment. For example, the qualitative component of our study suggested that parents were concerned about safety of their child, they were not in equipoise and they were not clear on the potential harms and benefits of the interventions that might be used. A decision on progression to the main trial could be taken 6 months after recruitment to the pilot has started.

Reflections on this project

We conclude with some reflections on this project. We had originally aimed to complete this project within 12 months and it was costed at a modest level because we underpinned some of the salary costs with resources from the Healing Foundation Cleft and Craniofacial Clinical Research Centre.

Unfortunately, the study over-ran by 16 months. Although the components directed at the qualitative research and health economics were completed inside the proposed time scale, there were major problems with the engagement from the clinical community which was needed to complete both the clinician survey and the COS project (although some centres were fully engaged and provided the information we required

in a short time scale). In effect, we underestimated the time required to gain sufficient input from the clinical community. Despite all our efforts to facilitate this, the engagement was still slow and co-operation was not always forthcoming. We recommend that careful consideration be given to this in future studies that require extensive engagement with the cleft clinical community given how diverse it is.

Conclusion

There is need for further study of the management of OME in children with CP. This research should be a randomised trial based in eight of the UK cleft centres that compares the effectiveness of VTs with HAs. Children will enter the trial when they are 2 years old and followed for 3 years. An initial calculation suggests that the trial should enrol a sample of at least 90 children. The outcomes should be based on the COS that has been developed, with a primary outcome of hearing. However, there is uncertainty about the required sample size and likely recruitment rate for a trial.

As a result, we recommend that additional data should be obtained from a note review of hospital records to inform the sample size calculation.

Concerns about the recruitment rate could be addressed by having an internal pilot in the trial, in order to identify likely recruitment rates and, through a qualitative research component, to identify barriers to recruitment and optimise methods of recruitment. For example, the qualitative component of our study suggested that parents were concerned about the safety of their child, they were not in equipoise and they were not clear on the potential harms and benefits of the interventions that might be used. A decision on progression to the main trial could be taken 6 months after recruitment to the pilot has started.

Acknowledgements

This project was carried out by the Healing Foundation Cleft and Craniofacial Clinical Research Centre at the University of Manchester. The centre is funded by The Healing Foundation supported by Vocational Training Charitable Trust (VTCT).

This study was part-funded by the Healing Foundation supported by VTCT who funded trial staff including the study co-ordinator, information systems developer, study statistician, administrator and supervisory staff.

We acknowledge both the time and the efforts from:

Joyce Russell, Jeanette Mooney and Haydn Bellardie for their support and contribution to the qualitative protocol.

Study Advisory Group members: Debbie Sell (Senior Speech and Language Therapist, Great Ormond Street Hospital, London, UK), Anne Harding-Bell (Senior Speech and Language Therapist, Addenbrookes Hospital, Cambridge, UK), Raouf Chorbachi (Consultant Audiologist, Great Ormond Street Hospital for Children, London, UK), Per Hall (Lead Surgeon and Clinical Director, Plastic Surgeon, Addenbrookes Hospital, Cambridge, UK), Ravi Sharma (Consultant ENT surgeon, Alder Hey Children's Hospital Trust, Liverpool, UK), Nichola Rumsey (Professor of Appearance Research, University of West of England, Bristol, UK) and Alex Bennett (Consultant ENT Surgeon, Royal Hospital for Sick Children, Edinburgh, Scotland).

Study Steering Committee members: Mike Clarke (Professor/Director of Medical Research Council Network of Hubs for Trials Methodology Research), Professor Anne Schilder (National Institute for Health Research Professor Director), Professor Andrea Manca (Professor of Health Economics, Centre for Health Economics) and Rosanna Preston (Chief Executive of the Cleft Lip and Palate Association).

Duncan Appelbe and Melanie Harper-Jones (Clinical Trials Research Centre, University of Liverpool) for their contribution to the design of the online system for the Delphi survey of health professionals and online survey of parents and children with CP, and Patricia Barnby (School of Dentistry/Healing Foundation Cleft and Craniofacial Clinical Research Centre, University of Manchester) for her contribution to the design of the online system for the Delphi survey of health professionals. Joyce Russell, Jeanette Mooney and Haydn Bellardie for their support and contribution to the qualitative protocol. Tri Tat (School of Dentistry/Healing Foundation Cleft and Craniofacial Clinical Research Centre, University of Manchester) for comments on an earlier version of the Delphi survey for health professional's protocol. Claire Bennett (School of Dentistry/Healing Foundation Cleft and Craniofacial Clinical Research Centre, University of Manchester) for administrative support provided to the study and assisting with the formatting of the final report.

Contributions of authors

Iain Bruce contributed to the design of the study, acquisition, analysis and interpretation of the data (clinician survey, COS, health economics, final recommendations), drafted and revised the manuscript.

Nicola Harman contributed to the design of the study, acquisition, analysis and interpretation of the data (clinician survey, qualitative study, COS, final recommendations), drafted and revised the manuscript.

Paula Williamson contributed to the design of the study, acquisition, analysis and interpretation of the data (clinician survey, COS, final recommendations), drafted and revised the manuscript.

Stephanie Tierney contributed to the design of the study, acquisition, analysis and interpretation of the data (qualitative study, COS), drafted and revised the manuscript.

Peter Callery contributed to the design of the study, acquisition, analysis and interpretation of the data (qualitative study, COS, final recommendations), drafted and revised the manuscript.

Syed Mohuiddin contributed to the design of the study, acquisition, analysis and interpretation of the data (health economics), drafted and revised the manuscript.

Katherine Payne contributed to the design of the study, acquisition, analysis and interpretation of the data (health economics), drafted and revised the manuscript.

Elisabeth Fenwick contributed to the design of the health economics study, acquisition, analysis and interpretation of the data, and drafted and revised the health economics section of the manuscript.

Jamie Kirkham had input into the design and data analysis of the Delphi.

Kevin O'Brien contributed to the design of the study, acquisition, analysis and interpretation of the data (clinician survey, qualitative study, COS, final recommendations), drafted and revised the manuscript. He is the guarantor.

Data sharing statement

All data can be obtained from the corresponding author.

References

1. Ponduri S, Bradley R, Ellis PE, Brookes ST, Sandy JR, Ness AR. The management of otitis media with early routine insertion of grommets in children with cleft palate – a systematic review. *Cleft Palate Craniofac J* 2009;**46**:30–8. http://dx.doi.org/10.1597/07-219.1
2. Flynn T, Möller C, Jönsson R, Lohmander A. The high prevalence of otitis media with effusion in children with cleft lip and palate as compared to children without clefts. *Int J Pediatr Otorhinolaryngol* 2009;**73**:1441–6. http://dx.doi.org/10.1016/j.ijporl.2009.07.015
3. Goldman JL, Martinez SA, Ganzel TM. Eustachian tube dysfunction and its sequelae in patients with cleft palate. *South Med J* 1993;**86**:1236–7. http://dx.doi.org/10.1097/00007611-199311000-00010
4. Maheshwar AA, Milling MAP, Kumar M, Clayton MI, Thomas A. Use of hearing aids in the management of children with cleft palate. *Int J Pediatr Otorhinolaryngol* 2002;**66**:55–62. http://dx.doi.org/10.1016/S0165-5876(02)00206-9
5. Civelek B, Celebioglu S, Sagit M, Akin I. Ventilation tubes in secretory otitis media associated with cleft palate: a retrospective analysis. *Turk J Med Sci* 2007;**37**:223–6.
6. Flynn T, Moller C, Lohmander A, Magnusson L. Hearing and otitis media with effusion in young adults with cleft lip and palate. *Acta Otolaryngol* 2012;**132**:959–66. http://dx.doi.org/10.3109/00016489.2012.669497
7. Handzic-Cuk J, Cuk V, Gluhinic M, Risavi R, Stajner-Katusic S. Tympanometric findings in cleft palate patients: influence of age and cleft type. *J Laryngol Otol* 2001;**115**:91–6. http://dx.doi.org/10.1258/0022215011907668
8. Moller P. Long-term otologic features of cleft palate patients. *Arch Otolaryngol* 1975;**101**:605–7. http://dx.doi.org/10.1001/archotol.1975.00780390019005
9. Sheahan P, Blayney AW. Cleft palate and otitis media with effusion: a review. *Rev Laryngol Otol Rhinol (Bord)* 2003;**124**:171–7.
10. Harman NL, Bruce IA, Callery P, Tierney S, Sharif MO, O'Brien K, *et al.* MOMENT – management of otitis media with effusion in cleft palate: protocol for a systematic review of the literature and identification of a core outcome set using a Delphi survey. *Trials* 2013;**14**:70. http://dx.doi.org/10.1186/1745-6215-14-70
11. National Institute for Health and Care Excellence (NICE). *Surgical Management of Otitis Media With Effusion in Children*. Clinical guidelines, CG60. London: NICE; 2008.
12. McCulloch P, Taylor I, Sasako M, Lovett B, Griffin D. Randomised trials in surgery: problems and possible solutions. *BMJ* 2002;**324**:1448–51. http://dx.doi.org/10.1136/bmj.324.7351.1448
13. Nelson P, Glenny AM, Kirk S, Caress AL. Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. *Child Care Health Dev* 2012;**38**:6–20. http://dx.doi.org/10.1111/j.1365-2214.2011.01244.x
14. Sharif MO, Callery P, Tierney S. The perspectives of children and young people living with cleft lip and palate: a review of qualitative literature. *Cleft Palate Craniofac J* 2013;**50**:297–304. http://dx.doi.org/10.1597/12-054
15. Murphy E, Dingwall R, Greatbatch D, Parker S, Watson P. Qualitative research in health technology assessment: a review of the literature. *Health Technol Assess* 1998;**2**(16).

16. Spencer L, Ritchie J, O'Connor W. Analysis: Practices, Principles and Processes. In Ritchie J, Lewis J, editors. *Qualitative Research Practice: A Guide for Social Science Students and Researchers*. Thousand Oaks, CA: Sage Publications; 2003. pp. xv, 336.

17. National Collaborating Centre for Women's and Children's Health. *Surgical Management of Otitis Media with Effusion in Children*. London: National Collaborating Centre for Women's and Children's Health; 2008.

18. Ritchie J, Lewis J, Elam G. Designing and Selecting Samples. In Ritchie J, Lewis J, editors. *Qualitative Research Practice*. London: Sage Publications; 2003. pp. 77–108.

19. Shilling V, Williamson P, Hickey H, Sowden E, Smyth R, Young B. Processes in recruitment to randomised controlled trials of medicines for children (RECRUIT): a qualitative study. *Health Technol Assess* 2011;**15**(15). http://dx.doi.org/10.3310/hta15150

20. Irwin LG, Johnson J. Interviewing young children: explicating our practices and dilemmas. *Qual Health Res* 2005;**15**:821–31. http://dx.doi.org/10.1177/1049732304273862

21. O'Kane C. The Development of Participatory Techniques: Facilitating Children's Views About Decisions Which Affect Them. In Christensen PM, James A, editors. *Research with Children: Perspectives and Practices*. London: RoutledgeFalmer; 2000. pp. 136–59.

22. Darbyshire P, Macdougall C, Schiller W. Multiple methods in qualitative research with children: more insight or just more? *Qual Res* 2005;**5**:417–36. http://dx.doi.org/10.1177/1468794105056921

23. Hill M. Participatory research with children. *Child Fam Soc Work* 1997;**2**:171–83. http://dx.doi.org/10.1046/j.1365-2206.1997.00056.x

24. Ritchie J, Lewis J. *Qualitative Research Practice: A Guide for Social Science Students and Researchers*. Thousand Oaks, CA: Sage Publications; 2003. pp. xv, 336.

25. Srivastava A, Thomson SB. Framework analysis: a qualitative methodology for applied policy research. *J Admin Govern* 2009;**4**:72–9.

26. Ward DJ, Furber C, Tierney S, Swallow V. Using framework analysis in nursing research: a worked example. *J Adv Nurs* 2013;**69**:2423–31. http://dx.doi.org/10.1111/jan.12127

27. Elliott R, Fischer CT, Rennie DL. Evolving guidelines for publication of qualitative research studies in psychology and related fields. *Br J Clin Psychol* 1999;**38**:215–29. http://dx.doi.org/10.1348/014466599162782

28. Long T, Johnson M. Rigour, reliability and validity in qualitative research . . . including commentary by Burnard P with author response. *Clin Eff Nurs* 2000;**4**:30–7. http://dx.doi.org/10.1054/cein.2000.0106

29. Corbett F, Oldham J, Lilford R. Offering patients entry in clinical trials: preliminary study of the views of prospective participants. *J Med Ethics* 1996;**22**:227–31. http://dx.doi.org/10.1136/jme.22.4.227

30. McDonald AM, Knight RC, Campbell MK, Entwistle VA, Grant AM, Cook JA, *et al.* What influences recruitment to randomised controlled trials? A review of trials funded by two UK funding agencies. *Trials* 2006;**7**:9. http://dx.doi.org/10.1186/1745-6215-7-9

31. Medical Research Council. *Clinical Trials for Tomorrow*. London: Medical Research Council; 2003.

32. Edwards SJ, Lilford RJ, Braunholtz DA, Jackson JC, Hewison J, Thornton J. Ethical issues in the design and conduct of randomised controlled trials. *Health Technol Assess* 1998;**2**(15).

33. Featherstone K, Donovan JL. 'Why don't they just tell me straight, why allocate it?' The struggle to make sense of participating in a randomised controlled trial. *Soc Sci Med* 2002;**55**:709–19. http://dx.doi.org/10.1016/S0277-9536(01)00197-6

34. Daugherty C, Ratain MJ, Grochowski E, Stocking C, Kodish E, Mick R, *et al.* Perceptions of cancer patients and their physicians involved in phase I trials. *J Clin Oncol* 1995;**13**:1062–72.

35. Shilling V, Young B. How do parents experience being asked to enter a child in a randomised controlled trial? *BMC Med Ethics* 2009;**10**:1. http://dx.doi.org/10.1186/1472-6939-10-1

36. Zupancic JA, Gillie P, Streiner DL, Watts JL, Schmidt B. Determinants of parental authorization for involvement of newborn infants in clinical trials. *Pediatrics* 1997;**99**:E6. http://dx.doi.org/10.1542/peds.99.1.e6

37. Sammons H, Atkinson M, Choonara I, Stephenson T. What motivates British parents to consent for research? A questionnaire study. *BMC Pediatr* 2007;**7**:12. http://dx.doi.org/10.1186/1471-2431-7-12

38. Glogowska M, Roulstone S, Enderby P, Peters T, Campbell R. Who's afraid of the randomised controlled trial? Parents' views of an SLT research study. *Int J Lang Commun Disord* 2001;**36**:499–504. http://dx.doi.org/10.3109/13682820109177936

39. Kass NE, Sugarman J, Faden R, Schoch-Spana M. Trust, the fragile foundation of contemporary biomedical research. *Hastings Cent Rep* 1996;**26**:25–9. http://dx.doi.org/10.2307/3528467

40. Eder ML, Yamokoski AD, Wittmann PW, Kodish ED. Improving informed consent: suggestions from parents of children with leukemia. *Pediatrics* 2007;**119**:e849–59. http://dx.doi.org/10.1542/peds.2006-2208

41. Postlethwaite RJ, Reynolds JM, Wood AJ, Evans JH, Lewis MA, Eminson DM. Recruiting patients to clinical trials: lessons from studies of growth hormone treatment in renal failure. *Arch Dis Child* 1995;**73**:30–5. http://dx.doi.org/10.1136/adc.73.1.30

42. Sweetow RW, Rosbe KW, Philliposian C, Miller MT. Considerations for cochlear implantation of children with sudden, fluctuating hearing loss. *J Am Acad Audiol* 2005;**16**:770–80. http://dx.doi.org/10.3766/jaaa.16.10.2

43. Paediatric European Network for Treatment of AIDS. Parents' attitudes to their HIV-infected children being enrolled into a placebo-controlled trial: the PENTA 1 trial. *HIV Med* 1999;**1**:25–31. http://dx.doi.org/10.1046/j.1468-1293.1999.00005.x

44. Singhal N, Oberle K, Burgess E, Huber-Okrainec J. Parents' perceptions of research with newborns. *J Perinatol* 2002;**22**:57–63. http://dx.doi.org/10.1038/sj.jp.7210608

45. *Oxford English Dictionary*. URL: www.oed.com (accessed April 2014).

46. Wells GA. Patient-driven outcomes in rheumatoid arthritis. *J Rheumatol Suppl* 2009;**82**:33–8. http://dx.doi.org/10.3899/jrheum.090129

47. Fitzpatrick E, Graham ID, Durieux-Smith A, Angus D, Coyle D. Parents' perspectives on the impact of the early diagnosis of childhood hearing loss. *Int J Audiol* 2007;**46**:97–106. http://dx.doi.org/10.1080/14992020600977770

48. Sinha I, Jones L, Smyth RL, Williamson PR. A systematic review of studies that aim to determine which outcomes to measure in clinical trials in children. *PLOS Med* 2008;**5**:e96. http://dx.doi.org/10.1371/journal.pmed.0050096

49. Clarke M. Standardising outcomes for clinical trials and systematic reviews. *Trials* 2007;**8**:39. http://dx.doi.org/10.1186/1745-6215-8-39

50. Cooney R, Warren B, Altman D, Abreu M, Travis S. Outcome measurement in clinical trials for ulcerative colitis: towards standardisation. *Trials* 2007;**8**:17. http://dx.doi.org/10.1186/1745-6215-8-17

51. Chan A-W, Altman DG. Identifying outcome reporting bias in randomised trials on PubMed: review of publications and survey of authors. *BMJ* 2005;**330**:753. http://dx.doi.org/10.1136/bmj.38356.424606.8F

52. Tugwell P, Boers M, Brooks P, Simon L, Strand V, Idzerda L. OMERACT: an international initiative to improve outcome measurement in rheumatology. *Trials* 2007;**8**:38. http://dx.doi.org/10.1186/1745-6215-8-38

53. Lux AL, Osborne JP. A proposal for case definitions and outcome measures in studies of infantile spasms and West syndrome: consensus statement of the West Delphi group. *Epilepsia* 2004;**45**:1416–28. http://dx.doi.org/10.1111/j.0013-9580.2004.02404.x

54. Ruperto N, Ravelli A, Murray KJ, Lovell DJ, Andersson-Gare B, Feldman BM, *et al.* Preliminary core sets of measures for disease activity and damage assessment in juvenile systemic lupus erythematosus and juvenile dermatomyositis. *Rheumatology* 2003;**42**:1452–9. http://dx.doi.org/10.1093/rheumatology/keg403

55. Chen JL, Messner AH, Curtin G. Newborn hearing screening in infants with cleft palates. *Otol Neurotol* 2008;**29**:812–15. http://dx.doi.org/10.1097/MAO.0b013e318180a4e0

56. Simpson SA, Thomas CL, van der Linden MK, Macmillan H, van der Wouden JC, Butler C. Identification of children in the first four years of life for early treatment for otitis media with effusion. *Cochrane Database Syst Rev* 2007;**1**:CD004163. http://dx.doi.org/10.1002/14651858.cd004163.pub2

57. Thomas CL, Simpson S, Butler CC, van der Voort JH. Oral or topical nasal steroids for hearing loss associated with otitis media with effusion in children. *Cochrane Database Syst Rev* 2006;**3**:CD001935. http://dx.doi.org/10.1002/14651858.cd001935.pub2

58. van den Aardweg MT, Schilder AG, Herkert E, Boonacker CW, Rovers MM. Adenoidectomy for otitis media in children. *Cochrane Database Syst Rev* 2010;**1**:CD007810. http://dx.doi.org/10.1002/14651858.cd007810.pub2

59. Griffin GH, Flynn C, Bailey RE, Schultz JK. Antihistamines and/or decongestants for otitis media with effusion (OME) in children. *Cochrane Database Syst Rev* 2006;**4**:CD003423. http://dx.doi.org/10.1002/14651858.cd003423.pub2

60. Browning GG, Rovers MM, Williamson I, Lous J, Burton MJ. Grommets (ventilation tubes) for hearing loss associated with otitis media with effusion in children. *Cochrane Database Syst Rev* 2010;**10**:CD001801. http://dx.doi.org/10.1002/14651858.cd001801.pub3

61. Perera R, Haynes J, Glasziou P, Heneghan CJ. Autoinflation for hearing loss associated with otitis media with effusion. *Cochrane Database Syst Rev* 2006;**4**:CD006285. http://dx.doi.org/10.1002/14651858.cd006285

62. Møller P. Seletive use of ventilating tubes in the treatment of secretory otitis media and retractions of the ear drum. *Acta Otolaryngol* 1982;**93**:158–60. http://dx.doi.org/10.3109/00016488209108504

63. Potsic WP, Cohen M, Randall P, Winchester R. Retrospective study of hearing impairment in 3 groups of cleft-palate patients. *Cleft Palate J* 1979;**16**:56–8.

64. Paradise JL, Bluestone CD. Early treatment of the universal otitis media of infants with cleft palate. *Pediatrics* 1974;**53**:48–54.

65. Møller P. Hearing, middle-ear pressure and otopathology in a cleft-palate population. *Acta Otolaryngol* 1981;**92**:521–8. http://dx.doi.org/10.3109/00016488109133291

66. Smith TL, DiRuggiero DC, Jones KR. Recovery of eustachian tube function and hearing outcome in patients with cleft palate. *Otolaryngol Head Neck Surg* 1994;**111**:423–9.

67. Hormann K, Roehrs M. Middle-ear findings in young cleft lip and palate children. Comparison of two treatment clinics. *Dtsche Z Mund Kiefer Gesichtschir* 1991;**15**:149–52.

68. Broen PA, Moller KT, Carlstrom J, Doyle SS, Devers M, Keenan KM. Comparison of the hearing histories of children with and without cleft palate. *Cleft Palate Craniofac J* 1996;**33**:127–33. http://dx.doi.org/10.1597/1545-1569(1996)033<0127:COTHHO>2.3.CO;2

69. Frable MA, Brandon GT, Theogaraj SD. Velar closure and ear tubings as a primary procedure in the repair of cleft palates. *Laryngoscope* 1985;**95**:1044–6. http://dx.doi.org/10.1288/00005537-198509000-00004

70. Sheahan P, Blayney AW, Sheahan JN, Earley MJ. Sequelae of otitis media with effusion among children with cleft lip and/or cleft palate. *Clin Otolaryngol* 2002;**27**:494–500. http://dx.doi.org/10.1046/j.1365-2273.2002.00607.x

71. Hubbard TW, Paradise JL, McWilliams BJ, Elster BA, Taylor FH. Consequences of unremitting middle-ear disease in early life. Otologic, audiologic, and developmental findings in children with cleft palate. *N Engl J Med* 1985;**312**:1529–34. http://dx.doi.org/10.1056/NEJM198506133122401

72. Gordon ASD, Jeanlouis F, Morton RP. Late ear sequelae in cleft-palate patients. *Int J Pediatr Otorhinolaryngol* 1988;**15**:149–56. http://dx.doi.org/10.1016/0165-5876(88)90066-3

73. Robson AK, Blanshard JD, Jones K, Albery EH, Smith IM, Maw AR. A conservative approach to the management of otitis-media with effusion in cleft-palate children. *J Laryngol Otol* 1992;**106**:788–92. http://dx.doi.org/10.1017/S0022215100120894

74. Greig AV, Papesch ME, Rowsell AR. Parental perceptions of grommet insertion in children with cleft palate. *J Laryngol Otol* 1999;**113**:1068–71.

75. Shaw R, Richardson D, McMahon S. Conservative management of otitis media in cleft palate. *J Craniomaxillofac Surg* 2003;**31**:316–20. http://dx.doi.org/10.1016/S1010-5182(03)00074-X

76. Freeland AP, Evans DM. Middle-ear disease in the cleft-palate infant – its effect on speech and language-development. *Br J Plast Surg* 1981;**34**:142–3. http://dx.doi.org/10.1016/S0007-1226(81)80081-1

77. Zheng Q, Xu H, He Y. Effects of tympanotomy and pressure equilibrium tube insertion during palatoplasty on prognoses of otitis media with effusion. *Hua Xi Kou Qiang Yi Xue Za Zhi* 2003;**21**:28–30.

78. Liu L, Sun Y-G, Ma L, Zhao W, Wu R. Effect of ventilation tube insertion on otitis media with effusion in cleft palate children. *Zhonghua er bi yan hou ke za zhi* 2004;**39**:216–18.

79. Tanpowpong K, Saisukul I, Kittimanont H, Rattanasiri S. Outcome of myringotomy with ventilation tube for otitis media with effusion in Thai children: Ramathibodi experiences. *J Med Assoc Thai* 2007;**90**:1866–71.

80. Phua YS, Salkeld LJ, de Chalain TM. Middle ear disease in children with cleft palate: protocols for management. *Int J Pediatr Otorhinolaryngol* 2009;**73**:307–13. http://dx.doi.org/10.1016/j.ijporl.2008.10.026

81. Reiter R, Haase S, Brosch S. Repaired cleft palate and ventilation tubes and their associations with cholesteatoma in children and adults. *Cleft Palate Craniofac J* 2009;**46**:598–602. http://dx.doi.org/10.1597/08-166.1

82. Szabo C, Langevin K, Schoem S, Mabry K. Treatment of persistent middle ear effusion in cleft palate patients. *Int J Pediatr Otorhinolaryngol* 2010;**74**:874–7. http://dx.doi.org/10.1016/j.ijporl.2010.04.016

83. Hornigold R, Morley A, Glore RJ, Boorman J, Sergeant R. The long-term effect of unilateral t-tube insertion in patients undergoing cleft palate repair: 20-year follow-up of a randomised controlled trial. *Clin Otolaryngol* 2008;**33**:265–8. http://dx.doi.org/10.1111/j.1749-4486.2008.01670.x

84. Merrick GD, Kunjur J, Watts R, Markus AF. The effect of early insertion of grommets on the development of speech in children with cleft palates. *Br J Oral Maxillofac Surg* 2007;**45**:527–33. http://dx.doi.org/10.1016/j.bjoms.2007.02.003

85. Curtin G, Messner AH, Chang KW. Otorrhea in infants with tympanostomy tubes before and after surgical repair of a cleft palate. *Arch Otolaryngol Head Neck Surg* 2009;**135**:748–51. http://dx.doi.org/10.1001/archoto.2009.106

86. Mandel EM, Casselbrant ML, Rockette HE, Fireman P, Kurs-Lasky M, Bluestone CD. Systemic steroid for chronic otitis media with effusion in children. *Pediatrics* 2002;**110**:1071–80. http://dx.doi.org/10.1542/peds.110.6.1071

87. Casselbrant ML, Mandel EM, Rockette HE, Kurs-Lasky M, Fall PA, Bluestone CD. Adenoidectomy for otitis media with effusion in 2–3-year-old children. *Int J Pediatr Otorhinolaryngol* 2009;**73**:1718–24. http://dx.doi.org/10.1016/j.ijporl.2009.09.007

88. Koivunen P, Uhari M, Luotonen J, Kristo A, Raski R, Pokka T, *et al.* Adenoidectomy versus chemoprophylaxis and placebo for recurrent acute otitis media in children aged under 2 years: randomised controlled trial. *BMJ* 2004;**328**:487–90. http://dx.doi.org/10.1136/bmj.37972.678345.0D

89. Mattila PS, Joki-Erkkila VP, Kilpi T, Jokinen J, Herva E, Puhakka H. Prevention of otitis media by adenoidectomy in children younger than 2 years. *Arch Otolaryngol Head Neck Surg* 2003;**129**:163–8. http://dx.doi.org/10.1001/archotol.129.2.163

90. Paradise JL, Bluestone CD, Rogers KD, Taylor FH, Colborn DK, Bachman RZ, *et al.* Efficacy of adenoidectomy for recurrent otitis-media in children previously treated with tympanostomy-tube placement – results of parallel randomized and nonrandomized trials. *JAMA* 1990;**263**:2066–73. http://dx.doi.org/10.1001/jama.1990.03440150074029

91. Paradise JL, Bluestone CD, Colborn DK, Bernard BS, Smith CG, Rockette HE, *et al.* Adenoidectomy and adenotonsillectomy for recurrent acute otitis media – parallel randomized clinical trials in children not previously treated with tympanostomy tubes. *JAMA* 1999;**282**:945–53. http://dx.doi.org/10.1001/jama.282.10.945

92. Rynneldagoo B, Ahlbom A, Schiratzki H. Effects of adenoidectomy – controlled 2-year follow-up. *Ann Otol Rhinol Laryngol* 1978;**87**:272–8. http://dx.doi.org/10.1177/000348947808700223

93. Gates GA, Avery CA, Cooper JC Jr, Prihoda TJ. Effectiveness of adenoidectomy and tympanostomy tubes in the treatment of chronic otitis media with effusion. *New Engl J Med* 1987;**317**:1444–51. http://dx.doi.org/10.1056/NEJM198712033172305

94. Hammaren-Malmi S, Saxen H, Tarkkanen J, Mattila PS. Adenoidectomy does not significantly reduce the incidence of otitis media in conjunction with the insertion of tympanostomy tubes in children who are younger than 4 years: a randomized trial. *Pediatrics* 2005;**116**:185–9. http://dx.doi.org/10.1542/peds.2004-2253

95. Roydhouse N. Adenoidectomy for otitis-media with mucoid effusion. *Ann Otol Rhinol Laryngol* 1980;**89**:312–15.

96. Black NA, Sanderson CFB, Freeland AP, Vessey MP. A randomized controlled trial of surgery for glue ear. *BMJ* 1990;**300**:1551–6. http://dx.doi.org/10.1136/bmj.300.6739.1551

97. Dempster JH, Browning GG, Gatehouse SG. A randomized study of the surgical-management of children with persistent otitis-media with effusion associated with a hearing impairment. *J Laryngol Otol* 1993;**107**:284–9. http://dx.doi.org/10.1017/S0022215100122844

98. Maw R, Wilks J, Harvey I, Peters TJ, Golding J. Early surgery compared with watchful waiting for glue ear and effect on language development in preschool children: a randomised trial. *Lancet* 1999;**353**:960–3. [Erratum in *Lancet* 1999;**354**:1392.] http://dx.doi.org/10.1016/S0140-6736(98)05295-7

99. Rach GH, Zielhuis GA, Vanbaarle PW, Vandenbroek P. The effect of treatment with ventilating tubes on language-development in preschool-children with otitis-media with effusion. *Clin Otolaryngol* 1991;**16**:128–32. http://dx.doi.org/10.1111/j.1365-2273.1991.tb01960.x

100. Rovers MM, Straatman H, Ingels K, van der Wilt GJ, van den Broek P, Zielhuis GA. The effect of ventilation tubes on language development in infants with otitis media with effusion: a randomized trial. *Pediatrics* 2000;**106**:E42. http://dx.doi.org/10.1542/peds.106.3.e42

101. Johnston LC, Feldman HM, Paradise JL, Bernard BS, Colborn DK, Casselbrant ML, *et al.* Tympanic membrane abnormalities and hearing levels at the ages of 5 and 6 years in relation to persistent otitis media and tympanostomy tube insertion in the first 3 years of life: a prospective study incorporating a randomized clinical trial. *Pediatrics* 2004;**114**:E58–67. http://dx.doi.org/10.1542/peds.114.1.e58

102. Paradise JL, Feldman HM, Campbell TF, Dollaghan CA, Colborn DK, Bernard BS, *et al.* Effect of early or delayed insertion of tympanostomy tubes for persistent otitis media on developmental outcomes at the age of three years. *N Engl J Med* 2001;**344**:1179–87. http://dx.doi.org/10.1056/NEJM200104193441601

103. Maw AR. Chronic otitis-media with effusion (glue ear) and adenotonsillectomy – prospective randomized controlled-study. *BMJ* 1983;**287**:1586–8. http://dx.doi.org/10.1136/bmj.287.6405.1586

104. Zielhuis GA, Rach GH, Vandenbroek P. Screening for otitis-media with effusion in preschool-children. *Lancet* 1989;**1**:311–14. http://dx.doi.org/10.1016/S0140-6736(89)91317-2

105. Fiellau-Nikolajsen M, Falbe-Hansen J, Knudstrup P. Adenoidectomy for middle-ear disorders – a randomized controlled trial. *Clin Otolaryngol Allied Sci* 1980;**5**:323–7. http://dx.doi.org/10.1111/j.1365-2273.1980.tb00898.x

106. Nguyen LHP, Manoukian JJ, Yoskovitch A, Al-Sebeih KH. Adenoidectomy: selection criteria for surgical cases of otitis media. *Laryngoscope* 2004;**114**:863–6. http://dx.doi.org/10.1097/00005537-200405000-00014

107. Paradise JL, Feldman HM, Campbell TF, Dollaghan CA, Rockette HE, Pitcairn DL, *et al.* Tympanostomy tubes and developmental outcomes at 9 to 11 years of age. *N Engl J Med* 2007;**356**:248–61. http://dx.doi.org/10.1056/NEJMoa062980

108. Paradise JL, Campbell TF, Dollaghan CA, Feldman HM, Bernard BS, Colborn DK, *et al.* Developmental outcomes after early or delayed insertion of tympanostomy tubes. *N Engl J Med* 2005;**353**:576–86. http://dx.doi.org/10.1056/NEJMoa050406

109. Paradise JL, Dollaghan CA, Campbell TF, Feldman HM, Bernard BS, Colborn DK, *et al.* Otitis media and tympanostomy tube insertion during the first three years of life: developmental outcomes at the age of four years. *Pediatrics* 2003;**112**:265–77. http://dx.doi.org/10.1542/peds.112.2.265

110. Paradise JL. Mix of middle ear effusions in infants with cleft palate. *Ann Otol Rhinol Laryngol Suppl* 1976;**85**:285–8.

111. National Institute of Adult Continuing Education. *SMOG Calculator*. 2014. URL: www.niace.org.uk/misc/SMOG-calculator/smogcalc.php (accessed 4 April 2014).

112. Guyatt GH, Oxman AD, Kunz R, Atkins D, Brozek J, Vist G, *et al.* GRADE guidelines: 2. Framing the question and deciding on important outcomes. *J Clin Epidemiol* 2011;**64**:395–400. http://dx.doi.org/10.1016/j.jclinepi.2010.09.012

113. Grant HR, Quiney RE, Mercer DM, Lodge S. Cleft-palate and glue ear. *Arch Dis Child* 1988;**63**:176–9. http://dx.doi.org/10.1136/adc.63.2.176

114. Britten N, Ukoumunne O. The influence of patients' hopes of receiving a prescription on doctors' perceptions and the decision to prescribe: a questionnaire survey. *BMJ* 1997;**315**:1506–10. http://dx.doi.org/10.1136/bmj.315.7121.1506

115. Edwards PJ, Roberts I, Clarke MJ, Diguiseppi C, Wentz R, Kwan I, *et al.* Methods to increase response to postal and electronic questionnaires. *Cochrane Database Syst Rev* 2009;**3**:MR000008. http://dx.doi.org/10.1002/14651858.mr000008.pub4

116. John A, Sell D, Sweeney T, Harding-Bell A, Williams A. The cleft audit protocol for speech-augmented: a validated and reliable measure for auditing cleft speech. *Cleft Palate Craniofac J* 2006;**43**:272–88. http://dx.doi.org/10.1597/04-141R.1

117. Shaw WC, Semb G, Nelson P, Brattstrom V, Molsted K, Prahl-Andersen B, *et al.* The Eurocleft Project 1996–2000: overview. *J Cranio-MaxilloFac Surg* 2001;**29**:131–40. http://dx.doi.org/10.1054/jcms.2001.0217

118. World Health Organization (WHO). *Global Strategies Towards Reducing the Health-Care Burden of Craniofacial Anomalies*. Report of WHO meetings on International Collaborative Research on Craniofacial Anomalies. Geneva: WHO; 2002.

119. Offcom. *The Communications Market Report: United Kingdom*. 2011. URL: http://stakeholders.ofcom.org.uk/market-data-research/market-data/communications-market-reports/cmr11/uk/ (accessed April 2014).

120. Khanna R, Lakhanpaul M, Bull PD, Group GD. Surgical management of otitis media with effusion in children: summary of NICE guidance. *Clin Otolaryngol* 2008;**33**:600–5. http://dx.doi.org/10.1111/j.1749-4486.2008.01844.x

121. Berman S, Roark R, Luckey D. Theoretical cost effectiveness of management options for children with persisting middle ear effusions. *Pediatrics* 1994;**93**:353–63.

122. Hartman M, Rovers MM, Ingels K, Zielhuis GA, Severens JL, van der Wilt GJ. Economic evaluation of ventilation tubes in otitis media with effusion. *Arch Otolaryngol Head Neck Surg* 2001;**127**:1471–6. http://dx.doi.org/10.1001/archotol.127.12.1471

123. Mui S, Rasgon BM, Hilsinger RL Jr, Lewis B, Lactao G. Tympanostomy tubes for otitis media: quality-of-life improvement for children and parents. *Ear Nose Throat J* 2005;**84**:418, 420–2, 424.

124. Sheahan P, Miller I, Sheahan JN, Earley MJ, Blayney AW. Incidence and outcome of middle ear disease in cleft lip and/or cleft palate. *Int J Pediatr Otorhinolaryngol* 2003;**67**:785–93. http://dx.doi.org/10.1016/S0165-5876(03)00098-3

125. Gould HJ. Hearing loss and cleft palate: the perspective of time. *Cleft Palate J* 1990;**27**:36–9. http://dx.doi.org/10.1597/1545-1569(1990)027<0036:HLACPT>2.3.CO;2

126. National Institute of Health and Care Excellence (NICE). *Guide to the Methods of Technology Appraisal.* London: NICE; 2004.

127. Barton P, Bryan S, Robinson S. Modelling in the economic evaluation of health care: selecting the appropriate approach. *J Health Serv Res Policy* 2004;**9**:110–18. http://dx.doi.org/10.1258/135581904322987535

128. Kwan WMY, Abdullah VJ, Liu K, van Hasselt CA, Tong MCF. Otitis media with effusion and hearing loss in chinese children with cleft lip and palate. *Cleft Palate Craniofac J* 2011;**48**:684–9. http://dx.doi.org/10.1597/10-006

129. Kay DJ, Nelson M, Rosenfeld RM. Meta-analysis of tympanostomy tube sequelae. *Otolaryngol Head Neck Surg* 2001;**124**:374–80. http://dx.doi.org/10.1067/mhn.2001.113941

130. Fior R, Veljak C. Late results and complications of tympanostomy tube insertion for prophylaxis of recurrent purulent otitis media in pediatric age. *Int J Pediatr Otorhinolaryngol* 1984;**8**:139–46. http://dx.doi.org/10.1016/S0165-5876(84)80062-2

131. Vlastarakos PV, Nikolopoulos TP, Korres S, Tavoulari E, Tzagaroulakis A, Ferekidis E. Grommets in otitis media with effusion: the most frequent operation in children. But is it associated with significant complications? *Eur J Pediatr* 2007;**166**:385–91. http://dx.doi.org/10.1007/s00431-006-0367-x

132. Gibb AG. President's address. Tympanosclerosis. *Proc R Soc Med* 1976;**69**:155–62.

133. Williamson I. Otitis media with effusion in children. *BMJ Clin Evid* 2011;**2011**:0502.

134. Lous J, Burton MJ, Felding JU, Ovesen T, Rovers MM, Williamson I. Grommets (ventilation tubes) for hearing loss associated with otitis media with effusion in children. *Cochrane Database Syst Rev* 2005;**1**:CD001801. http://dx.doi.org/10.1002/14651858.cd001801.pub2

135. NHS Choices. *Complications of Glue Ear*. 2013. URL: www.nhs.uk/Conditions/Glueear/Pages/Complications.aspx (accessed 10 July 2013).

136. Hoffmann KK, Thompson GK, Burke BL, Derkay CS. Anesthetic complications of tympanostomy tube placement in children. *Arch Otolaryngol Head Neck Surg* 2002;**128**:1040–3. http://dx.doi.org/10.1001/archotol.128.9.1040

137. Oxford Radcliffe Hospitals NHS Trust. *Grommets Surgery for Glue Ear – Information for Patients*. Oxford: Oxford Radcliffe Hospitals NHS Trust; 2012. URL: www.ouh.nhs.uk/patient-guide/leaflets/files%5C101018grommets.pdf (accessed 10 April 2013).

138. Gani B, Kinshuck AJ, Sharma R. A review of hearing loss in cleft palate patients. *Int J Otolaryngol* 2012;**2012**:548698. http://dx.doi.org/10.1155/2012/548698

139. Rovers MM, Schilder AGM, Zielhuis GA, Rosenfeld RM. Otitis media. *Lancet* 2004;**363**:465–73. http://dx.doi.org/10.1016/S0140-6736(04)15495-0

140. Rosenfeld RM, Kay D. Natural history of untreated otitis media. *Laryngoscope* 2003;**113**:1645–57. http://dx.doi.org/10.1097/00005537-200310000-00004

141. Rovers MM, Glasziou P, Appelman CL, Burke P, McCormick DP, Damoiseaux RA, *et al.* Antibiotics for acute otitis media: a meta-analysis with individual patient data. *Lancet* 2006;**368**:1429–35. http://dx.doi.org/10.1016/S0140-6736(06)69606-2

142. Gunasekera H, O'Connor TE, Vijayasekaran S, Del Mar CB. Primary care management of otitis media among Australian children. *Med J Aust* 2009;**191**:S55–9.

143. Russell C, Black O, Dutt D, Ray A, Devlin M, Wynne D. Are ventilation tubes (grommets) in cleft children truly associated with increased complication rates? Results of a nested case control study of cleft and non-cleft children. *Br J Oral Maxillofac Surg* 2012;**50**:S2–3. http://dx.doi.org/10.1016/j.bjoms.2012.04.151

144. Maw R, Bawden R. Spontaneous resolution of severe chronic glue ear in children and the effect of adenoidectomy, tonsillectomy, and insertion of ventilation tubes (grommets). *BMJ* 1993;**306**:756–60. http://dx.doi.org/10.1136/bmj.306.6880.756

145. Kubba H. *Quality of Life Assessment in Paediatric Otolaryngology*. MD thesis. Glasgow: University of Glasgow; 2004.

146. Department of Health. *NHS Reference Costs 2010–11*. URL: www.gov.uk/government/publications/2010-11-reference-costs-publication (accessed March 2013).

147. Personal Social Services Research Unit. *Unit Costs of Health and Social Care 2011*. URL: www.pssru.ac.uk/pdf/uc/uc2011/uc2011.pdf (accessed March 2013).

148. *Connevans Limited*. URL: www.connevans.co.uk (accessed 3 December 2012).

149. HAB. *Hearing Aid Batteries and Accessories*. URL: www.hearing-aid-batteries.org.uk (accessed 3 December 2012).

150. Rosenfeld RM, Bhaya MH, Bower CM, Brookhouser PE, Casselbrant ML, Chan KH, *et al.* Impact of tympanostomy tubes on child quality of life. *Arch Otolaryngol Head Neck Surg* 2000;**126**:585–92. http://dx.doi.org/10.1001/archotol.126.5.585

151. CRANE Database. *Annual Report on Cleft Lip and/or Palate 2013*. URL: www.craniofacialsociety.org.uk/downloads/CRANE%20Annual%20Report%202013%20(Final).pdf (accessed March 2014).

152. Doyle WJ, Cantekin EI, Bluestone CD. Eustachian-tube function in cleft-palate children. *Ann Otol Rhinol Laryngol* 1980;**89**:34–40.

153. Stool SE, Randall P. Unexpected ear disease in infants with cleft palate. *Cleft Palate J* 1967;**4**:99–103.

154. Armstrong BW. A new treatment for chronic secretory otitis media. *AMA Arch Otolaryngol* 1954;**59**:653–4. http://dx.doi.org/10.1001/archotol.1954.00710050665001

155. National Institute for Health and Care Excellence (NICE). *Guide to the Methods of Technology Appraisal 2013*. London: NICE; 2013.

156. Brennan A, Kharroubi S, O'hagan A, Chilcott J. Calculating partial expected value of perfect information via Monte Carlo sampling algorithms. *Med Decis Making* 2007;**27**:448–70. http://dx.doi.org/10.1177/0272989X07302555

157. Groot Koerkamp B, Myriam Hunink MG, Stijnen T, Weinstein MC. Identifying key parameters in cost-effectiveness analysis using value of information: a comparison of methods. *Health Econ* 2006;**15**:383–92. http://dx.doi.org/10.1002/hec.1064

158. Bluestone CD. Eustachian tube obstruction in infant with cleft palate. *Ann Otol Rhinol Laryngol* 1971;**80**:1–30.

159. Rawlins MD, Culyer AJ. National Institute for Clinical Excellence and its value judgments. *BMJ* 2004;**329**:224–7. http://dx.doi.org/10.1136/bmj.329.7459.224

160. Claxton K, Ginnelly L, Sculpher M, Philips Z, Palmer S. A pilot study on the use of decision theory and value of information analysis as part of the NHS Health Technology Assessment programme. *Health Technol Assess* 2004;**8**(31). http://dx.doi.org/10.3310/hta8310

161. Kalcioglu MT, Cokkeser Y, Kizilay A, Ozturan O. Follow-up of 366 ears after tympanostomy tube insertion: why is it draining? *Otolaryngol Head Neck Surg* 2003;**128**:560–4. http://dx.doi.org/10.1016/S0194-5998(03)00120-7

162. Schönweiler R, Lisson JA, Schönweiler B, Eckardt A, Ptok M, Tränkmann J, *et al.* A retrospective study of hearing, speech and language function in children with clefts following palatoplasty and veloplasty procedures at 18–24 months of age. *Int J Pediatr Otorhinolaryngol* 1999;**50**:205–17. http://dx.doi.org/10.1016/S0165-5876(99)00243-8

163. Schönweiler R, Schönweiler B, Schmelzeisen R. Hearing capacity and speech production in 417 children with facial cleft abnormalities. *HNO* 1994;**42**:691–6.

164. Macfarlane J, Holmes W, Macfarlane R, Britten N. Influence of patients' expectations on antibiotic management of acute lower respiratory tract illness in general practice: questionnaire study. *BMJ* 1997;**315**:1211–14. http://dx.doi.org/10.1136/bmj.315.7117.1211

165. NHS England. *NHS Standard Contract for Cleft Lip and/or Palate Services Including Non-Cleft Velopharyngeal Dysfunction (VPD) (All Ages)*. URL: www.england.nhs.uk/wp-content/uploads/2013/06/d07-cleft-lip.pdf (accessed April 2014).

166. Rovers MM, Straatman H, Ingels K, van der Wilt GJ, van den Broek P, Zielhuis GA. The effect of short-term ventilation tubes versus watchful waiting on hearing in young children with persistent otitis media with effusion: a randomized trial. *Ear Hear* 2001;**22**:191–9. http://dx.doi.org/10.1097/00003446-200106000-00003

Appendix 1 mOMEnt clinician survey V1.0 30 October 2013

mOMEnt
Management of Otitis Media with Effusion in childreN with ClefT Palate

The MOMENT study is a feasibility study which aims to find out whether a study of the treatments of OME is feasible. As part of this we would like to obtain information on the care pathways in each Network/Centre.

We would like to ask you to complete this questionnaire based on current practice within your centre.

Could you please complete all of the questions that you feel in a position to answer. Tick boxes can be completed by double clicking on the appropriate box and selecting "checked"

If you are able to answer a question please add your initials in the box provided for each question. If you feel the question would be better answered by another member of your team, please add their name and contact details to the appropriate question.

Completion of the questionnaire may require input from more than one person in your team.
When you have provided your answers please return the questionnaire by email to: [redacted] ([redacted]). We will contact any other team members as indicated in your responses.

Section 1 – General information

i. Name of cleft network	
1.2 Name of cleft centre	
1.3 Name and clinical role of person/s completing this questionnaire on behalf of the centre/network	
1.4 contact details (email/tel) of person/s completing this questionnaire on behalf of the centre/network	
1.5 Name of cleft service coordinator	
1.6 email address of cleft service coordinator	

Section 2 – Clinical Provision and Practice

2.1 Does your cleft service have dedicated audiology input based at your centre?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Audiologist	Yes ☐	No ☐
Clinical Scientist (audiology)	Yes ☐	No ☐
Paediatrician in audiology	Yes ☐	No ☐
Audiovestibular Physician	Yes ☐	No ☐

2.2 Does your cleft service have a dedicated ENT clinician?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

ENT consultant	Yes ☐	No ☐
Associate Specialist/staff grade	Yes ☐	No ☐
Other (please specify grade):	Yes ☐	No ☐

2.3 Is there anyone else in your centre that is involved in the management of OME in children with cleft? *For example, community paediatrician (audiology) or audiovestibular physician*

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Yes ☐ No ☐
If yes please give details:

2.4 How often do children with cleft palate receive routine audiological assessment at your cleft centre. If assessment varies by age please give frequency of routine audiological assessment and age ranges.

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.5 What tests do you routinely use to diagnose and guide the subsequent management of OME. If you have a protocol please attach it to this questionnaire. Please tick all that apply.

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Otoscopy ☐

Tympanometry ☐

Age appropriate hearing test ☐

Which of the following age appropriate tests are you able to provide at your centre?

Automated Brainstem audiometry (ABR) ☐

Visual Reinforced Audiometry (VRA) ☐

Distraction testing (DT) ☐

Performance / play audiometry ☐

Speech testing ☐

Pure Tone audiogram ☐

Other, *please give details:* ☐

2.6 At primary cleft palate repair how is the decision made to insert ventilation tubes or not and who is involved in the decision making process?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.7 What factors influence the decision to insert ventilation tubes at primary cleft repair?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.8 **After what period of time would a conductive hearing loss >25–30dBHL trigger 'active' intervention (referral for/decision to insert ventilation tubes or prescribe hearing aids) at your centre**

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.9 **Please describe the decision making process to provide hearing aids or to insert/refer to ENT for consideration of ventilation tubes as the first line treatment for persistent OME. Please include any involvement of parents and/or the child.**

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.10 Please describe the decision making process to provide hearing aids or ventilation tubes if the child has already received treatment and has persistent OME.

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

For patients who have received hearing aids as initial treatment

For patients who have received ventilation tubes as initial treatment

2.11 Under what specific circumstances would you advise against inserting VTs to treat persistent OME in a child with OME?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.12 What would be the maximum number of sets of ventilation tubes that you would consider in a child before advising against further ventilation tube insertion? Please explain your answer

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

1 ☐ 2 ☐ 3☐ 4 ☐ 5 ☐ 6☐ 7☐ 8 ☐ >8 ☐

2.13 What is your view on the optimum age for inserting ventilation tubes?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.14 In what circumstances would you consider offering long term ventilation t-tubes?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.15 In the last five years what is the average yearly number of patients with cleft palate who receive hearing aids as primary management of conductive hearing loss secondary to OME?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Where possible please complete the annual figures in the table below:

Year	Number of referrals
2012	
2011	
2010	
2009	
2009	

1.16 Does your centre offer hearing aid technology other than BTE hearing aids, for example, bone conduction hearing aids?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Yes ☐ No ☐
If yes please give details:

2.17 Please describe the management of children with cleft palate and OME who are not offered ventilation tubes or hearing aids

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

2.18 Up to what age (in years) does your cleft service actively monitor a child's hearing thresholds irrespective of OME?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

0 ☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5 ☐ 6 ☐ 7 ☐ 8 ☐ 9 ☐ 10 ☐ 11 ☐ 12 ☐ 13 ☐ 14 ☐ 15 ☐

16 ☐ 17 ☐ 18 ☐ adulthood ☐

How regularly do these assessments take place?

Does your answer change depending on OME history?

Yes ☐ No ☐
If yes please describe how:

Section 3. Spoke audiological/ENT services

3.1 Do children in the service attend cleft clinics at any other sites outside of your trust?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Yes ☐ No ☐
If yes how many sites provide audiological/ENT services?

3.2 What, if any, are your recommendations to local Audiology and or ENT services regarding the frequency of hearing tests and management of OME in patients with cleft palate outside visits to your cleft service

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Do you receive a copy of the results?
Yes ☐ No ☐

Does the local hospital make decisions regarding ventilation tubes and/or hearing aids?
Yes ☐ No ☐
If yes please give details

Section 5. Information

5.1 What information do you issue to your families about OME and the impact of OME

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Please attach a copy of any written documentation to this questionnaire

5.2 When is the risk of OME and signs of OME first discussed with your families

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

5.3 Who in your network gives advice and discusses OME with families

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Section 6: Service Delivery

6.1 Are there any barriers to the care that you would like to provide to patients with cleft and OME?

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

Yes ☐ **No** ☐
If yes please give details

6.2 If there is anything else that you would like to tell us about the management of OME in your centre please enter it here.

Answered by: (initials)

This question will be better answered by:
Name:
Contact details:

If you have written protocols for the treatment of OME in your centre please return a copy with this questionnaire.

Thank you for completing this questionnaire

Appendix 2 Full results of clinician survey

The survey closed end of working Monday 3 February 2014 and final data entry occurred on Tuesday 4 February 2014.

The statistical analysis was completed on 4 February 2014 and disseminated on the 5 February 2014.

Statistical summaries

Sections refer to sections of the questionnaire sent to cleft sites (see *Appendix 1*).

Section 2: clinical provision and practice

2.1: Does your cleft service have dedicated audiology input based at your centre?

Centre ID	Audiologist: dedicated audiology input	Clinical scientist: dedicated audiology input	Paediatrician: dedicated audiology input	Audiovestibular physician: dedicated audiology input	Number of professions
13	Yes	Yes	No	No	2
10	Yes	No	No	No	1
12	No	No	Yes	No	1
16	Yes	Yes	No	Yes	3
1	No	Yes	No	No	1
17	Yes	No	No	No	1
8	Yes	No	Yes	Yes	3
7	Yes	No	No	No	1
2	Yes	Yes	Yes	Yes	4
6					
11					
4	No	No	No	No	0
5					
15					
14					
9					
Overall *n/N* (%)	7/10 (70.0)	4/10 (40.0)	3/10 (30.0)	3/10 (30.0)	None = 1/10 (10.0) One profession = 5/10 (50.0) Two professions = 1/10 (10.0) Three professions = 2/10 (20.0) Four professions = 1/10 (10.0)

2.2: Does your cleft service have a dedicated ENT clinician?

2.3: Is there anyone else in your centre that is involved in the management of OME in children with cleft? For example, community paediatrician (audiology) or audiovestibular physician.

	Question 2.2			Question 2.3
Centre ID	ENT consultant: dedicated ENT clinician	Associate specialist/staff dedicated: ENT clinician	Other dedicated ENT clinician/text	Anyone else involved in management of OME/text
13	Yes	No	No	Yes/all other ENT consultants, associate specialists, specialist trainees and audiologists in [location]
10	Yes	No	No[a]	No
12	Yes	No	No[a]/[name] as general ENT adviser but children more likely to be seen by local ENT services	Yes[b]/I and my team offer follow-up to all CLEFT children . . . the team comprises audiologists and community paediatricians
16	Yes	No	Yes/ENT fellow	No
1	No	No	Yes/associate specialist ENT	Yes/audiologists, paediatricians (audiology) and ENT at each of the spoke hospital centres
17	Yes	No	Yes/SPR/post-CCT ENT fellow	No
8	No	No	No	No
7	Yes	No	No	Yes
2	Yes	No	No	No
6				
11				
4				No
5				
15				
14				
9				
Overall *n*/*N* (%)	7/9 (77.8)	0/9 (0.0)	3/9 (33.3)	4/10 (40.0)

CCT, controlled clinical trial; SPR, specialist registrar.
a Imputed with 'no' based on answers to previous questions.
b Imputed with 'yes' based on free-text answer that includes 'community paediatricians'.

2.4: How often do children with CP receive routine audiological assessment at your cleft centre. If assessment varies by age please give frequency of routine audiological assessment and age ranges.

Individual listing of answers.

Centre ID	Text answer to 'how often receive routine audiological assessment?'
13	NBHS. 8–10 Months. 18 months. 3 yrs. School screen. 5, 10, 15 and 20 yrs @ audit clinics
10	Normal hearing – 6 mthly if preschool and annually if school age. If a hearing loss is identified these children will follow pathway for CDHL
12	Children seen at 8 months age and then as frequently as clinical needs directs
16	All children seen at 8 months/3 yrs/5 yrs/7 yrs/10 yrs. Seen as clinical need dictates between these times for management of any hearing issues
1	All receive audiological assessment one week prior to primary palate surgery (approx 6 mths – 12 mths depending of palate surgery timing). They receive behavioural assessment follow up before 12 months as per national NHSP protocol requirements. Between 1–4 yrs: they are assessed locally until normal hearing or a sensorineural hearing loss is established bilaterally without intervention (i.e. grommets). Follow up timing is determined on a case by case basis depending on hearing results, timings are likely to be 3 months, 6 months or annual review depending on the hearing results, hearing history, speech and language therapists feedback and parental views. Assessments are 'open' and can be rearranged as and when in response to parental/professional concerns, i.e. families can access services before the scheduled review date. All children with a palatal cleft are recalled for a 5 year audit assessment between 5 : 0 to 5 : 11 as per national cleft service requirements. The 5 yr assessment is reported nationally as part of the cleft service quality dash board. 5yr+ audiological assessment and management as required and/or indicated, in cases of persistent conductive (fluctuating temporary or permanent) or sensorineural hearing loss. There is a requirement for 10 year audit, in our experience this is difficult to achieve in families with no hearing concerns, families with hearing concerns are already in the system. Due to the 'burden' of appointments families rarely want to come at 10 years for a hearing assessment if there are not hearing concerns, particularly if the child is involved in orthodontic care, and are missing school on a regular basis for ortho appointments
17	Post Palate repair (approx 9m), 18M, 2y, 5y, 10y
8	Audiological assessment will take place when babies/children attend the cleft centre at [centre]. I.e. Post palate repair, 18 months, 3 years, 5 years. We do not attend the clinics for children older than 7 years
7	Under 2y every 6mths, over 2y every 12mths. (if hearing is normal). If hearing is down or glue ear, then can change to 3 or 6 mths depending on loss or uni or bil glue
2	3m, 8m, 12m, 18m, 3yr, then as per CSAG
6	
11	
4	
5	
15	
14	
9	

CDHL, conductive hearing loss; CSAG, Clinical Studies Advisory Group; NBHS, newborn hearing screening; NHSP, newborn hearing screening programme.

Schematic summary: how often receive routine audiological assessment?

Timings of routine audiological assessment									
Time point	**13**	**10**	**12**	**16**	**1**	**17**	**8**	**7**	**2**
Newborn	✗	Every 6 months					✗	Every 6 months	✗
9 months	✗		✗	✗	✗	✗	✗		✗
18 months	✗		As frequently as clinical needs directs	✗	✗	✗	✗		✗
3 years	✗			✗	✗	✗ (at 2 years)	✗	Every 12 months	✗
5 years	✗	Every 12 months		✗	✗	✗	✗		As per CSAG
7 years				✗					
10 years	✗			✗	✗	✗			
15 years	✗								
20 years	✗								

CSAG, Clinical Studies Advisory Group.
Note
Missing data for centres: 6, 11, 4, 5, 15, 14, 9.

2.5: What tests do you routinely use to diagnose and guide the subsequent management of OME? If you have a protocol please attach it to this questionnaire. Please tick all that apply.

				Breakdown of age-appropriate hearing test						
Centre ID	Otoscopy	Tympanometry	Age-appropriate hearing test	Automated brainstem audiometry	Visual reinforced audiometry	Distraction testing	Performance/play audiometry	Speech testing	Pure tone audiogram	Other test/text
13	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
10	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
12	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
16	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes/OAEs
1	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes/age-appropriate speech assessments
17	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes	No
8	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes	Yes	Yes/OAEs
7	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes/OAEs
2	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No
6	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	No[a]
11	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes[b]
4	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes/OAEs
5	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes/OAE
15										
14	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes[c]
9										
Overall *n/N* (%)	14/14 (100.0)	14/14 (100.0)	14/14 (100.0)	13/14 (92.9)	14/14 (100.0)	13/14 (92.3)	14/14 (100.0)	13/14 (92.3)	14/14 (100.0)	8/14 (57.1)

OAE, otoacoustic emission.

a Only tymps and pure tone audiogram available at the twice monthly Thursday morning cleft review clinic. All other audiological investigations are arranged subsequently with paediatric audiology/ENT as required. Imputed to 'no' as tests mentioned in text already covered in preceding questions.

b Automated auditory brainstem response; true auditory brainstem response; otoacoustic emissions; electocochleography; speech-in-noise testing; auditory process disorder testing; various tinnitus tests; auditory steady state response testing.

c Otoacoustic emissions. Cochlear microphonics (test rarely performed).

Whether or not protocol was provided?

Centre ID	Protocol provided
13	No
10	No
12	No
16	No
1	Yes
17	No
8	No
7	No
2	No
6	No
11	No
4	No
5	No
15	
14	Yes
9	
Overall *n/N* (%)	2/14 (14.3)

2.6: At primary CP repair how is the decision made to insert VTs or not and who is involved in the decision-making process?

Individual listing of answers.

Centre ID	Text answer to 'how is decision made to insert VTs and who decides?'
13	Not routinely used, as no evidence to support this. Each decision is made on an individual need basis but at this age it is likely that a soft band will be used and not grommet insertion. Decision to insert grommets would be made by ENT consultant, based on info provided by Speciaist [Specialist] audio team and following discussion with parents
10	ENT consultant decides
12	Unlikely unless primary repair very late
16	Dependent on clinical need as assessed by local or regional ENT
1	Audiological assessment and audiological history acts as a gateway for ENT to be present at the same time as palate surgery. ENT decision based on audiological report and clinical presentation
17	We don't insert VT at primary cleft repair
8	We follow the NICE guidelines. Children will be seen in their local audiology departments twice (usually at 8 months and 11 months) and the clinician will recommend grommets or not at palate repair if this has not already happened
7	In the [region], the local audiologist does the hearing assessments (following the hearing assessment and the treatments are discussed by them. If parents show an inclination for grommets, then the audiologists lets me [name] and the cleft coordinator know. I would normally see the patient during their admission or if parents want, arrange to see them in my clinic. I would liaise with the Cleft surgeons and arrange for the Grommets to be inserted and send a letter back to the referring audiology department to pick up the hearing assessments following grommets. If the patient is found to have other pathology – airway/cholesteatoma/Retraction pockets, then I would arrange a follow up appointment in my clinic
2	Audiological Physicians and ENT
6	
11	
4	
5	
15	
14	
9	

Schematic summary: how is decision made to insert VTs and who decides?

Centre ID	No VT at primary	ENT consultant	Audiological physician	Single or joint professional decision		
				ENT alone	Audio alone	Both ENT + audio
13		✗	✗			✗
10		✗		✗		
12	✗					
16		✗		✗		
1		✗	✗			✗
17	✗					
8			✗		✗	
7		✗	✗			✗
2		✗	✗			✗
Overall *n*/*N* (%)	2/9 (22.2)	6/7 (85.7)	5/7 (71.4)	2/7 (28.6)	1/7 (14.3)	4/7 (57.1)

2.7: What factors influence the decision to insert VTs at primary cleft repair?

Individual listing of answers.

Centre ID	Text answer to 'what factors influence the decision to insert VTs at primary cleft repair?'
13	Individual clinics need e.g. recurrent OME, but rarely used at this age!!
10	ENT consultant decides
12	
16	Any hearing loss and impact on development
1	Newborn hearing screen results. Parental concerns. Otology history to date. Audiological assessment results (Otoscopy, tympanometry, behavioural audiometry assessment)
17	
8	Hearing levels raised (the actual level that is used as the threshold can differ from clinician to clinician and it may also depend on the general health of the child, eg always full of a cold) and presence of middle ear fluid
7	Nice guidelines with hearing assessments. Discussion between audiologist and parents. Parents choice. Child's ear canal anatomy (Narrow ear canals in syndromic kids, making access difficult)
2	Hearing levels, parent informed choice, practical clinical considerations
6	
11	
4	
5	
15	
14	
9	

Summary: most common factor is hearing loss with some centres referencing to NICE (CG60).[11] Other contributing factors including parental choice and concerns, otoscopy history, anatomical abnormality restricting surgical access and practice of individual surgeons.

2.8: After what period of time would a conductive hearing loss > 25–30 dBHL trigger 'active' intervention (referral for/decision to insert VTs or prescribe HAs) at your centre?

Individual listing of answers.

Centre ID	Text answer to 'time period of conductive hearing loss > 25–30 dBHL that triggers "active" intervention'
13	NO specific timeframe but if after > 3/12 documented HL [hearing loss] and history of symptoms/parental or professional concerns, then intervention would be discussed/considered
10	2 consecutive hearing tests showing CD hearing loss
12	Dependent on needs of the child – is it delaying language, social interaction, is it associated with ear infections etc – intervention decision would be discussed with family?
16	Any child seen within Audiology service with a loss gets referral to ENT straight away. Management then depends on clinical need on an individual basis
1	When persistent hearing loss demonstrated by two tests 3 mths apart. Where specialist cleft speech and language therapists identify hearing related speech characteristics with audiological assessment demonstrating hearing loss. Multiple/repeated ear infections/tympanic membrane perforations over a short period of time – decision made by ENT – seek clarification from ENT lead
17	Minimum 3m observation and only if the clinical history correlates with the audiological data
8	3 months
7	Persistent hearing loss beyond 3 months
2	Depends on additional clinical factors, speech and middle ear health
6	
11	
4	If the OME is persistent over approximately 2 month period. Following the referral to ENT (about 6 wks wait) evidence of 3 months of persistent OME will have been recorded and can inform the intervention decision as per Nice guidelines
5	
15	
14	
9	

CD, conductive hearing loss.

Schematic summary: time period of conductive hearing loss > 25–30 dBHL that triggers 'active' intervention.

Centre ID	One hearing test showing hearing loss	Two hearing tests showing hearing loss		Depends on factors (in addition to hearing loss)
		3 months apart	Unknown months apart	
13		✗		
10			✗	
12				✗
16	✗			
1		✗		
17		✗		
8		✗		
7		✗		
2				✗
4		✗		
Overall *n*/*N* (%)	1/10 (10.0)	6 (60.0)	1/10 (10.0)	2/10 (20.0)

2.9: Please describe the decision-making process to provide HAs or to insert/refer to ENT for consideration of VTs as the **first-line treatment** for persistent OME. Please include any involvement of parents and/or the child.

Individual listing of answers.

Centre ID	Text answer to 'how decide to provide HAs or insert VTs as first-line treatment'
13	At this age the patient is usually being seen by the Specialist team including Audiologists and community paediatrics +/– ENT consultant. The need for amplification based on neuralplasitcity is discussed. Usually parents are asked to try a softband before contemplating Grommet insertion, because of the expected need for treatment over a long period of time/limitation to number of grommets might arise
10	Parents always contribute to this decision, and to date they have decided VT for management
12	Monitor hearing loss. Assess receptive speech . . . and expressive speech. School concerns? History of ear infections? Otoscopy. Ask parents. Ask child if old enough
16	All children referred to ENT, all options discussed with family – who then make their choice
1	All hearing management options are explained to parents with the pros and cons of each approach by the audiologist. Often parents choose grommets as a first line approach
17	If child has persistent OME and there is both clinical and audiological evidence of hearing loss; the parents are given a choice of either trial BC hearing aid or if > 4y VT tube insertion. VT tube insertion for < 4y is limited to children who have failed a trial of BC/AC hearing aid, who have recurrent AOM or have evidence of Tympanic membrane (TM) damage
8	If hearing levels are raised (usually 45 dBHL or greater) on two occasions, 12 weeks apart, we will discuss the management options with parents and child if old enough
7	The first time a child is seen with glue ear and reduced hearing, results are explained, NDCS glue ear booklet given and a review appt booked. Its explained to parents that to start with we always follow WW and hopefully it will clear on its own. If at next appt things haven't changed then we will discuss the different management options, these are outlined in the booklet given. At next appt [appointment], again results explained and if both h.aids [hearing aids] or grommets are an option, they are discussed and explained then its parents who decide which to go for. (or child if old enough)
2	Discussions with the parents about the impact of the hearing loss and the benefits, risks, and alternatives of treatment, between Grommets vs Hearing aids and Hearing tactics (only)
6	
11	
4	Presence of persistent OME (as described earlier) confirmed through age appropriate hearing assessment and tympanometry. ENT consultant will discuss surgical intervention vs mangment with hearing aids so parents can make an informed choice regarding further management
5	
15	
14	

AC, air conduction; BC, bone conduction; NDCS, National Deaf Children's Society; WW, watchful waiting.

Schematic summary: how decide to provide HAs or insert VTs as first-line treatment.

Decision process	13	10	12	16	1	17	8	7	2	4
Seen by specialist team (audiologist, community paediatrics, ENT consultant)	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗
Monitor hearing loss										
Discuss with family (parents/child if old enough)	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗
Assess receptive speech and expressive speech			✗							
School concerns			✗							
History of ear infections			✗							
Tests (otoscopy, tympanometry, age-appropriate hearing tests)			✗							✗

2.10: Please describe the decision-making process to provide HAs or VTs if the child has already received treatment and has persistent OME.

Individual listing of answers.

	Text answer to 'how do you decide to provide HAs or insert VTs if child has already received treatment'	
Centre ID	For patients who have received HAs as initial treatment	For patients who have received VTs as initial treatment
13	See centre response for 2.9, for very young. Later on if hearing rehabilitation needs to be discussed both Has and vent tubes will be discussed. The pros and cons of each are discussed with parents, who can then consider the options based on their child and likelihood of utising the Has etc.	See centre response for 2.9, but the risks associated with repeated grommet insertion may need to be discussed in more details depending on clinical findings on examination
10		Evidence of ongoing OME over a period of time.
12	Monitor hearing loss. Assess receptive speech . . . and expressive speech. School concerns? History of ear infections? Otoscopy. Ask parents. Ask child if old enough	And again . . . as above
16	Down to patient choice	Patient choice
1	VT would be discussed where the child is unable to consistently use the hearing aids, either due to non-compliance with hearing aid use, because the hearing loss is no longer manageable with hearing aids, the tympanic membranes have become 'unsafe' and require VTs to prevent damage or active infection as a result of severe retraction or because hearing aids can no longer be used due to multiple ear infections or external auditory canal inflammation (otitis externa) through allergy or contact dermatitis or because the amplification available via the aid is no longer adequate to manage the hearing loss. Clarify with lead ENT – Children with persistent OME and had received three grommet/VT insertion should ideally be given hearing aid unless parents are not happy for their child using HA or clinically not suitable for using hearing aid	Hearing aids would be discussed as per with the first line treatment/management discussions, hearing aids as with grommets are discussed as a treatment option with each episode of OME where the clinical decision it to actively manage the OME. Families may choose hearing aids for treatment after previously choosing VTs depending on their experience of VTs. Hearing aids may be the recommended management because of a contraindication to further VT insertion for example if the child suffered with multiple ear infections and required the grommets (VTs) to be removed previously or if they have already had 3 sets of VTs or if the VTs lasted only a very short period of time and were found to be a less effective management option for the individual

	Text answer to 'how do you decide to provide HAs or insert VTs if child has already received treatment'	
Centre ID	For patients who have received HAs as initial treatment	For patients who have received VTs as initial treatment
17	Continue with hearing aid until OME resolves or the child stops using the Aid. If OME persists then VT tubes are offered	Continue with VT tubes. If parents choose hearing aids then the child is observed closely for evidence of TM damage. IF TM damage then VT tubes are placed
8	If already using hearing aids, they will be under regular review by their local audiology department and any changes in their hearing thresholds will be discussed and hearing aids reprogrammed if necessary. If they have previously had hearing aids but these were removed because their hearing improved but has now got worse again, I would discuss management options with parents/child. It probably depends what their experience of hearing aids was like whether they decide to go for aids again or try grommets. Some parents are happier to consider grommets once their child is a little older	If they have previously had grommets, I would discuss the options emphasising the problems of repeated grommet insertion
7	Children are always reviewed and if the child is not getting on with the aids, different management options are discussed and again parents decide what to go for.(for example, referral to ENT, different type of h.aid [hearing aid]). [name]	If ventilation tubes have come out or are blocked and OME is back, we would again follow WW. If after 3 mth [months] it had not cleared, then we would discuss all options again (grommets, hearing aids). For some children a second set of grommets may not be appropriate but that would be the decision of ENT [name]
2	See centre response for 2.9, with more emphasis on the importance of follow up and engagement with hearing aid use, also to monitor middle ear health	See centre response for 2.9, but in addition will highlight the small increase in complications with repeated grommets
6		
11		
4	Presence of persistent OME (as described earlier) confirmed through age appropriate hearing assessment and tympanometry. ENT consultant will discuss surgical intervention vs management with hearing aids so parents can make an informed choice regarding further management	Presence of persistent OME (as described earlier) confirmed through age appropriate hearing assessment and tympanometry. ENT consultant will discuss surgical intervention vs management with hearing aids so parents can make an informed choice regarding further management. The increased risks of TM perforation with repeat sets of grommets will be discussed
5		
15		
14		
9		

TM, tympanic membrane; WW, watchful waiting.

Schematic summary: how decide to provide HAs or insert VTs if child has already received **HAs as initial treatment**.

Decision process	13	10	12	16	1	17	8	7	2	4
Similar to decision process for first-line treatment using HAs or insert VTs	✗		✗						✗	✗
Pros and cons of HAs and VTs are discussed with parents	✗									
Patient choice	✗		✗	✗	✗					
VTs discussed where non-compliance with HA use					✗	✗				
VTs discussed where the amplification available via the aid is no longer adequate to manage the hearing loss					✗	✗				
VTs discussed where tympanic membranes have become 'unsafe' and require VTs to prevent damage or active infection as a result of severe retraction					✗	✗	✗			
VTs discussed where HAs can no longer be used due to multiple ear infections or external auditory canal inflammation (otitis externa) through allergy or contact dermatitis					✗	✗				
Regular review by their local audiology department and any changes in their hearing thresholds will be discussed and hearing aids reprogrammed if necessary							✗			
Referral to ENT								✗		
Different type of HA								✗		
Monitor middle ear health									✗	

Schematic summary: how to decide to provide HAs or insert VTs if child has already received **VTs as initial treatment**.

Decision process	13	10	12	16	1	17	8	7	2	4
Similar to decision process for first-line treatment using HAs or insert VTs	✗		✗		✗				✗	✗
Risks associated with repeated grommet insertion discussed with parents	✗						✗		✗	✗
Patient choice	✗		✗	✗	✗	✗				
Children with persistent OME and had received three grommet/VT insertions should ideally be given HAs unless parents are not happy for their child using a HA or clinically not suitable for using a HA					✗					
HAs may be the recommended management because of a contraindication to further VT insertion for example if the child suffered with multiple ear infections and required the grommets (VTs) to be removed previously					✗					
Continue with VTs. If parents choose HAs then the child is observed closely for evidence of TM damage. IF TM damage then VTs are placed						✗				
If VTs have come out or are blocked and OME is back, we would again follow WW. If after 3 months it had not cleared, then we would discuss all options again (grommets, HAs). For some children a second set of grommets may not be appropriate but that would be the decision of ENT								✗		

TM, tympanic membrane; WW, watchful waiting.

2.11: Under what specific circumstances would you advise against inserting VTs to treat persistent OME in a child with OME?

Individual listing of answers.

Centre ID	Text answer to 'advise against inserting VTs'
13	Very young – recommend trial of softband first. Very atelectatic TM, with possible increased risk of TM Perfoatation development, which might make HA use difficult at a later date
10	Early extruders, thinning of TM
12	If greater risk from GA.?syndromic child, particularly if swallowing problems. Small ear canals. History of previous discharging ears with grommets
16	Where there are anatomical restrictions or history of repeated infections with previous VTs
1	VTs leading to multiple ear infections. VT insertion contraindicated by tympanic membrane appearance/ structure for example too thin to maintain the VTs. I agree with above statement – SH
17	If child is under < 4y and hasn't been tried with hearing aid. Syndromic child who is immune compromised
8	If they have had grommet(s) in the past. If they have dead ear on one side
7	Ent decision, or if child can't have GA. [name]
2	If the ear canal access is difficult or there is an underlying sensorineural hearing loss
6	
11	
4	
5	
15	
14	
9	

GA, general anaesthetic; TM, tympanic membrane.

Schematic summary: advise against inserting VTs.

Specific circumstance	13	10	12	16	1	17	8	7	2
Very young – recommend trial of softband first	✗					✗			
If child is aged < 4 years and has not been tried with a HA									
Very atelectatic TM, with possible increased risk of TM perfoatation development, which might make HA use difficult at a later date	✗	✗			✗				
Thinning of TM									
VT insertion contraindicated by tympanic membrane appearance/structure for example too thin to maintain the VTs									
Early extruders		✗							
Greater risk from GA			✗					✗	
If child cannot have GA									
Syndromic child, particularly if swallowing problems			✗			✗			
Syndromic child who is immune compromised									
Small ear canals			✗	✗					✗
Anatomical restrictions									
If the ear canal access is difficult									
History of previous discharging ears with grommets			✗	✗	✗				
History of repeated infections with previous VTs									
VTs leading to multiple ear infections									
If they have had grommet(s) in the past							✗		
If they have dead ear on one side							✗		
Underlying sensorineural hearing loss									✗

GA, general anaesthetic; TM, tympanic membrane.

2.12: What would be the maximum sets of VTs that you would consider in a child before advising against further VT insertion? Please explain your answer.

Individual listing of answers.

Centre ID	Maximum sets of VT	Explanation of answer
13	Three sets[a]	3 usually, but this would depend on findings at examination and other influencing factors
10		ENT consultant decides
12	Four sets	Depends on gain V [versus] risk benefits
16		No specific number depends on individual patients
1	Three sets	I agree with the above response. Too many myringotomies increases the risk of tympanic membrane perforation, tympano sclerosis. Perforated tympanic membrane increases the risk of ME infection with discharge making the use of HA difficult. SH
17		No maximum. After 2 sets of short term tubes we would place long term tubes
8	One set	I have seen children who have had grommet insertion repeated a number of times and it is likely to affect the function of the TM. Also increasing the risk of chronic ear infections
7	Three sets	Or if the patient develops thin atrophic drum following extrusion of the last set of grommest. Or patient/parent choice
2		There is no absolute number, it is a clinical decision, I have seen patients who had 5–6 grommets elsewhere but these are the minority
6		
11		
4		
5		
15		
14		
9		

TM, tympanic membrane.
a Imputed to '3 sets' based on text answer.

Schematic summary: maximum sets of VTs.

	Maximum			
Centre ID	One set	Three sets	Four sets	No maximum
13		✗		
10				✗
12			✗	
16				✗
1		✗		
17				✗
8	✗			
7		✗		
2				✗
Overall *n*/*N* (%)	1/9 (11.1)	3/9 (33.3)	1/9 (11.1)	4/9 (44.4)

2.13: What is your view on the optimum age for inserting VTs?

Individual listing of answers.

Centre ID	Text answer to 'optimum age for inserting VTs'
13	No optimum age, but I find we recommend them most from 3–4 yrs on
10	Decision should be based on clinical findings.
12	VT are of benefit for a limited period of time . . . so it is a judgment as to when is the best time for an individual child
16	None – when clinical need dictates
1	There is no age limitation in children with cleft palate, usually we prefer to perform myringotomy and grommet insertion when they undergo their first cleft surgery which is around six months. SH
17	Around age 5yr [years]
8	I don't like to see them done too early but this is weighed against the convenience of putting in grommets at the time of palate repair although in practice this does not happen often now because of the NICE guidelines meaning that the repair is often done before the second hearing assessment
7	For OME in cleft children – generally beyond 2 years, when speech is developing
2	This is a clinical decision, I have not seen in my practice a child having grommets younger than 3 months
6	Difficult procedure in small syndromic infants with narrow ear canals. Individual decision based on Otoscopy and hearing results and speech and language. No age limits
11	Depends on indication – e.g. conductive hearing loss secondary to OME or recurrent acute AOM. I would insert them as soon as clinically indicated (with parental consent)
4	–9
5	If needed early then 18–24 months
15	
14	We perform ventilation tube insertion with that of primary cleft palate repair surgery (8 months – 1 year), if persistent glue ear is confirmed. Alternatively if persistent glue ear is confirmed at any ventilation tubes are considered an option age as per national guidelines
9	

Schematic summary: optimum age for inserting VTs.

	Age			
Centre ID	< 1 year (palate repair)	1–5 years	> 5 years	No optimum age
13		✗ (most from 3–4 years onwards)		
10				✗ (based on clinical findings)
12				✗ (clinical decision)
16 [locaton]				✗ (when clinical need dictates)
1	✗ (myringotomy and grommet insertion when they undergo their first cleft surgery which is around 6 months)			
17			✗	
8		✗		
7		✗		
2				✗ (clinical decision)
6				✗ (individual decision based on otoscopy and hearing results and speech and language)
11				✗ (insert them as soon as clinically indicated)
5		✗ (if needed early then 18–24 months)		
14	✗ [insertion with that of primary CP repair surgery (8 months–1 year)]			
Overall *n/N* (%)	2/13 (15.4)	4/13 (30.8)	1/13 (7.7)	6/13 (46.2)

2.14: In what circumstances would you consider offering long-term ventilation T-tubes?

Individual listing of answers.

Centre ID	Text answer to 'circumstances to consider offering long-term ventilation T-tubes'
13	On third set of grommets (so usually about 7 years +) and patient did not get on with trial of HA in past. Parents understand and accept the associated risks
10	ENT consultant decides
12	If grommets/aids not alternatives
16	After at least one previous set of VTs dependent on child's age
1	In children who had persistent OME and had standard grommet insertions previously and who is not an appropriate candidate for hearing aid use.SH
17	Persistent OME following 2 sets of short term tubes or if there is evidence of TM damage (grade > 2)
8	That would not be my decision. I might say to parents that ENT may consider it but would never recommend it
7	Not for OME. Consider for persistent retraction pockets after first set grommets
2	In our practice in has not been necessary, and we have not had any
6	
11	
4	
5	
15	
14	
9	

TM, tympanic membrane.

Schematic summary: circumstances to consider offering long-term ventilation T-tubes.

Circumstance	13	10 h	12	16	1	17	8	7	2
On third set of grommets (so usually about ≥ 7 years) and patient did not get on with trial of HA in past	✗		✗		✗				
If grommets/aids not alternatives									
In children who had persistent OME and had standard grommet insertions previously and who is not an appropriate candidate for hearing aid use									
ENT consultant decides		✗					✗		
That would not be my decision. I might say to parents that ENT may consider it but would never recommend it									
After at least one previous set of VTs dependent on child's age				✗					
Persistent OME following two sets of short-term tubes or if there is evidence of TM damage (grade > 2)						✗			
Not for OME. Consider for persistent retraction pockets after first set grommets								✗	✗
In our practice in has not been necessary, and we have not had any									

TM, tympanic membrane.

2.15: In the last 5 years what is the average yearly number of patients with CP who receive HAs as primary management of conductive hearing loss secondary to OME?

Individual listing of answers.

Centre ID	Text answer to 'average yearly number of patients with CP who receive HAs as primary management of conductive hearing loss secondary to OME'
13	NO idea. This data is not available to me at present, as ENT care is delivered locally (as per NICE guidelines)
10	None
12	Sorry do not have the ability to answer this
16	Data not available
1	We do not have this data as children are seen across several hospitals
17	There were originally 23 but some do not require them now or have tried them and then had grommets fitted. Audiology's hearing aid database currently reports 12 Cleft hearing aid patients
8	No idea as hearing aids fitted in local services
7	
2	About 5%
6	
11	
4	Unable to extract data
5	
15	
14	
9	

Schematic summary: average yearly number of patients with CP who receive HAs as primary management of conductive hearing loss secondary to OME.

Centre ID	Average yearly number of patients
13	Unknown number of patients
10	0
12	Unknown number of patients
16	Unknown number of patients
1	Unknown number of patients
17	12
8	Unknown number of patients
7	Unknown number of patients
2	Unknown number of patients (but 5% of unknown total)
[Location]	Unknown number of patients

2.16: Does your centre offer HA technology other than behind the ear HAs, for example, bone conduction HAs?

Centre ID	Centre offers HA technology other than BTE HAs	Details
13	Yes	Considered on individual need basis
10	Yes	Spectacle, BC and CROS. Refer to other service for BAHA
12	Yes	
16	Yes	BAHA on softband
1	Yes	BTE, bone conduction hearing aids, and soft band hearing aid technology available depending on the degree of hearing loss and the child
17	Yes	BC on soft band/hard band, BAHA softband, or implant
8	No[a]	Hearing aids are not fitted at the centre, all done at local audiology service
7	Yes	Contact mini soft/hard bands [name]
2	Yes	
6		
11		
4	Yes	Bone Conduction Aids
5		
15		
14		
9		
Overall *n*/*N* (%)	9/10 (90.0)	

BAHA, bone anchored hearing aid; BC, bone conduction; BTE, behind the ear; CROS, contralateral routing of sound.
a Imputed to 'no' based on text answer.

2.17: Please describe the management of children with CP and OME who are not offered VTs or Has.

Individual listing of answers.

Centre ID	Text answer to 'management of children with CP and OME who are not offered VTs or HAs'
13	Some children with be monitored with watchful waiting if appropriate for their individual clinical needs
10	Regular monitoring
12	If mild . . . advice to parents and school re 'hearing tactics' and good acoustic environment
16	Treatment is offered if required – otherwise hearing tactics for home/school given when parents decline treatment
1	All families are given the NDCS glue ear leaflet and locally written leaflets on cleft palate and hearing loss and early listening skills at their primary palate preadmission assessment appointment. The lead audiologist for the network explains the mechanism and impact of OME at the appointment using the NDCS leaflet diagrams, this allows parents to take the annotated diagrams home for future reference. Where grommets are required at the same time as surgery parents are given a leaflet about grommet inserion [insertion] addressing commonly asked questions associated with this procedure. SLT and audiology discuss communciatoin [communication] tactics at the regular SLT and Audiology appointments throughout early childhood. Families are given communication tactics regarding mild and or fluctuating hearing loss tailored to the individuals hearing loss and hearing history as part of their audiological assessment debrief/individual management plan setting. [location] run a specialsist [specialist] speech and hearing clinic run by the lead audiologist and specialist speech and language therapists. This clinic is for children with hearing and speech concerns and children/families from across the network can be referred and assessed in this clinic. The main aim is to explore hearing impact on speech and to tailor future therapy, communication, listening and hearing management advice using an MDT approach building a joint speech and audiological individual management plan
17	Observation for those who have OME but no clinical history of hearing issues. Observation and support for those syndromic children who have OME and VT or hearing aids are not appropriate
8	Continue watchful waiting, give parents advice about talking to child in quiet, ie turn off TV, child should sit at front of class, teacher should be aware that there is a hearing problem, talk to them from front etc, we have a leaflet with this advice
7	If a child has persistent glue ear and reduced hearing they would always be offered h.aid or grommets assessment [name]
2	Hearing tactics and Listening strategies
6	
11	
4	
5	
15	
14	
9	

MDT, multidisciplinary meeting; NDCS, National Deaf Children's Society; SLT, speech and language therapy.

Schematic summary: management of children with CP and OME who are not offered VTs or HAs.

Circumstance	13	10	12	16	1	17	8	7	2
Watchful waiting	✗	✗				✗	✗		
Regular monitoring									
Observation for those who have OME but no clinical history of hearing issues									
Observation and support for those syndromic children who have OME and VT or HAs are not appropriate						✗			
Hearing tactics and listening strategies			✗	✗	✗		✗		✗
Written leaflets					✗		✗		
If a child has persistent glue ear and reduced hearing they would always be offered HA or grommets assessment								✗	

2.18: Up to what age (in years) does your cleft service actively monitor a child's hearing thresholds irrespective of OME?

Individual listing of answers.

Centre ID	Maximum age monitor hearing (years)	How regularly assessments take place	Does answer change depending on OME history	Description of change depending on OME history
13	Adulthood	See question above asking about 'routine monitoring'	Yes	Will be monitored into adulthood if clinical need e.g. persistent ear problems (including retraction pockets, OME etc) are present
10	16	6 mthly for preschool and annually thereafter sooner if a hearing loss is identified	No	Always dependent on hearing thresholds obtained
12		Cleft audit clinic at 5 10 and 15 years	Yes	Active monitoring dictated by clinical need
16	10	See question 1	Yes	If OME at 10 – this is monitored/ treated as required
1	5	Audiological assessments are available for all up to 18 yrs and beyond this via the GP. If a child continues to have OME at 5 yrs then they would be monitored until this has resolved and their hearing is stable. Active management for all regardless of hearing status is until the 5 year audit point. All children are seen until hearing is within the normal range and stable at that level. The regularity of appointments is individaully set as per answer in 2.4	No	Children are seen until hearing is within the normal range and stable at that level
17		Standard – 2y, 5y, 10y. If issues up to 16y frequency depends on history and progress		As above
8	7	They take place at the times that the child comes to the cleft unit (see above)	No	No because this will be done locally and the frequency of assessments will vary depending on the hearing thresholds, the degree of difficulty the child is experiencing etc.
7	18	See 2.4 [name]	Yes	More frequent review depending on hearing loss and bil or uni OME [name]
2	Adulthood	Please see answer 2.4	Yes	And other otological and Audiological presentation of the case
6				
11				
4				
5				
15				
14				
9				

Section 3: spoke audiological/ear, nose and throat services

3.1: Do children in the service attend cleft clinics at any other sites outside of your trust?

Centre ID	Attend other sites outside of your trust?	How many other sites outside of your trust provide audiological/ENT services?	
		Number of sites	Text response
13	Yes	7	These are different questions!! Attend cleft clinics: 1 other site. The team go to [location] to hold clinics there for patients living nearer that area of [location]. No of Sites providing ENT/Audiology services: this equates to the number of healthe boards across [location] (7 I think) as ENT and Audiology services are provided locally (as per NICE)
10	No	0	
12	Yes	6	[locations]
16	Yes	Missing	All spokes in [location]region have audiology
1	Yes	14	14 hospital sites
17	Yes	12	Approx 12 sites
8	Yes[a]	Missing	Do you mean dedicated cleft clinics within audiology depts. [departments]? I do not think this happens anywhere, children are put into regular assessment clinics
7	Yes	Missing	Audiology is provided at local services. [name]
2	Yes	2	2
6	Yes	3	Probably [locatons], and [location] – 3 other sites outside our trust
11	Yes	7	At least 7 spoke sites in the [location]. We call them all back to [location]for audit clinics though
4	Yes		
5			
15			
14	Yes	9	Approx. 8 – 10 sites
9			
Overall *n*/*N* (%)	12/13 (92.3)	*N* = 9	
		Median = 7 spokes	
		Minimum = 0 spokes	
		Maximum = 14 spokes	

a Missing data imputed to be 'yes' given text answer to how many sites provide audiological/ENT services.

3.2: What, if any, are your recommendations to local audiology and or ENT services regarding the frequency of hearing tests and management of OME in patients with CP outside visits to your cleft service?

	Recommendations to local audiology and/or ENT services			Local hospital make decisions regarding VTs and/or HAs	
Centre ID	Text answer	Spoke protocol: same as hub or spoke specific?	Receive a copy of the results?	Spoke decides?	Details
13	See answers on monitoring schedule above. This has been agreed with local Audiology and ENT services	Same as hub	Yes	Yes	This is very repetitive. All answers given above apply here also
10	6 mthly for preschool and annually for school age, sooner if a hearing loss is identified	Same as hub	No	Yes	Unable to specify
12	Dependent on need	Same as hub	Missing	Yes	
16	All spokes are advised to follow same protocol as described above	Same as hub	No	Yes	Request insertion of VTs at same time as palate surgery in hub or insert within own institution
1	See attached network protocol	Same as hub	Yes	Yes	They make decisions as per the individual cases, in line with NICE guidelines and according to locally available technology in the case of hearing aids
17	No direct recommendations. They usually continue with the management recommended by us but do support and manage patients independently if appropriate	Same as hub	Yes	Yes	Depends if local ENT are involved
8	Our regional policy states 6 monthly assessments if hearing is normal but this will change if hearing affected. I think this pathway needs to be revised. In practice services see children at 8 months initially if newborn screen normal	Same as hub	Yes	Yes	The majority of hearing assessments are carried out locally, usually 6 monthly until about 7 years of age by which time most services report that if the hearing is normal they have usually DNA'd by then
7	None, they have their own policies. [name]	Spoke specific	Yes	Yes	They are their patients so they make all decisions. [name]
2	As per protocol 2.4	Same as hub	Yes	Yes	
6					
11					
4					
5					
15					
14					
9					
Overall *n*/*N* (%)	Protocol same as hub = 8/9 (88.8); spoke-specific protocol = 1/9 (11.1)		6/8 (75.0)	9/9 (100.0)	

DNA, did not attend.

Section 4: case load

4.1: What is the average number of new CP (cleft lip and palate, and palate only) patients referred to your service each year?

Centre ID	2012	2011	2010	2009	2008	Annual average
13	45	55	43	48	45	47.2
10	29	29	38	40	44	36
12[a]	51	47	48	39	60	49
16						
1[b]	178	147	188	182	210	181
17	117	108	107	108	106	109.2
8	CP = 28; BCLP = 11; UCLP = 19; all types = 58	CP = 36; BCLP = 16; UCLP = 12; all types = 64	CP = 38; BCLP = 15; UCLP = 22; all types = 75	CP = 40; BCLP = 4; UCLP = 19; all types = 63	CP = 37; BCLP = 9; UCLP = 12; all types = 58	CP = 35.8; BCLP = 11.0; UCLP = 16.8; all types = 63.6
7	CP = 37; BCLP = 8; UCLP = 13; all types = 58	CP = 40; BCLP = 9; UCLP = 14; all types = 63	CP = 43; BCLP = 10; UCLP = 21; all types = 74	CP = 46; BCLP = 10; UCLP = 12; all types = 68	CP = 41; BCLP = 7; UCLP = 26; all types = 74	CP = 41.4; BCLP = 8.8; UCLP = 17.2; all types = 67.4
2[c]	233	132	119	104	86	134.8
6	29	31	36	33	37	33.2
11	46	41	45	56	60	49.6
4[d]						
5[e]	73	67	73	74	76	72.6
15	33 (including 1 SMCP)	45 (including 4 SMCP)	44 (including 4 SMCP)	48 (including 9 SMCP)	44 (including 12 SMCP)	42.8 (including SMCP); 37.2 (excluding SMCP)
14	43	40 (including 1 SMCP)	47 (including 5 SMCP)	35 (including 4 SMCP)	46 (including 3 SMCP)	42.2 (including SMCP); 39.6 (excluding SMCP)
9	155	146	120	131	131	136.6

BCLP, bilateral cleft lip and palate; MDT, multidisciplinary meeting; SLT, speech and language therapy; SMCP, submucous cleft palate; UCLP, unilateral cleft lip and palated.
Additional text provided by centre:

a [location] (Excludes [location]). CPs (all types). NB: number is for patients referred up to age of 12 months only.
b Our acting database manager has just done a quick calculation on the database. This includes all new referrals – antenatal, postnatal and GP/SLT type referrals to the service.
c Number of 'new' MDT appointments each year.
d Thank you for your enquiry. I regret that I am unable to provide this information as I believe that we are not currently registered as participating in this study. However this information is submitted to CRANE and this is available to view. [name], Cleft Co-ordinator.
e Please note these figures do not include non-cleft VPI referrals. (CB – we asked whether to include lip involvement, transfers in and late referrals in these figures. Replied yes.)

Section 5: information

5.1: What information do you issue to your families about OME and the impact of OME?

Centre ID	Information to your families		Attached written documentation
	Text answer	Information booklet	
13	Baby booklets provided to parents at birth. Info leaflets e.g. that provided by NDCS on HAs, OME, are provided where appropriate	Own leaflets; NDCS	No
10	NDCS documentation	NDCS	No
12	Our own leaflet – sorry don't have access to it electronically	Own leaflets	No
16	Families given NDCS leaflets – please see NDCS website for copies	NDCS	No
1	NDCS glue ear/cleft palate leaflet. Early listening skills. Cleft palate and hearing loss. Grommet insertion at the same time as primary palate repair	NDCS	No
17		Missing	No
8	There is a section on hearing, OME and treatment options in the information given to all parents of babies with cleft palate. We sometimes hand out the NDCS booklet on cleft palate but usually use our in house generic leaflet on glue ear	Own leaflets; NDCS	Yes
7	NDCS glue gear booklet. NDCS cleft palate booklet. [name]	NDCS	No
2	Local patient information leaflets, in additional to NDCS and CLAPA information	Own leaflets; NDCS; CLAPA	No
6			
11			
4	I cannot answer for patients attending ENT services of cleft appointments, however in audiology NDCS info on OME is available		No
5			
15			
14			
9			
Overall *n/N* (%)	Own leaflets = 4/9 (44.4); NDCS = 8/9 (88.9); CLAPA = 1/9 (11.1)		1/10 (10.0)

NDCS, National Deaf Children's Society.

5.2: When is the risk of OME and signs of OME first discussed with your families?

Individual listing of answers.

Centre ID	Text answer to 'when is the risk of OME and signs of OME first discussed with your families?'
13	Mentioned in Baby booklet. By Cleft Nursing staff at time of home visits. Advised baby clinic, e.g. 6/52 age, by cleft surgeon. By SLT at Babble therapy workshops at 6/12. S/B [seen by] Audioogists [audiologists] at 8–10 months and further advice given
10	As soon as OME is confirmed
12	At first visit hearing is discussed
16	At every routine audiology appointment
1	OME is discussed actively from the early contacts and repeatedly by team members through the early years. It is FIRST discussed by the cleft nurses in the early home based appointments, then in the combined cleft MDT clinic appointments when the cleft diagnosis is clarified and surgery first discussed with the consultant surgeon, then again by the SLTs at the babble appointment (3mths of age) and again in detail at the preadmission appointment by the lead audiologist (6mths) one week before palate surgery when the information documented above is issued
17	At first ENT appointment (post palate repair) or when they first present with OME
8	It will usually all be mentioned at the first meeting with parents when they come to the cleft unit soon after birth. Virtually all the children have OME by the time they come for their assessment at 8 months (at local clinic) so we talk about effects this can have on the hearing. Risks are always discussed when it becomes necessary to discuss treatment options for OME with associated hearing loss
7	At first appt. [name]
2	Antenatal visit and in the initial attendance at the MDT
6	
11	
4	
5	
15	
14	
9	

MDT, multidisciplinary meeting; SLT, speech and language therapy.

Schematic summary: when is the risk of OME and signs of OME first discussed with your families?

Centre ID	Antenatal	Newborn	6 weeks	3 months	6 months	9 months
13		Mentioned in Baby booklet Cleft Nursing staff at time of home visits	Advised baby clinic, e.g. 6/52 age, by cleft surgeon		SLT at Babble therapy workshops	Audioogists at 8–10 months
10		As soon as OME is confirmed				
12		At first visit hearing is discussed				
16		At every routine audiology appointment				
1		Cleft nurses in the early home based appointments	Combined cleft MDT clinic appointments when the cleft diagnosis is clarified and surgery first discussed with the consultant surgeon	SLTs at the babble appointment (3mths of age)	Preadmission appointment by the lead audiologist (6mths) one week before palate surgery	
17					At first ENT appointment (post palate repair) or when they first present with OME	
8		First meeting with parents when they come to the cleft unit soon after birth				Virtually all the children have OME by the time they come for their assessment at 8 months (at local clinic) so we talk about effects this can have on the hearing
7		At first appt. [appointment]				
2	Antenatal visit		Initial attendance at the MDT			

MDT, multidisciplinary meeting; SLT, speech and language therapy.

5.3: Who in your network gives advice and discusses OME with families?

Individual listing of answers.

Centre ID	Text answer to: Who in your network gives advice and discusses OME with families
13	All professionals in contact with patient
10	Depends on clinic attended, sometimes the audiologist other times the ENT consultant – whoever leads the clinic
12	Cleft surgeons at initial visit . . . . . SLT Audiology
16	All audiology/ENT services
1	The whole team as required: Nurses, SLTs, audiologist, psychologists, consultant plastic surgeon. The written information and discussion around this is given by the lead audiologist
17	ENT doctor or audiologist seeing child
8	All of us involved in the audiology assessments i.e. consultants and audiologists. The cleft nurses give advice as do SALTS and cleft surgeons to some extent
7	Audiology team, ENT and cleft team. [name]
2	Audiological Physician and ENT, and on follow visits Audiologists
6	
11	
4	Audiologist/ENT cons [consultants]/Cleft Team
5	
15	
14	
9	

SALTS, speech and language therapists; SLT, speech and language therapy.

Schematic summary: who in your network gives advice and discusses OME with families?

Centre ID	Cleft nurse	Cleft surgeon	SLT	Audiologists	ENT consultant	Psychologists
13	✗	✗	✗	✗		
10				✗	✗	
12		✗	✗	✗		
16				✗	✗	
1	✗	✗	✗	✗		✗
17				✗	✗	
8	✗	✗	✗	✗		
7	✗	✗		✗	✗	
2				✗	✗	
4	✗	✗		✗	✗	

SLT, speech and language therapy.

Section 6: service delivery

6.1: Are there any barriers to the care that you would like to provide to patients with cleft and OME?

Individual listing of answers.

	Text answer to 'barriers to the care'	
Centre ID	Yes/no	Details
13	Yes	Integrated records: each dept has it's own records. This means I rely on professionals remembering to copy letters to me for all cleft patients seen. Financial/Time: I would like to review every cleft child in ENT clinics from an early age but there is not the resource to allow this at present. Currently they are seen by my only at the audit clinics 5, 10 yrs [years] etc. Currently there is co co-ordination between local departments and the cleft team however!
10	No	
12	No	
16	Yes	Funding issues with unilateral ventilation
1	Yes	Lack of knowledge of the best practise approach to managing OME. Working across spoke hospitals ensures family centred care delivered close to home but makes collection of assessments and application of a single protocol or policy difficult to apply consistently, particularly in the absence of a nationally agreed protocol
17	Yes	BC hearing technology is still not as advanced as BAHA softband, but we can't provide BAHA soft bands to all our patients who have OME!
8	Missing	
7	No	
2	Yes	Competing staff, equipment and testing rooms/theatre time with other initiatives and services within the department and with other Units externally
6		
11		
4		
5		
15		
14		
9		

BAHA, bone anchored hearing aid; BC, bone conduction.

6.2: If there is anything else that you would like to tell us about the management of OME in your centre please enter it here.

Individual listing of answers.

Centre ID	Text answer to 'anything else about the management of OME in your centre'	Attached written protocol
13		No
10		No
12		No
16		No
1	We believe that the active early intervention model with all members of the team actively educating families on hearing improves the ability to inform individualised patient care and tailor individual management plans including how best to and when to manage OME. Working closely with specialist SLTs is critical for exploring the impact of OME on speech and listening skills. A close working relationship between the audiologist and SLT helps inform both audiological management and speech therapy advise planning and delivery. Giving parents information on signs to look out for with OME and raising their awareness of the presentation and time course of OME help to empower the parents/families to ask for hearing assessments/or ask for advice about hearing/behavioural presentations as they have concerns rather than waiting for set appointments	Yes
17		No
8		No
7		No
2		No
6		
11		
4		
5		
15		
14		
9		

SLT, speech and language therapy; VPI, velo pharyngeal insufficiency.

Appendix 3 Topic guides for qualitative interviews

mOMEnt
Management of Otitis Media with Effusion in childreN with ClefT Palate

The management of Otitis Media with Effusion in children with Cleft Palate – Qualitative Study

Initial Interview Topic Guide - Parents

Note: The following topic guide is indicative. Interviews will only be loosely structured moving between topics in response to what the parent is saying.

A. Parents/Carers will be asked to tell the story of their child's cleft

- I'd like to start by asking you to tell me about your child's cleft and associated glue ear.
- When did you first notice a problem with your child's ears? – what alerted you?

B. Discussion of the treatment received

- What treatments the child received for glue ear (eg one or more grommets operation)
- What other help they received to deal with symptoms (eg hearing aids)
- What choices were made available to you?
- How were decisions made about treatments (eg involvement of child and/or parent in decisions)
- What information was available to choose between treatments?
- Why did you choose the treatment you did?
- What would you advise another parent about treatment choices?

C. Views about parent's satisfaction with the treatment their child received

- Experiences of surgery: hospitalisation, general anaesthetic, post operative care, absence from school.
- If grommets fitted, have they fallen out, when?
- Experience of hearing aids: how they were used, how they do/do not help with hearing (eg parents, siblings, teachers, friends)
- What was helpful about surgery and/or hearing aids
- What problems were associated with surgery and/or hearing aids
- What would you advise another parent about treatment?

D. Views about parent's expectations from treatment and ideal outcomes

- What did you hope the treatment would do to help your child?
- What effect would a treatment have to be useful?
- What did you expect from the treatment?
- Which aspects are important in terms of determining the success/failure of treatment?
- What would you tell another parent about what to expect from treatment?

E. Discussion about the impact of glue ear and its management on the family's day-to-day routines

- What impact did/has glue ear had on the family's routines?

- What impact did it have on other children in the house?
- What impact did it have on other adults in the house?
- Can you rate the impact of your child's illness on the family's routines on a scale of 1 to 5 where 1 is a low impact and 5 the highest.
- What impact did it have on family expenditure?
- Were additional appointments needed at audiology for example, due to the treatment received – adjustment/changing of hearing aids? – if so what impact did this have?

F. Discussion about the impact of glue ear and its management on the child's day-to-day activities

- What impact did/has glue ear have on your child's routines?
- What impact did it have on your child's school performance and progression?
- What impact did it have on your child's relationships at home as well as at school?

G. Discussion about willingness to partake in a trial comparing grommets and hearing aids

- Would you be willing to allow your child to take part in a trial comparing two common treatments for glue ear (Grommets and hearing aids)?
- Would you be likely to participate if it meant that your child was randomly allocated to one of the treatments?
- What kind of information would you need to help you make this decision?

H. Views about any recommendations for other parents

- What is your main recommendation?
- What outcome of treatment has/would have had the most impact on you?

mOMEnt
Management of Otitis Media with Effusion in childreN with ClefT Palate

The management of Otitis Media with Effusion in children with Cleft Palate – Qualitative Study

Final Interview Topic Guide - Parents

Note: The following topic guide is indicative. Interviews will only be loosely structured moving between topics in response to what the parent is saying.

Topic guide for with parents

1. Thank the participant for agreeing to be interviewed
2. Brief outline of the purpose of the research – want to focus today on talking about your child's glue ear – realise there are other things that relate to having a cleft but for the purpose of this project is to focus on glue ear
3. Reassurance about anonymity, confidentiality, and non impact on service delivery – remind participant that you will only repeat what they have said to a member of their treatment team if they or others are thought to be at significant risk of harm
4. Check it is still OK to record the conversation (even though written consent will have been received)
5. Ask about any concerns before starting
6. Have they got any questions?
7. Make clear = OK to stop at any point or refuse to answer questions during interview – no right or wrong answers

A. Tell story of child's glue ear

- To start, can you tell me about your child's glue ear.
- When did you first notice a problem with your child's ears? What alerted you?
- How aware were you that glue ear was common among children with cleft palate?
- What problems did your child experience?
  - Pain, hearing, discomfort, bunged up, restriction of activities, time off school, social interactions, communication difficulties

B. Discuss treatment received

- What choices were made available to you? What options were you given?

- How were decisions made about treatments?
- What about taking no action – was that given as an option?
- How did this compare to other decisions you've had to make about your child's health?
- What information was available to you about different treatments?
- Why did you choose the treatment you did?
- How involved was your GP?
- How old was your child when he/she received the treatment?
- How long did it take to get treatment?
- What would you advise another parent about treatment choices?

C. Views about satisfaction with the treatment their child received

- Grommets: Can you tell me about the surgery – what was it like?
  - Hospitalisation, anaesthetic, post op care, school absence
- If grommets fitted, have they fallen out, when?
- How often did have to go for check-ups?
- Did it make things better or worse?
- Hearing aids: Can you tell me about getting a hearing aid?
  - How did your child get on with a hearing aid?
  - Did it make things better or worse?
- How often did you have to go to the hearing aid service?
- Both: What was helpful about surgery and/or hearing aids?
- What problems were associated with surgery and/or hearing aids?
- What changes have you noticed in your child following treatment?
- What things didn't change?
  - Number of ear infections, ear discharge

D. Views about expectations from treatment and ideal outcomes

- What did you hope the treatment would do to help your child?
- ON IPAD – If a child has treatment for glue ear, as a parent what would you be looking for in terms of results from that treatment? What things would you look to change or improve?
- **ASK PARENTS TO RANK IN IMPORTANCE**
- Why did these results matter to you?
- What were the drawbacks of any treatment your child has had?
- What are the long-term problems that might be a concern for you with your child's hearing?

E. Discuss impact of glue ear and its management on family's day-to-day routines

- What impact did/has glue ear had on the family's routines?

- What impact did it have on other children in the house?
- What impact did it have on other adults in the house?
- What impact did it have on family finances?
- Were additional appointments needed - at audiology for example, due to the treatment received – adjustment/changing of hearing aids? If so, what impact did this have?
    - Time off work?

F. Discuss impact of glue ear and its management on child's day-to-day activities

- What impact has glue ear had on your child's routines?
- What impact did it have on your child's school performance?
- What impact did it have on your child at home? At school?
    - Time off school?

G. Views about any recommendations for other parents

- If you knew another parent who was considering treatment for their child's glue ear, what would you recommend?

H. Discuss willingness to have child involved in a trial

I want to end by getting your advice on another study we hope to carry out in the future. We are not asking you to take part in this research but would value your views because it would involve children with glue ear. The study would be what is known as a clinical trial. **EXPLAIN THIS.**

At the moment it's not clear what treatment is best for glue ear. We would like to do a trial comparing two different treatments. The best way to do a fair test between two types of treatments is for there to be an equal chance of children receiving treatment A and B. This could be done by a computer programme or by rolling a dice - if an even number comes up the child receives treatment A, if an odd number comes up they receive treatment B.

So if a parent agreed to let their child be part of this type of trial it wouldn't be a doctor who decided what treatment they received or the parent and the child would have an equal chance of receiving treatment A or B. What they did receive would be down to chance. Does this make sense?

- Would you have let your child take part if you knew they would get a treatment depending on chance?
    - Why?
    - Is there anything that would change your view?
    - What would you want to know before you decided?
    - How would you want to find out about the study?
    - If there was some sort of pill your child could take and researchers wanted to test that how would you feel?

End question

- Is there something else that you want to say about your experiences of the treatment for glue ear received by your child that we haven't covered already?

Appendix 4 Core outcome set systematic review search strategies

Cochrane Central Register of Controlled Trials

URL: www.cochrane.org/editorial-and-publishing-policy-resource/cochrane-central-register-controlled-trials-central via Ovid search strategy https://ovidsp.ovid.com/

Date range searched: 2006 to 14 April 2011.

Date search performed: 14 April 2011.

Search strategy

#1 MeSH descriptor cleft palate explode all trees (92)

#2 MeSH descriptor cleft lip explode all trees (69)

#3 (cleft* in All Text near/6 palate* in All Text) (176)

#4 (cleft* in All Text near/6 lip* in All Text) (131)

#5 "hare lip*" in All Text (2)

#6 harelip* in All Text (2)

#7 Palatoschisis in All Text (1)

#8 (orofacial* in All Text near/6 cleft* in All Text) (6)

#9 (facial* in All Text near/6 cleft* in All Text) (13)

#10 (face* in All Text near/6 cleft* in All Text) (7)

#11 (#1 or#2 or#3 or#4 or#5 or#6 or#7 or#8 or#9 or#10) (204)

#12 MeSH descriptor middle ear ventilation explode all trees (200)

#13 MeSH descriptor otitis media explode all trees (820)

#14 grommet* in All Text (72)

#15 (ear in All Text near/6 ventilat* in All Text) (232)

#16 otitis next media in All Text (1527)

#17 (ventilat* in All Text near/6 tube* in All Text) (408)

#18 tympanostom* in All Text (135)

#19 (glue in All Text near/6 ear* in All Text) (65)

#20 (#12 or#13 or#14 or#15 or#16 or#17 or#18 or#19) (1908)

#21 (#11 and#20) (17)

Cumulative Index to Nursing and Allied Health Literature via Ovid

URL: www.ebscohost.com/nursing/products/cinahl-databases/cinahl-complete via Ovid search strategy – https://ovidsp.ovid.com/

Date range searched: 1982 to February week 2 2006.

Date search performed: September 2012.

Search strategy

1. Cleft Palate/ (686)
2. Cleft Lip/ (526)
3. (cleft$ adj3 lip$).tw. (495)
4. (cleft$ adj3 palat$).tw. (617)
5. hare lip$.tw. (1)
6. harelip$.tw. (0)
7. Palatoschisis.tw. (0)
8. (orofacial$ adj3 cleft$).tw. (29)
9. (facial adj3 cleft$).tw. (40)
10. (oral adj3 cleft$).tw. (49)
11. (craniofacial adj3 cleft$).tw. (47)
12. or/1-11 (817)
13. Middle Ear Ventilation/ (224)
14. exp Otitis Media/ (1259)
15. grommet$.tw. (21)
16. (ear$ adj3 ventilat$).tw. (80)
17. otitis media.tw. (898)
18. (ventilat$ adj3 tube$).tw. (99)
19. tympanostom$.tw. (90)
20. (glue adj3 ear$).tw. (32)
21. or/13-20 (1634)
22. 21 and 12 (17)
23. from 22 keep 1-17 (17)

Cumulative Index to Nursing and Allied Health Literature via EBSCO*host*

URL: www.ebscohost.com/nursing/products/cinahl-databases/cinahl-com via EBSCO*host* search strategy – www.ebsco.com/

Date range searched: February week 2, 2006 to 13 April 2011.

Date search performed: September 2012.

Search strategy

	CINAHL via Ovid			CINAHL via EBSCO*host*			
	Keyword	Field	Results	Keyword	Field	Results for 1982–14 April 2014	Results for 1982–February 2006
1	Cleft Palate	Thesaurus	686	Cleft Palate	Thesaurus	1436	751
2	Cleft Lip	Thesaurus	526	Cleft Lip	Thesaurus	1099	567
3	cleft$ adj3 lip$	Text Word (.tw.)	495	Cleft* N3 lip*	TI, AB,IN	995	530
4	cleft$ adj3 palat$	Text Word (.tw.)	617	Cleft* N3 palat*	TI, AB,IN	1232	663
5	hare lip$	Text Word (.tw.)	1	hare lip*	TI, AB,IN	1	1
6	harelip$	Text Word (.tw.)	0	Harelip*	TI, AB,IN	0	0
7	Palatoschisis	Text Word (.tw.)	0	Palatoschisis	TI, AB,IN	1	1
8	orofacial$ adj3 cleft$	Text Word (.tw.)	29	Orofacial* N3 cleft*	TI, AB,IN	93	33
9	facial adj3 cleft$	Text Word (.tw.)	40	facial N3 cleft*	TI, AB,IN	69	40
10	oral adj3 cleft$	Text Word (.tw.)	49	oral N3 cleft*	TI, AB,IN	115	50
11	craniofacial adj3 cleft$	Text Word (.tw.)	47	craniofacial N3 cleft*	TI, AB,IN	74	45
12	or/1-11		817	or/1-11		1741	889
13	Middle Ear Ventilation	Thesaurus	224	Middle Ear Ventilation	Thesaurus	410	264
14	exp Otitis Media	Thesaurus	1259	exp Otitis Media	Thesaurus	2403	1467
15	grommet$	Text Word (.tw.)	21	grommet*	TI, AB,IN	49	28
16	ear$ adj3 ventilat$	Text Word (.tw.)	80	ear* N3 ventilat*	TI, AB,IN	158	75
17	otitis media	Text Word (.tw.)	898	otitis media	TI, AB,IN	1646	1104
18	ventilat$ adj3 tube$	Text Word (.tw.)	99	ventilat* N3 tube*	TI, AB,IN	266	166
19	tympanostom$	Text Word (.tw.)	90	tympanostom*	TI, AB,IN	171	110
20	glue adj3 ear$	Text Word (.tw.)	32	glue N3 ear*	TI, AB,IN	42	33
21	or/13-20		1634	or/13-20		3210	
22	21 and 12		17	21 and 12		36	
23	from 22 keep 1-17		17	from 22 keep 1-17			

EMBASE

URL: www.elsevier.com/online-tools/embase via Ovid search strategy – https://ovidsp.ovid.com/

Date range searched: 2006 to 14 April 2011.

Date search performed: 14 April 2011.

Search strategy

1. cleft lip/ (3181)
2. Cleft Palate/ (5513)
3. Cleft Lip Face Palate/ (257)
4. Cleft Lip Palate/ (931)
5. (cleft$ adj3 lip$).tw. (4006)
6. (cleft$ adj3 palat$).tw. (5885)
7. hare lip$.tw. (23)
8. harelip$.tw. (31)
9. Palatoschisis.tw. (25)
10. (orofacial$ adj3 cleft$).tw. (243)
11. (facial adj3 cleft$).tw. (675)
12. (oral adj3 cleft$).tw. (290)
13. (craniofacial adj3 cleft$).tw. (259)
14. or/1-13 (9179)
15. middle ear ventilation/ (238)
16. exp Otitis Media/ (13,078)
17. tympanostomy tube/ (1111)
18. grommet$.tw. (305)
19. (ear$ adj3 ventilat$).tw. (901)
20. otitis media.tw. (9424)
21. (ventilat$ adj3 tube$).tw. (1075)
22. tympanostom$.tw. (640)
23. (glue adj3 ear$).tw. (216)
24. or/15-23 (16372)
25. 14 and 24 (206)
26. from 25 keep 1-206 (206)

MEDLINE

URL: www.nlm.nih.gov/bsd/pmresources.html via Ovid search strategy – https://ovidsp.ovid.com/

Date range searched: 2006 to 14 April 2011.

Date search performed: 14 April 2011.

Search strategy

1. Cleft Palate/ (11,628)
2. Cleft Lip/ (8131)
3. (cleft$ adj3 lip$).tw. (6156)
4. (cleft$ adj3 palat$).tw. (8901)
5. hare lip$.tw. (68)
6. harelip$.tw. (99)
7. Palatoschisis.tw. (69)
8. (orofacial$ adj3 cleft$).tw. (280)
9. (facial adj3 cleft$).tw. (759)
10. (oral adj3 cleft$).tw. (326)
11. (craniofacial adj3 cleft$).tw. (213)
12. or/1-11 (15,658)
13. Middle Ear Ventilation/ (1469)
14. exp Otitis Media/ (15,239)
15. grommet$.tw. (336)
16. (ear$ adj3 ventilat$).tw. (616)
17. otitis media.tw. (10,785)
18. (ventilat$ adj3 tube$).tw. (1163)
19. tympanostom$.tw. (651)
20. (glue adj3 ear$).tw. (273)
21. or/13-20 (18,849)
22. 21 and 12 (297)
23. exp animals/ not human/ (2,948,249)
24. 22 not 23 (286)
25. 22 not 24 (11)
26. from 25 keep 1-11 (11)

Appendix 5 Examples of invitations sent to parents and children via the Cleft Lip & Palate Association newsletter and social media

Web Version | Update preferences | Unsubscribe Like Tweet
Forward

Glue Ear Treatment - What Matters to YOU?

Would you like to take part in a survey to help decide what the important results of treatment for glue ear are?

If you are an adult with cleft palate or a parent of a child with cleft palate you can find out more and share your opinions using the link below.

Parents: if your child is aged between 7 and 16 we also want to know their opinions about what results of treatment for glue ear are important. For them to take part you will need to agree at the end of the survey, they may also need your help to complete the survey.

To find out more and to take part please click here >>

Registered Charity England & Wales (1108160) and Scotland (SC041034)

Edit your subscription | Unsubscribe

[redacted]

Appendix 6 Examples of invitation and reminder e-mails sent to health professionals to take part in the Delphi survey

Round 1 – Invitation email

Developing a core outcome set for children with cleft palate and otitis media with effusion.

Dear First Name

Thank you for your interest in our study.

Core outcome sets represent the minimum that should be measured and reported in all clinical trials of a specific condition. Currently there is no core outcome set for studies of OME in children with cleft palate which may hinder the comparison of the effectiveness of interventions across trials.

The aim of this study is to find out which outcomes are important to clinicians and which outcomes should be included in a core outcome set for use in future trials in children with cleft palate and OME.

This questionnaire is the first round of three rounds of the survey.

We would like you to complete this questionnaire within three weeks (by the dd-mm-yy).

Once we have received responses from all panellists we will collate and summarise the findings and prepare the second questionnaire. Your responses in each round will be anonymous.

It is very important that you complete the questionnaires in each round. The reliability of the results could be compromised if people drop out of the study before it is completed, because they feel that the rest of the group does not share their opinions. If people drop out because they feel their opinions are in the minority, the final results will overestimate how much the sample of participants agreed on this topic.

All responses will be anonymous. However, to help us track completion of each round

we would like you to initially register using your email address. Upon registration you will be allocated a unique identifier which will be used to process all data anonymously.

To access the survey please click on the following link: https://mcrnctu.org.uk/MomentDelphi/Login.aspx

If you are unable to enter the study by double clicking the link in this email, please right click on the link and use . This will enable you to paste the URL into your browser

Should you have any questions please contact: <contact details>

If you experience any difficulty accessing the online system please contact: <contact details>

Invitation to round 2

Developing a core outcome set for children with cleft palate and otitis media with effusion.

Dear FirstName

Thank you for your response to round 1 of the MOMENT study (Identifying a core outcome set for the management of otitis media with effusion).

We can now share with you the results from round 1 from your peer group in this second round.

It is really important that we get round 2 responses from as many people as possible who took part in round 1 so that all opinions can be considered. At the moment, only 37% overall of those who took part in round 1 have responded in round 2 and we really need your help to improve this figure.

Round 2 is available to complete until the dd-mm-yy.

The survey can be found at: https://mcrnctu.org.uk/MomentDelphi/Login.aspx

To log in you will need your unique identifier from round 1, your unique identifier is: MOnnnnnn

Should you have any questions please contact: <contact details>

If you experience any difficulty accessing the online system please contact: <contact details>

Invitation to round 3

STOP PRESS - ALL RESULTS NOW AVAILABLE FOR VIEWING

Dear First Name
Thank you for your responses to round 2 of the Delphi survey..

In this third and final round we would like to share with you the results from round 2. You will be shown the results from **all** stakeholder groups including health professionals, children with cleft palate and results of parents of children with cleft palate..

You will also be able to view your score from round 2, and to change it should you wish to do so after seeing others' responses

We do hope that you will be able to help with this **final stage of the online Delphi survey**. As a thank you, all those completing this third and final round will be entered into a prize draw to **win a bottle of champagne**!

To take part please go to: https://mcrnctu.org.uk/MomentDelphi/Login.aspx
Your unique identifier is: M0nnnnnn
your email address: _myEmail

The survey will close on the dd-mm-yy

If you are using a mac please avoid using the back button, if you use the back button inadvertently please check your summary of scores carefully before submitting

Best wishes

Nicola Harman, on behalf of the MOMENT study team

Should you have any questions please contact: <contact details>
If you experience any difficulty accessing the online system please contact: <contact details>

Example – reminder email round 3

STOP PRESS - PARENTS' AND CHILDREN'S OPINIONS NOW AVAILABLE FOR VIEWING

> DEADLINE EXTENDED to dd-mm-yy

Dear First Name,

We are pleased to be able to share with you the results from **all** stakeholder groups who have taken part in MOMENT including **children with cleft palate, parents of children with cleft palate and all health professionals.**

At present **only X%** of those who completed round 1 and 2 have given their final opinion and we need your help to ensure that we don't introduce bias from one particular group of clinicians.

Your opinion on important outcomes is very valuable and we want you to have your say, the difference in round 3 is that you can now see the results from all clinicians, parents and children to help you decide on what score to give.

We do hope that you will be able to help with this **final stage of the online Delphi survey.**

As an extra thank you, all those completing this third and final round will be entered into a prize draw to **win a bottle of champagne**!

To take part please go to: https://mcrnctu.org.uk/MomentDelphi/Login.aspx .

Your unique identifier is: M0nnnnnn
your email address: _myEmail

The survey takes about 15-20 minutes to complete.

To ensure you have time to give your opinion we have extended the survey to the dd-mm-yy. If you have experienced any difficulties completing the survey please get in touch so that we can help find a solution.

Thank you in anticipation of your ongoing support of the MOMENT study

Nicola

(cleftcollective@manchester.ac.uk)
Should you have any questions please contact: <contact details>

If you experience any difficulty accessing the online system please contact: <contact details>

Appendix 7 Documents provided to participants of the consensus meeting for core outcome set development

E-mail content

Dear <name>,

Thank you for agreeing to take to part in the MOMENT study event for the development of a core outcome set on the 6th March in Manchester.

The event will take place at the MacDonald Manchester Hotel and Spa, details of how to get the hotel are available on the hotel website: http://www.macdonaldhotels.co.uk/our-hotels/macdonald-manchester-hotel-spa/useful-information/

I have attached three documents for you to have a look at before the meeting, these are:

1. A summary of the day outlining who is involved and what the aims of the day are
2. A summary from an organisation called COMET describing what core outcome measures are.
3. An agenda for the day.

In the agenda there are two slots, one before and one after lunch, where we will present back to you the results of the surveys of health professionals, parents and children. These results will show how important people thought each of the outcomes of treatment for glue ear were.

In these results sessions we will ask you to think about the results we present and if you agree with them. We will ask everyone at the meeting, including those who have already completed the surveys, to score each of the outcomes. For some of these outcomes there may be some discussion needed within the group. For example, if it appears that there are large differences in opinions of how important an outcome is.

*<We would like to welcome you to our meeting at **10.30am** to meet with the team so that we can give you an overview of core outcome measures, introduce you to the study team and answer any questions that you might have.> – provided to parents only*

If you would like us to book your travel arrangements and/or overnight accommodation and we have yet to confirm the arrangements with you please contact Claire (email address) who will be able to help with the booking. It would also be helpful if you could let us know whether you have any dietary requirements and if you are bringing along a friend or relative so that we know final numbers.

The MOMENT study team would like to take this opportunity to thank you for your interest in the study and the core outcome set meeting. We greatly value the contribution of parents, patient representatives and health care professionals in this study and look forward to meeting you on the 6th March.

Best wishes

Nicola

On behalf of the MOMENT study team.

Documents provided with e-mail

COMET plain language summary, URL: www.comet-initiative.org/resources/PlainLanguageSummary (accessed 9 April 2014).

mOMEnt consensus meeting agenda V1.0 24 February 2014

mOMEnt consensus meeting summary V2.0 19 February 2014

mOMEnt
Management of Otitis Media with Effusion in childreN with ClefT Palate

Study event for the development of a core outcome set

6th March 2014.

Meeting room 11, MacDonald Manchester Hotel and Spa, London Road, Manchester, M1 2PG Tel 0161 272 3200.
Contact on the 6th March 2014—Nicola Harman
Hotel information: http://www.macdonaldhotels.co.uk/our-hotels/macdonald-manchester-hotel-spa/useful-information/

Agenda

	Arrival and registration (*refreshments on arrival).—Breakout space*
10.30	*Parallel session for parents/patient representatives with Heather Bagley (Patient and Public Coordinator) - 10.30-11.00 Meeting room 11*
11.00	Welcome and introductions
11.30	Introduction to the MOMENT Study
11.45	Introduction to core outcome sets and plan for the day
12.00	Methods—what we did and how we did it.
12.15	Results from qualitative interviews with parents and children
12.30	Results from the Delphi survey of health professionals and online surveys of parents, children and young persons. Part 1.
13.00	*Lunch— Hotel restaurant*
13.45	Results from the Delphi survey of health professionals and online surveys of parents, children and young persons. Part 2.
2.30	*Coffee Break—breakout space*
2.45	Summary
3.15	What happens next
3.45	Meeting close

MOMENT Consensus meeting agenda V1.0

mOMEnt
Management of Otitis Media with Effusion in childreN with CleFT Palate

Study event for the development of a core outcome set

Would you like to contribute more to the MOMENT study by using your experience to help with the final stages of the project?

Part of the MOMENT study involves the development of something called a core outcome set. An outcome is a result of treatment and can be thought of as the "what" to measure to find out if treatment has worked. The MOMENT study wants to find out what the most important outcomes/results of treatment are and should be included in future research.

Why do we need a core outcome set?

A core outcome set represents the minimum set of results that should be measured in future research. To decide which treatments are best for patients, it is important to look at the effect those treatments have on patients. Researchers do this by measuring an 'outcome.

By having a set of agreed outcomes that are "core" to all trials it will be easier to compare and combine results of studies so that all of the evidence available can be used to help determine if one treatment is better than another.

Event details

Date:
Thursday 6th March 2014

Venue:
MacDonald Manchester Hotel, London Road, Manchester.

Time:
11am-4pm ***(lunch provided)***

Who will be invited?

Parents of children with cleft palate and glue ear

Parents of children with cleft palate and glue ear who have taken part in qualitative interviews for the study, who have completed an online survey or who are on the CLAPA mailing list will receive an invitation to take part.

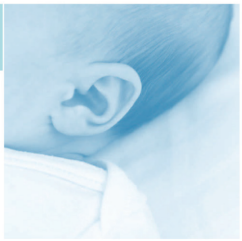
Health care professionals who provide care for children with cleft palate

Health care professionals who have taken part in all three rounds of an online survey and who have expressed an interest in taking part in further meetings about the core outcome set.

Cleft Surgeons | **ENT Surgeons** | **Specialist cleft nurses** | **Clinical Psychologists** | **Speech and Language therapists** | **Audiologists and audiological physicians**

MOMENT Consensus meeting summary V2.0 19-2-2014

What will I need to do?



If you decide to take part we would like you to attend the meeting in person and to join a discussion about which outcomes should be included in the core outcome set.

A list of 47 outcomes have already been scored by parents and health professionals. Each outcome was given a score from 1 (not at all important) to 9 (extremely important). At the meeting we will present the results for each of the outcomes and how important each group though it was. We will also send you these results before the meeting including a reminder of what you scored if you took part in the online survey.

At the meeting we will discuss each of the outcomes in turn to find out:

Do the results show that the outcome is felt to be important enough to be part of the core outcome set, and can those at the meeting confirm that

Do the results show that the outcome is not important and should not be part of the core outcome set, and can those at the meeting confirm that.

Where the results show that there are mixed feelings, we will discuss the outcome and vote, anonymously, on the inclusion or not of the outcome in the core outcome set.

What happens after the meeting?

The summary of the results and the summary of the meeting will be reviewed by two groups of people involved with the study, these are called the Study Advisory Group and an independent group called the Study Steering Committee. They will pay particular attention to the consistency of the results for each outcome to be included in the core outcome set. They will ensure the study results are presented transparently .

Useful Information

Travel expenses will be covered and if you prefer we are able to book your travel arrangements for you. Please note the current mileage rates are 40p for the first 150 miles of a round trip and 25p thereafter and we are able to cover up to the cost of standard train fare only. If you are attending as a parent of a child with cleft palate we are also able to cover the cost of any child care that you might need to attend the meeting. Travel (unless booked by ourselves) and childcare costs will be reimbursed following the meeting and expenses forms will be available to complete on the day.

What happens next?

If you would like to take part in the meeting please contact [redacted] to confirm your attendance .

Alternatively if you have any questions please contact [redacted]

Thank you for your support of the MOMENT Study!

MOMENT Consensus meeting summary V2.0 19-2

Appendix 8 Full responses to round 3 of the health professional Delphi survey and survey of parents and children with cleft palate

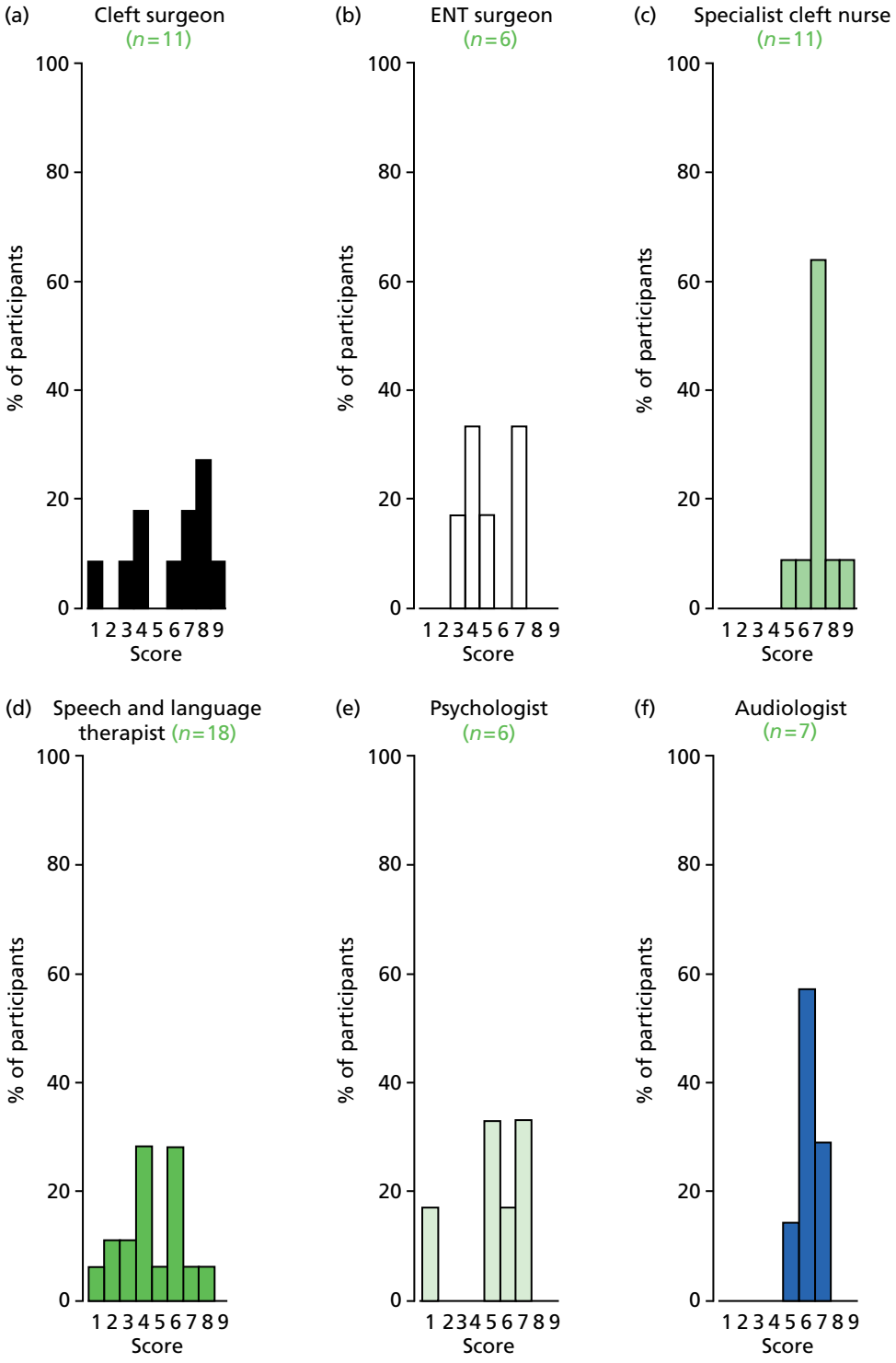


FIGURE 23 Outcome: tinnitus. (*continued*)

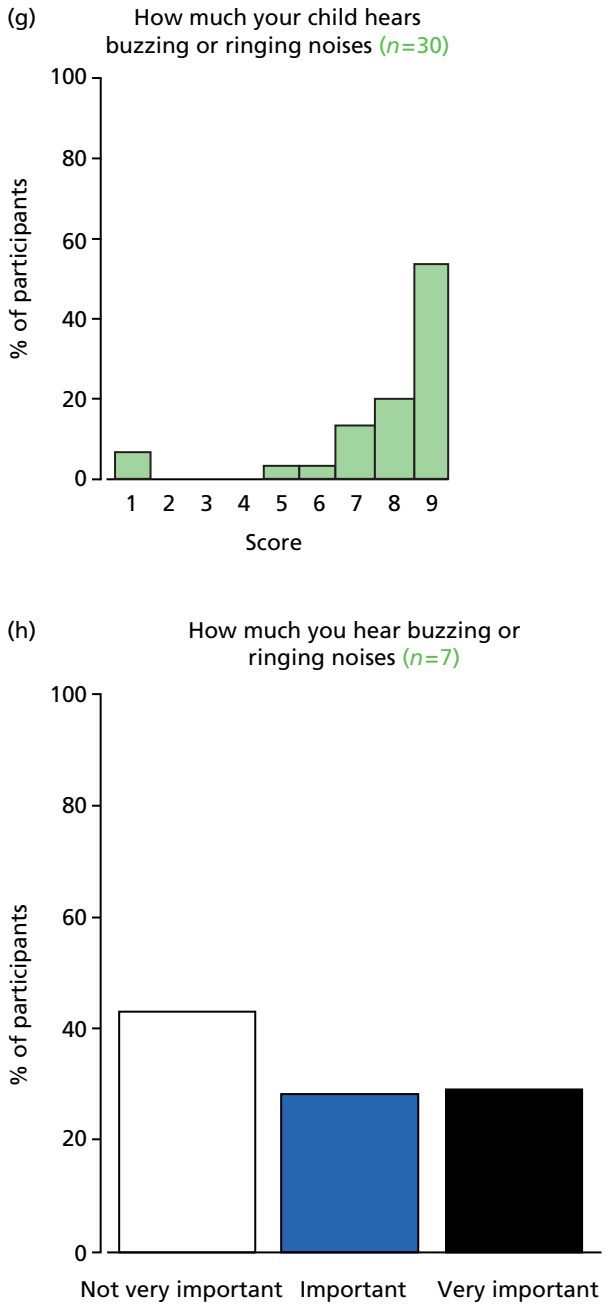


FIGURE 23 Outcome: tinnitus.

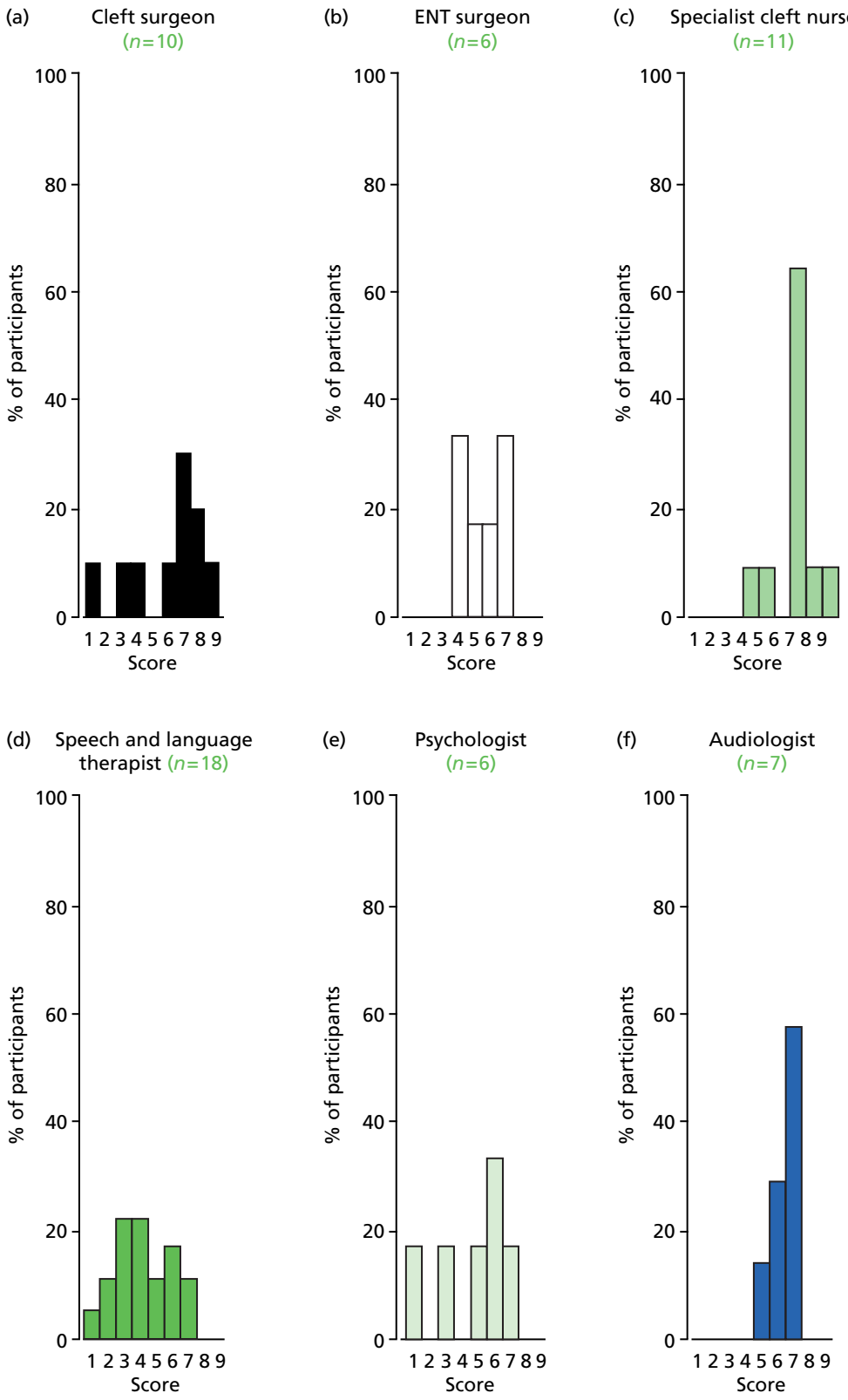


FIGURE 24 Outcome: vertigo. (*continued*)

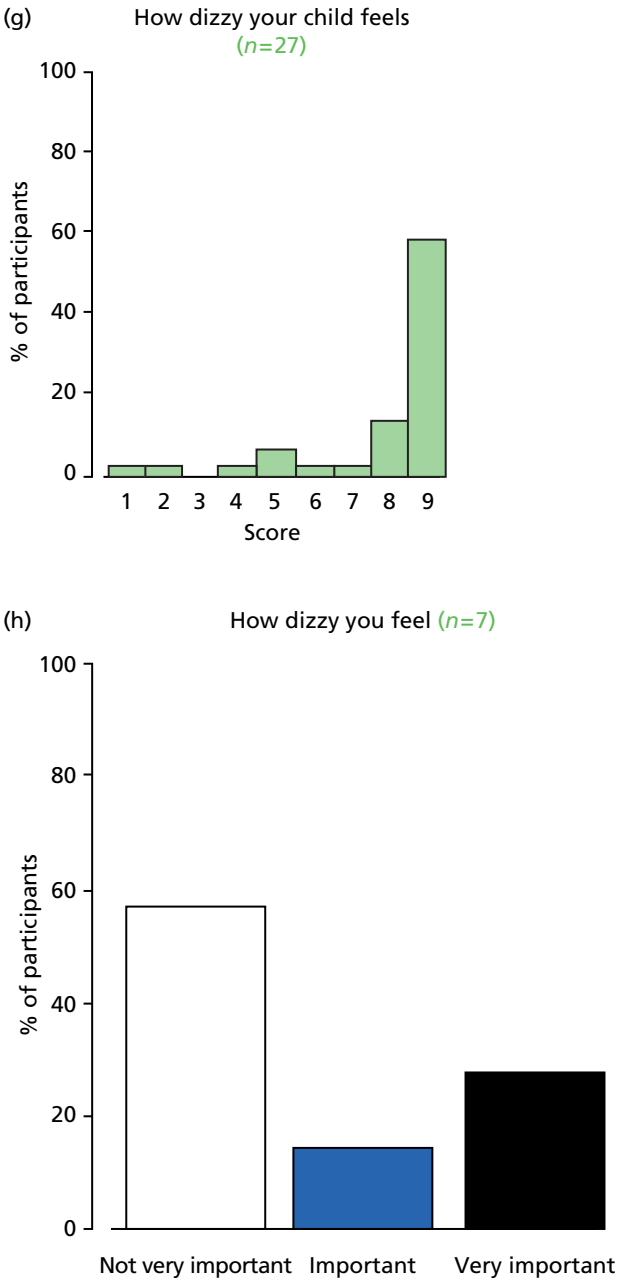


FIGURE 24 Outcome: vertigo.

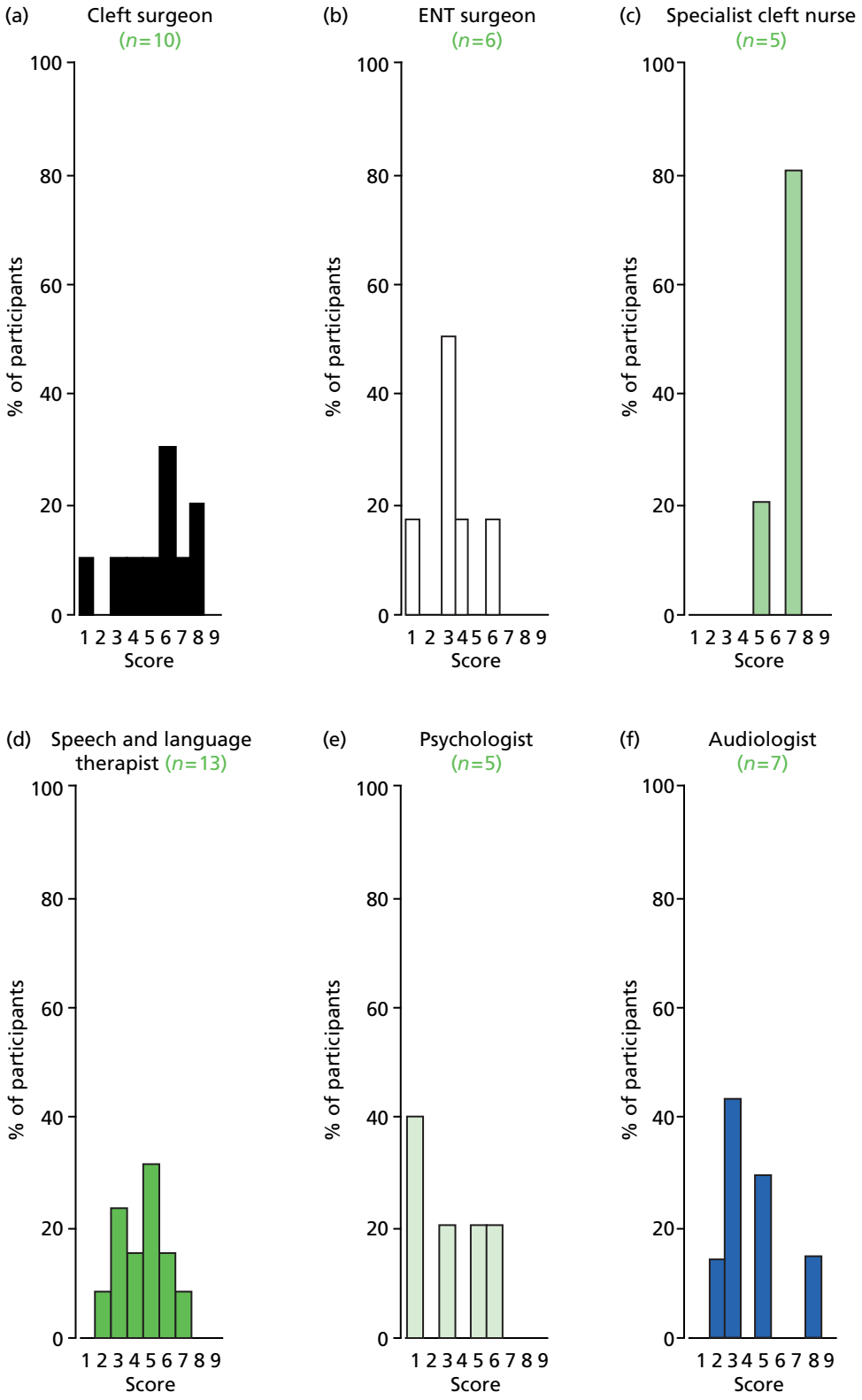


FIGURE 25 Outcome: stapedial reflex. (*continued*)

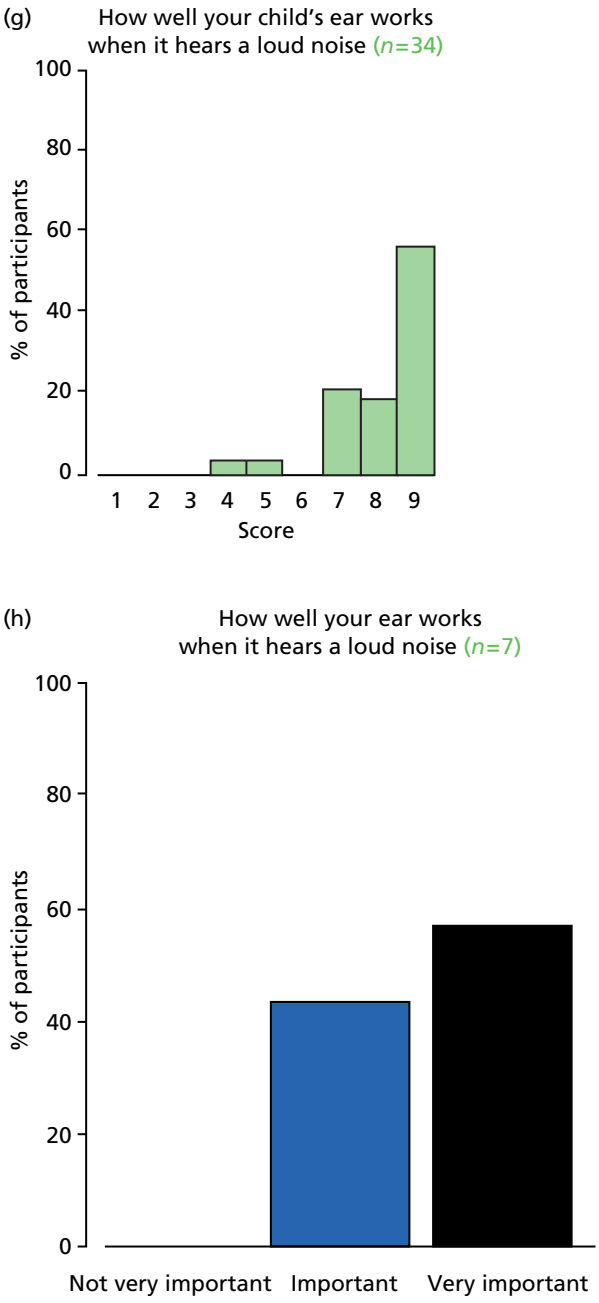


FIGURE 25 Outcome: stapedial reflex.

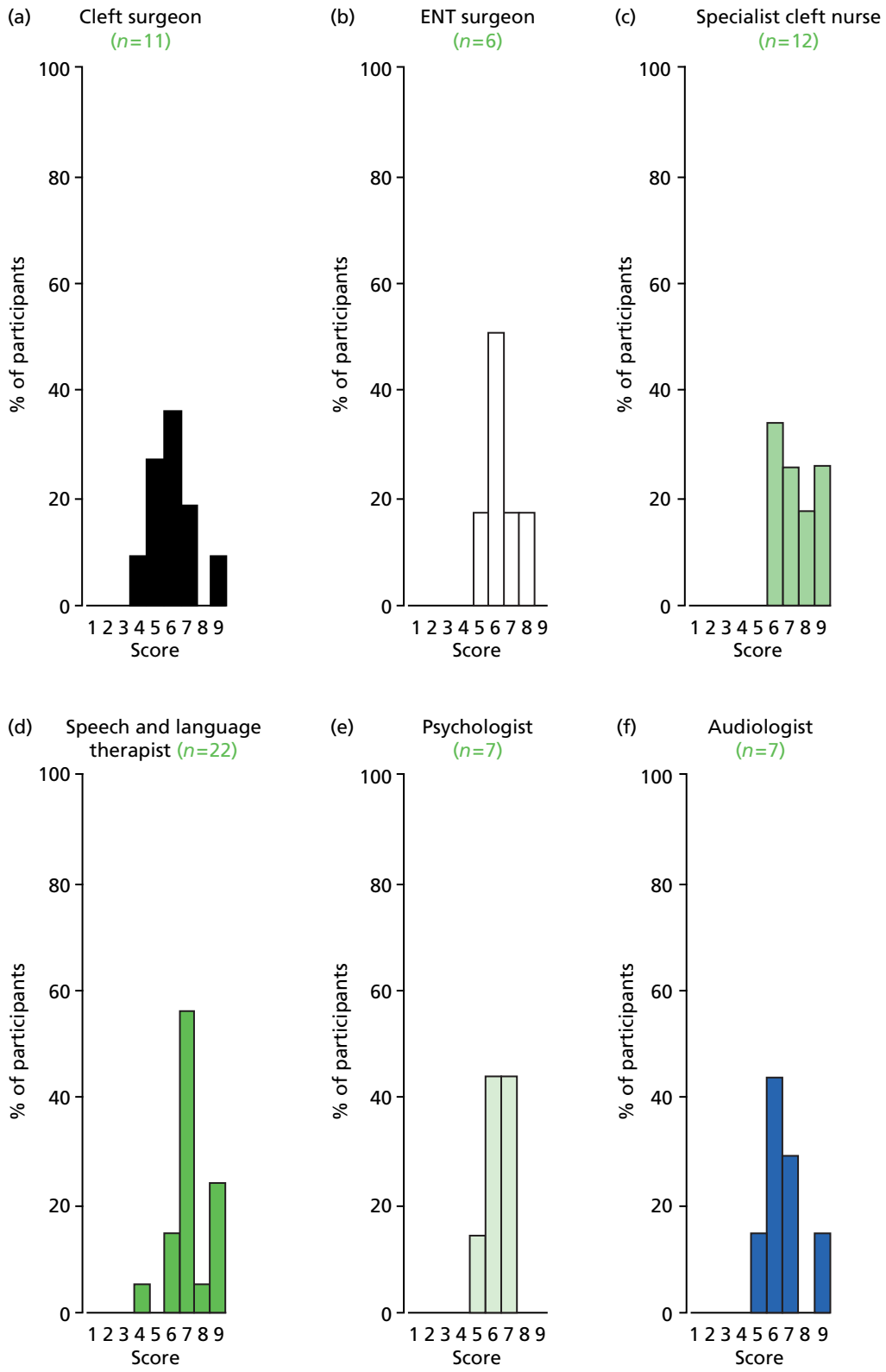


FIGURE 26 Outcome: parent's perspective of speech. (*continued*)

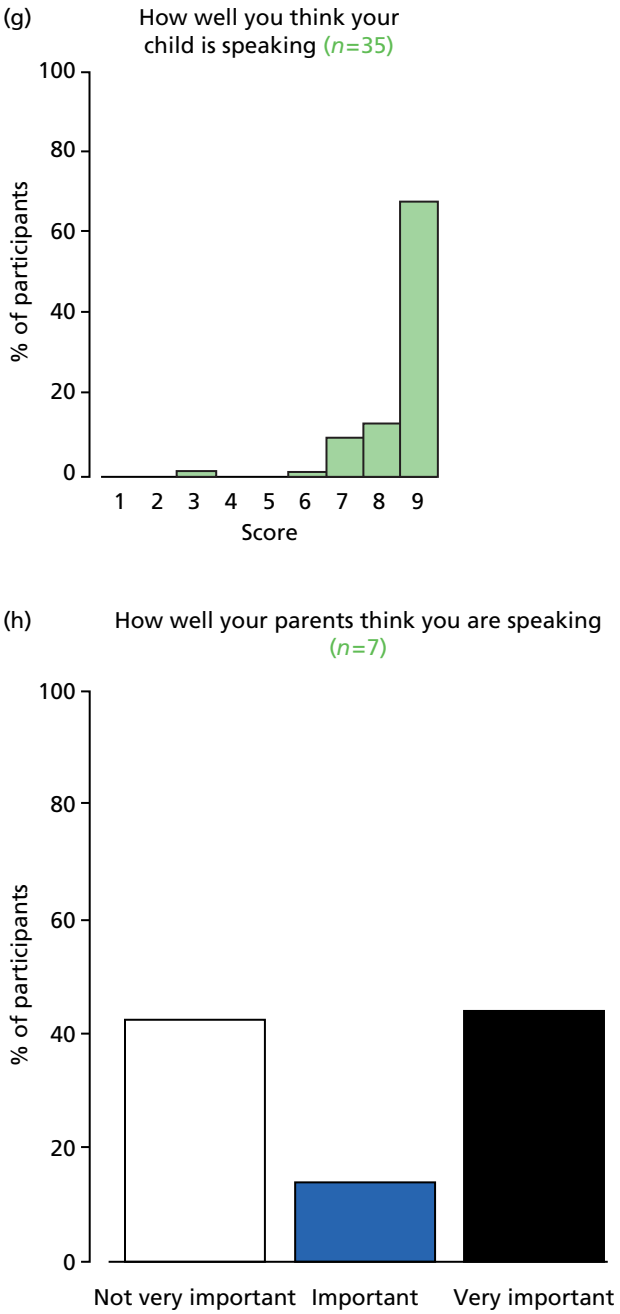


FIGURE 26 Outcome: parent's perspective of speech.

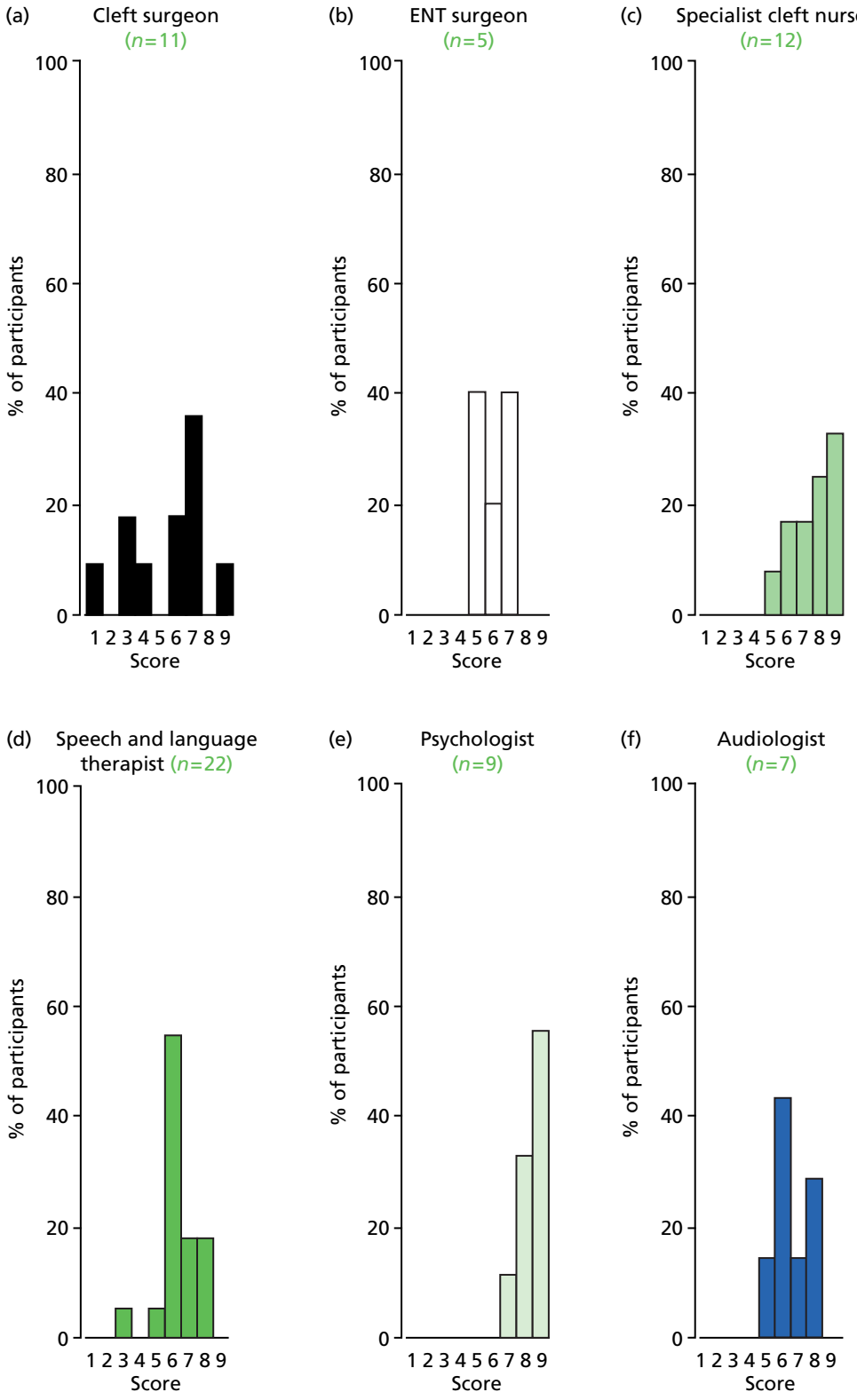


FIGURE 27 Outcome: parental stress. (*continued*)

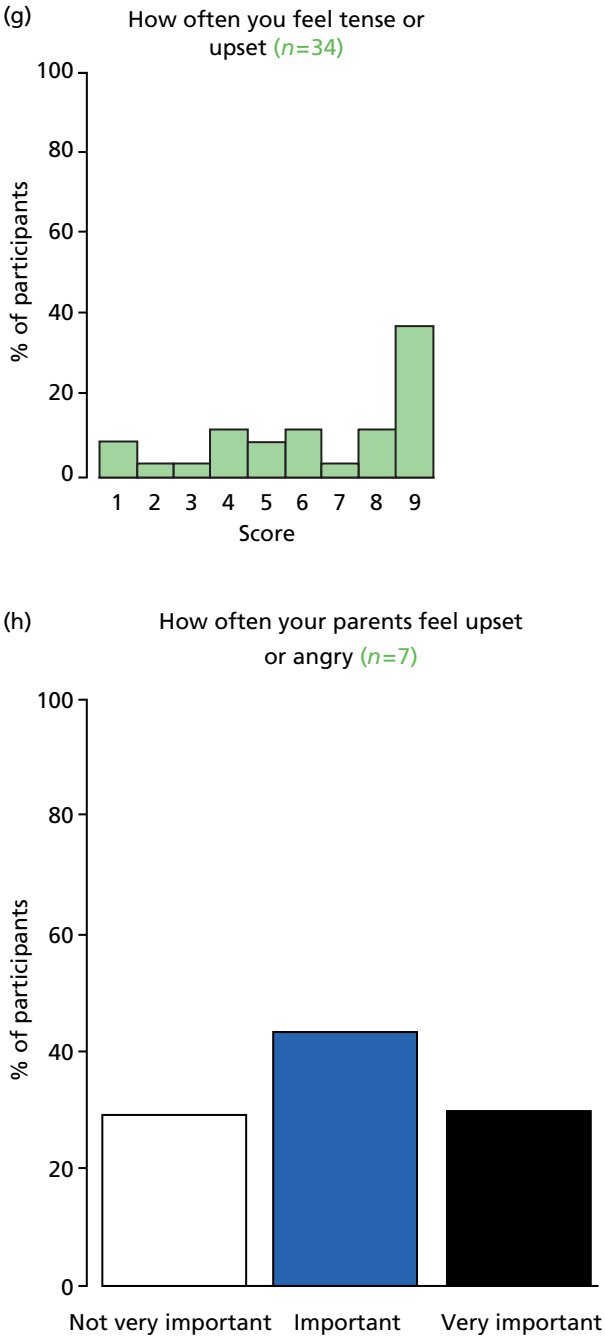


FIGURE 27 Outcome: parental stress.

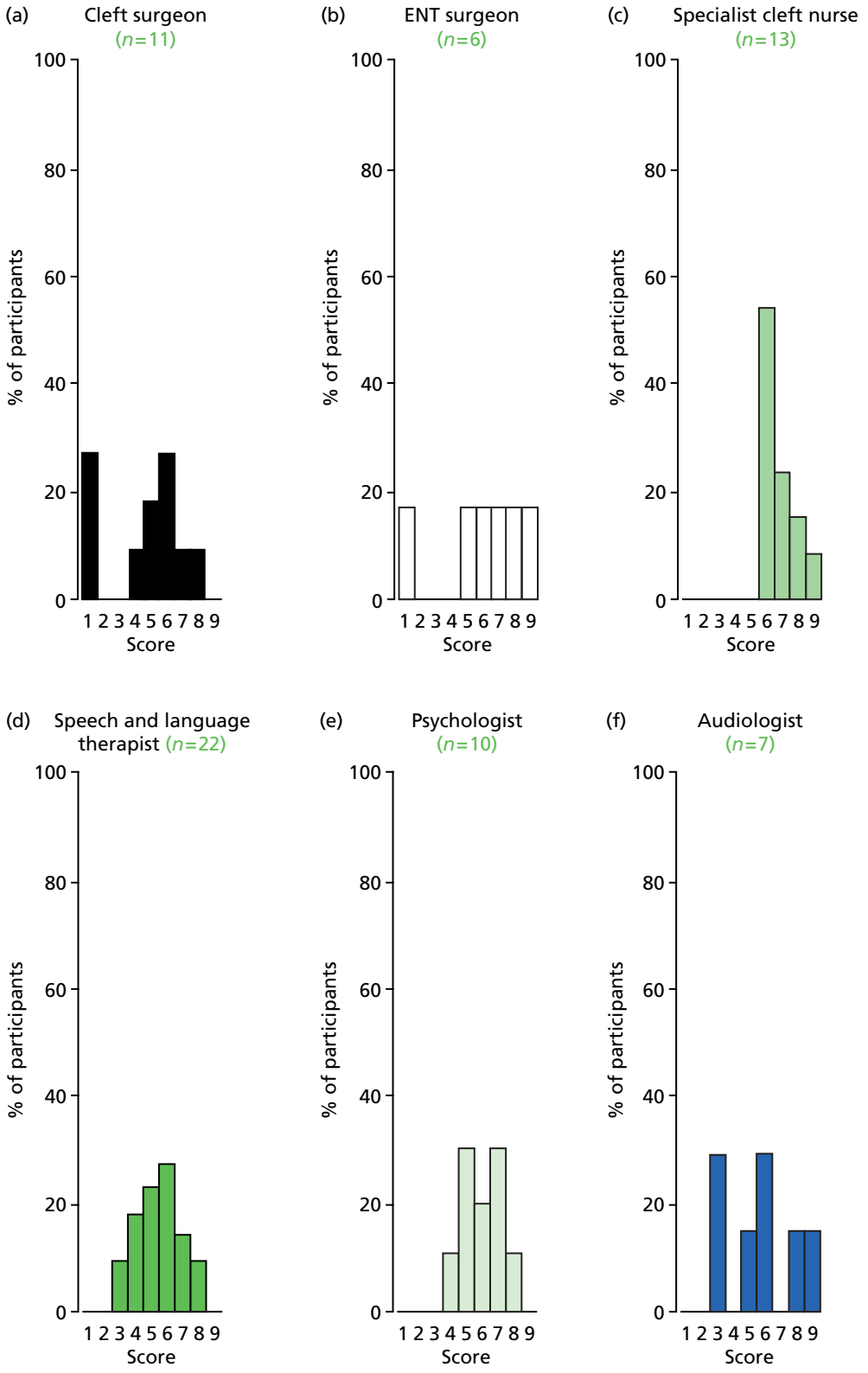


FIGURE 28 Outcome: intelligence. (*continued*)

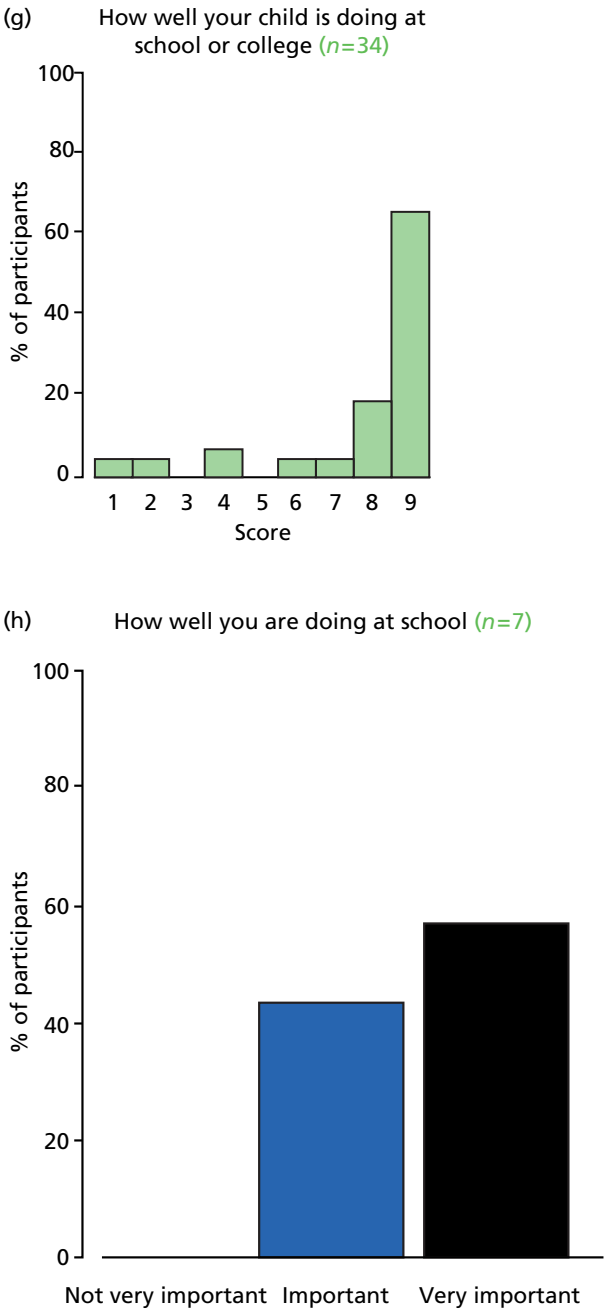


FIGURE 28 Outcome: intelligence.

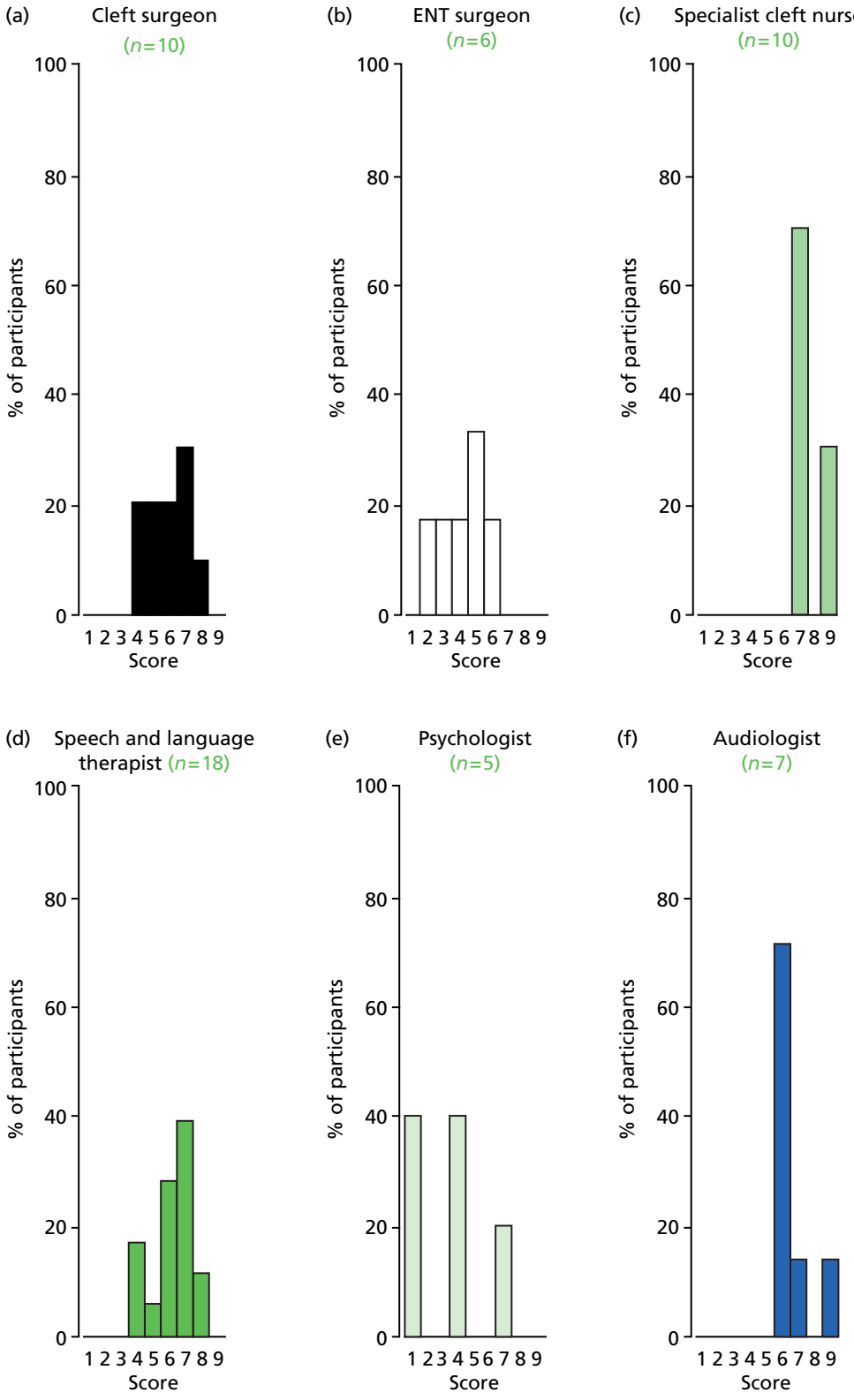


FIGURE 29 Outcome: temporary tympanic membrane perforation. (*continued*)

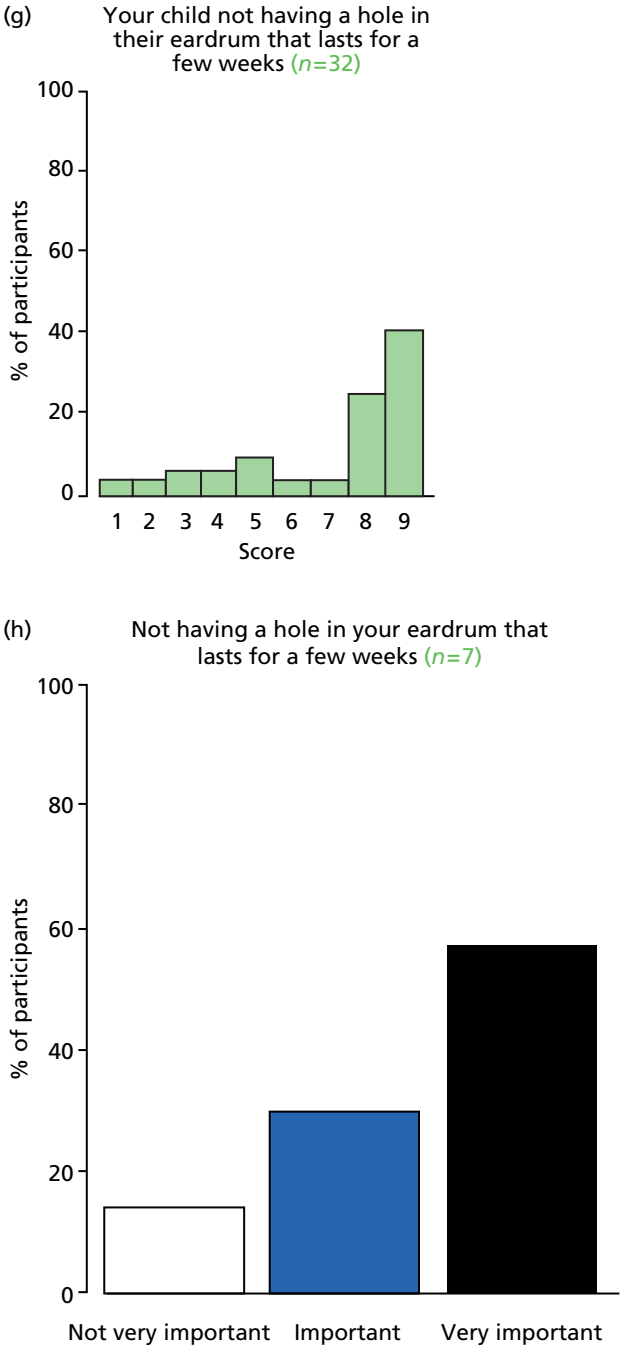


FIGURE 29 Outcome: temporary tympanic membrane perforation.

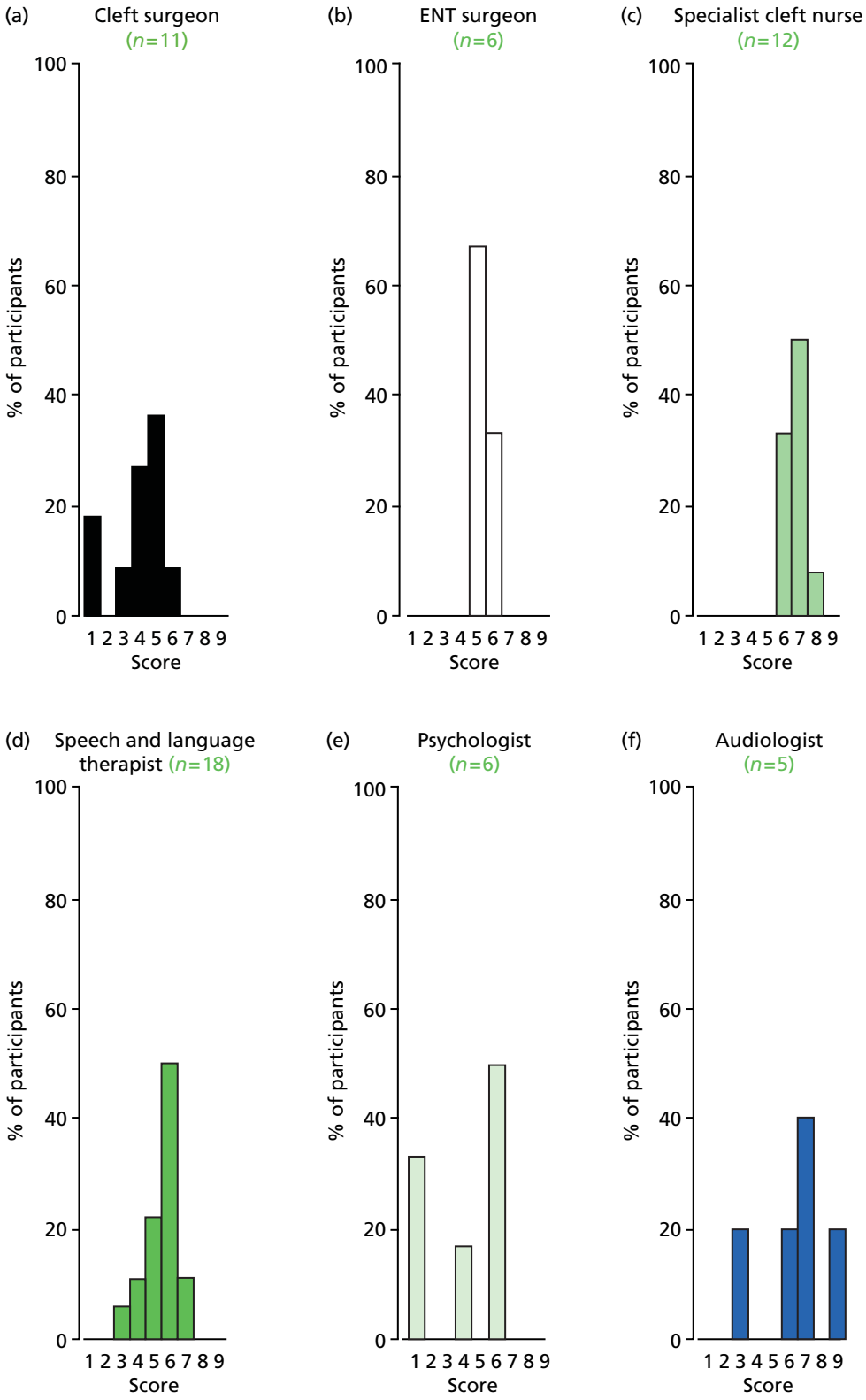


FIGURE 30 Outcome: upper respiratory tract infection. (*continued*)

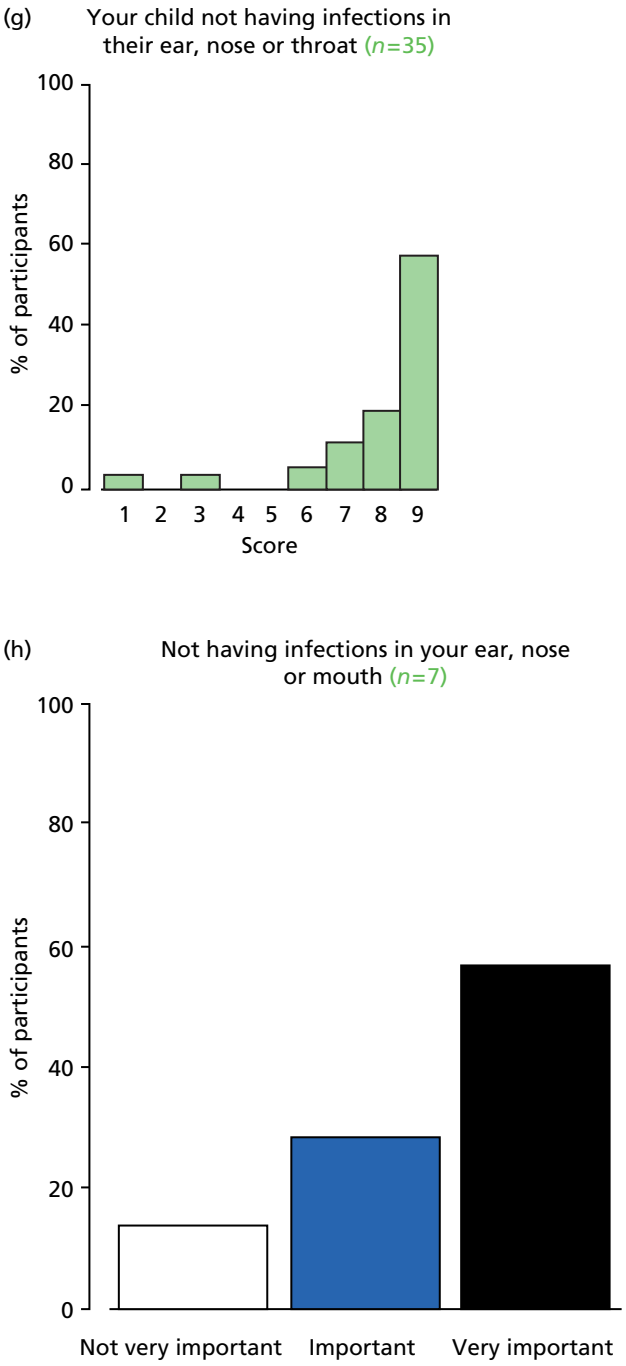


FIGURE 30 Outcome: upper respiratory tract infection.

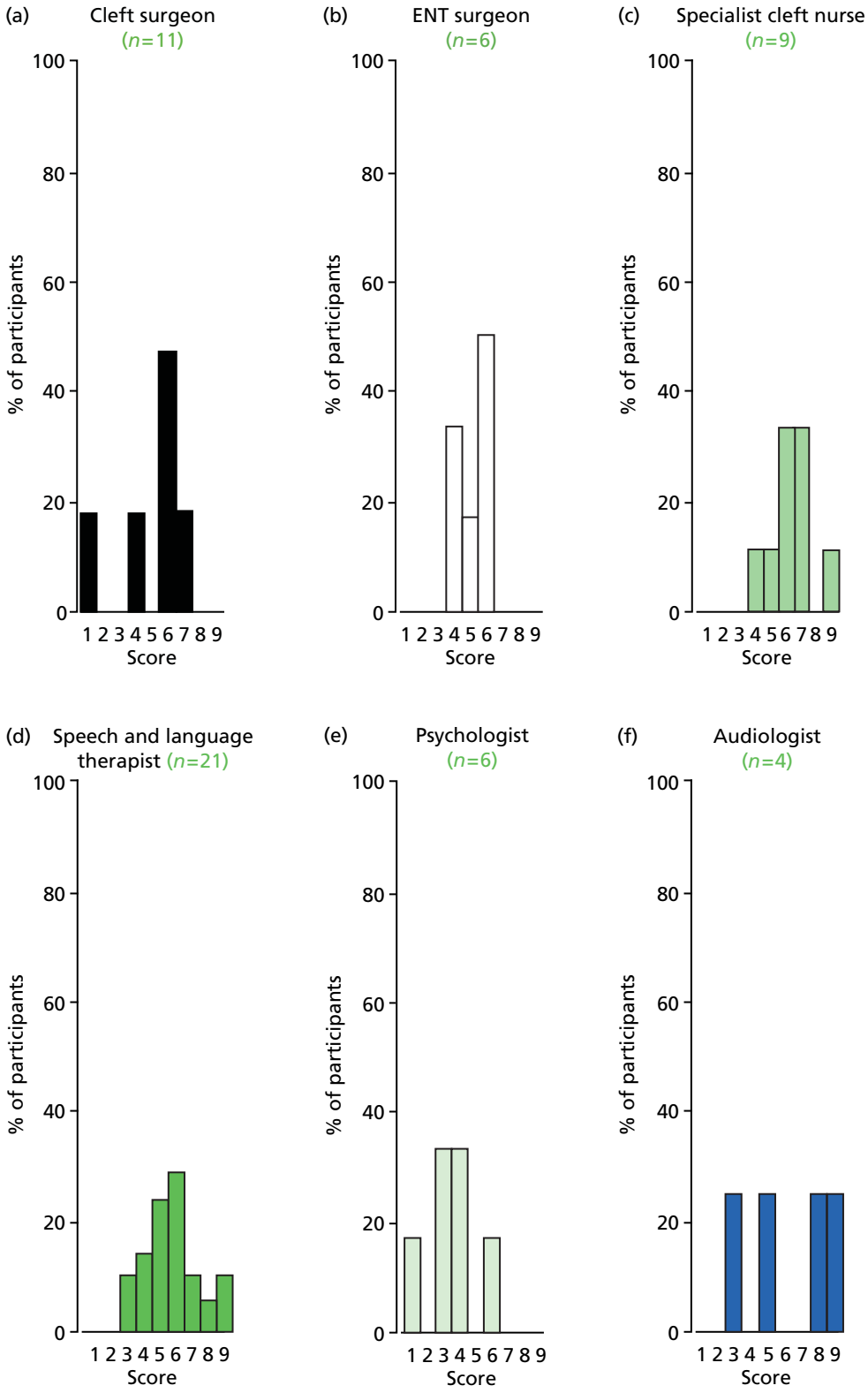


FIGURE 31 Outcome: nasal obstruction. (*continued*)

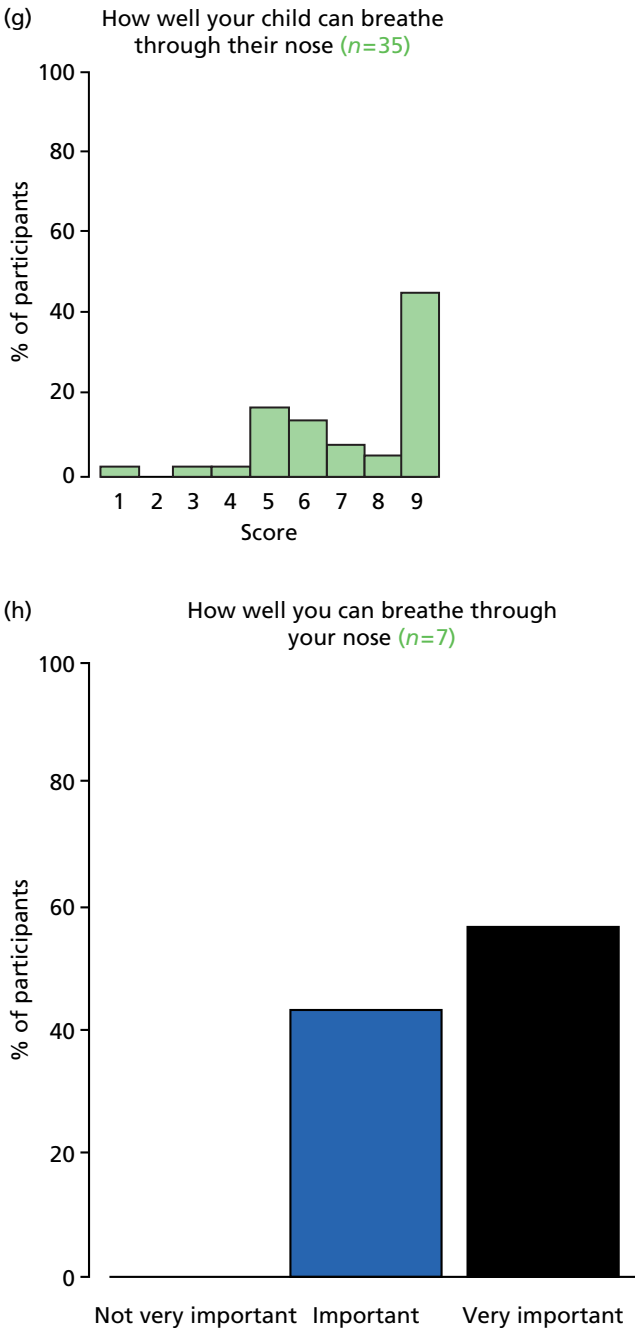


FIGURE 31 Outcome: nasal obstruction.

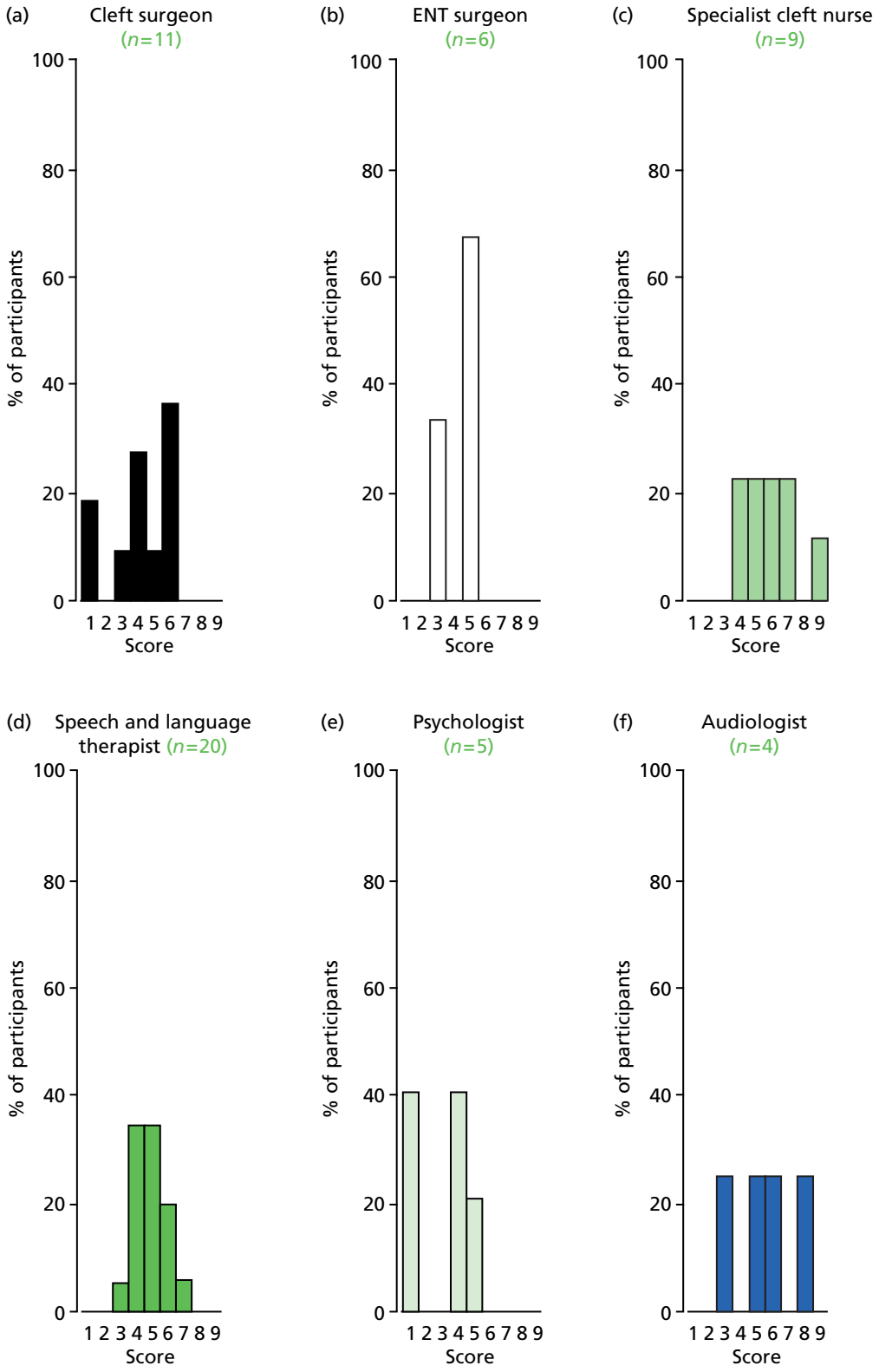


FIGURE 32 Outcome: rhinitis. (*continued*)

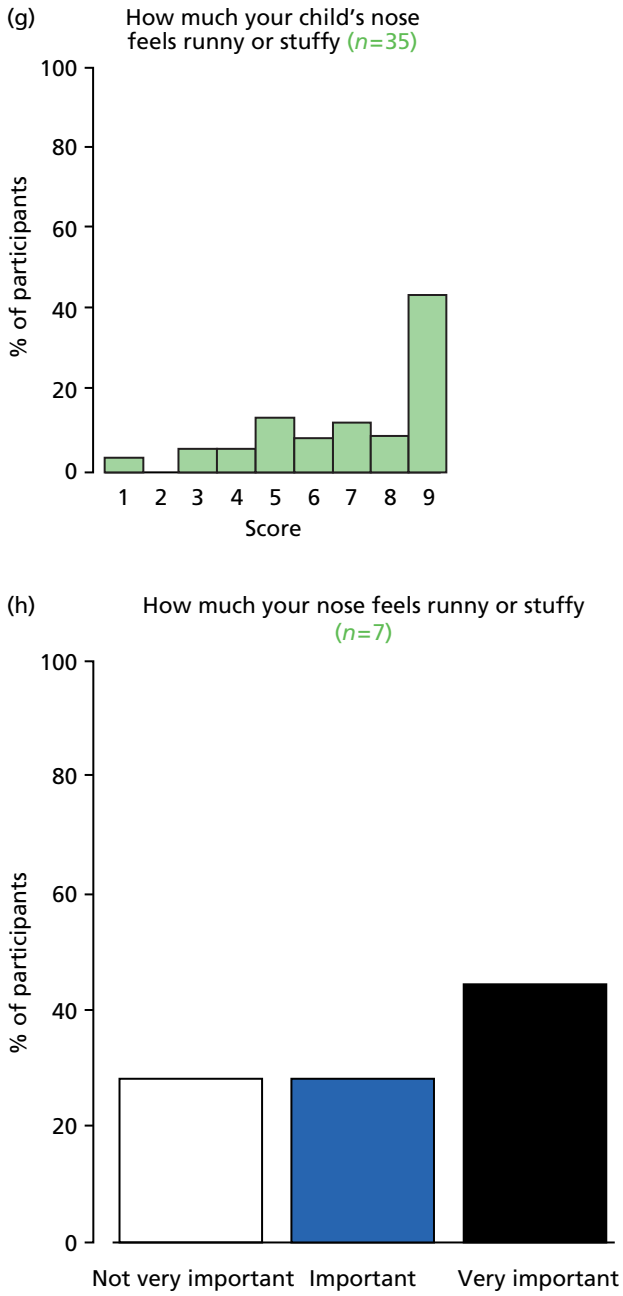


FIGURE 32 Outcome: rhinitis.

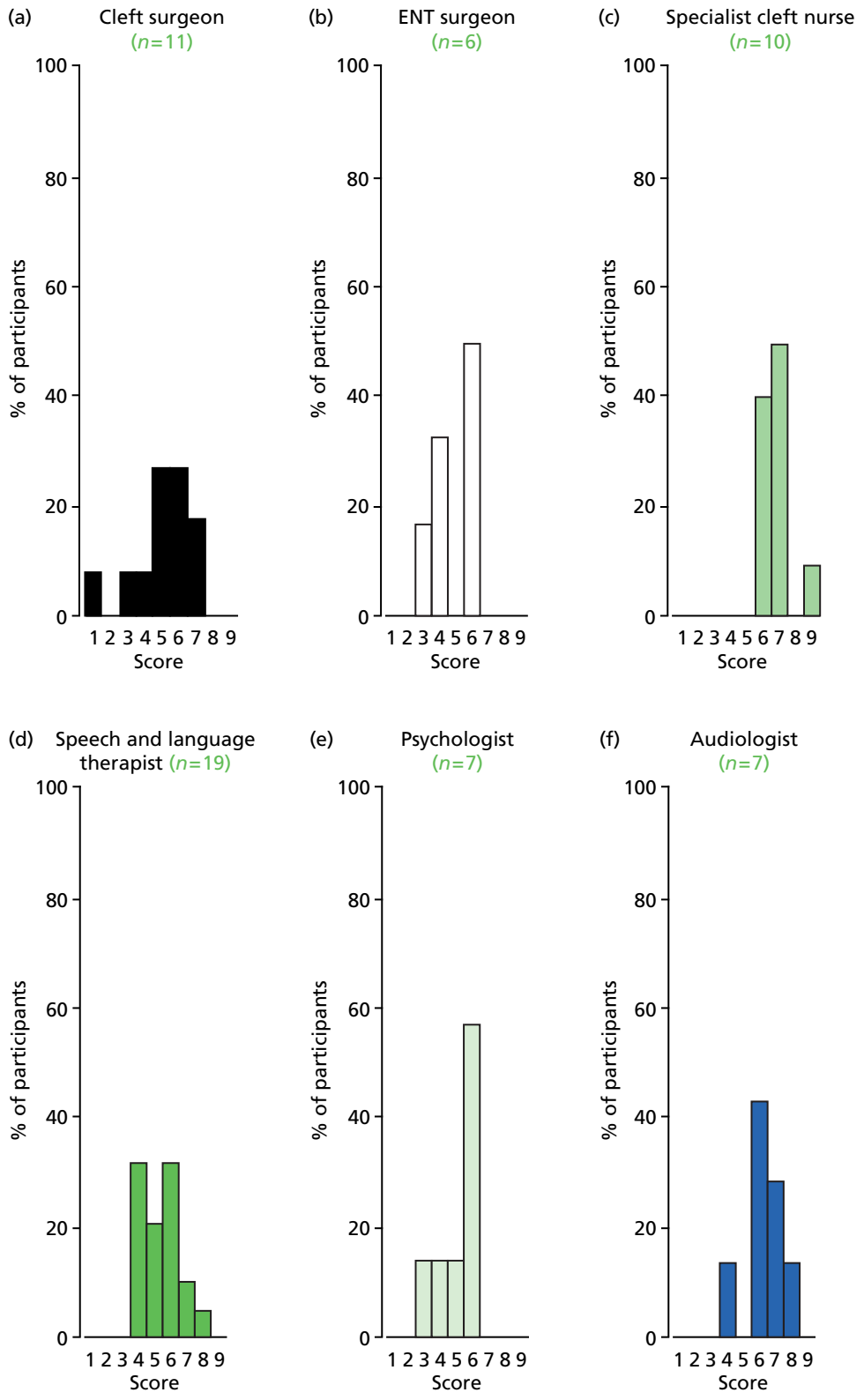


FIGURE 33 Outcome: hyperacusis.

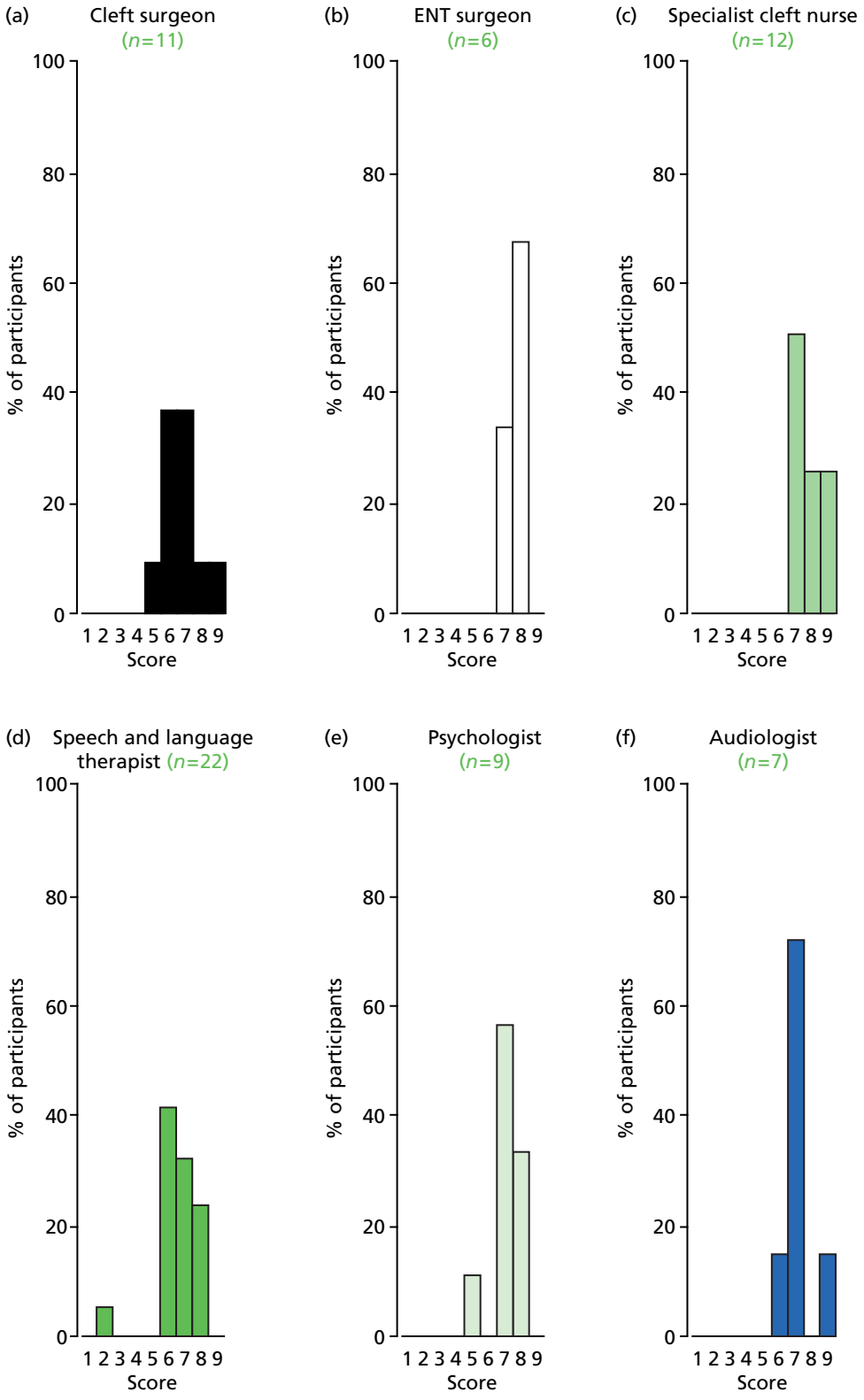


FIGURE 34 Outcome: parental satisfaction with treatment. (*continued*)

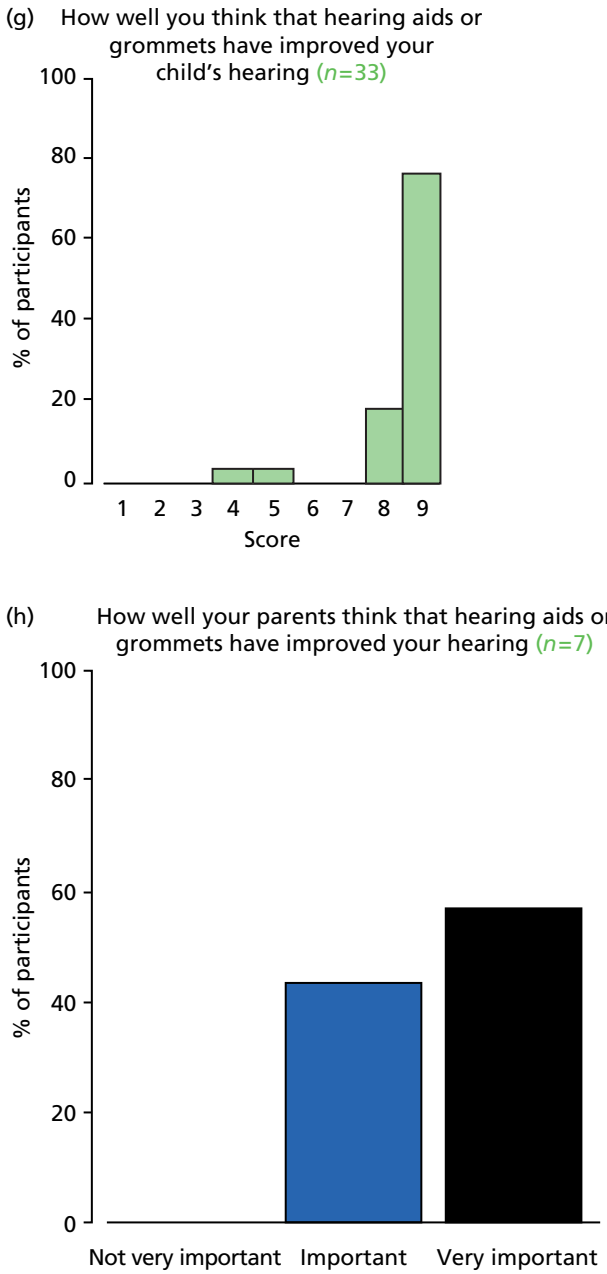


FIGURE 34 Outcome: parental satisfaction with treatment.

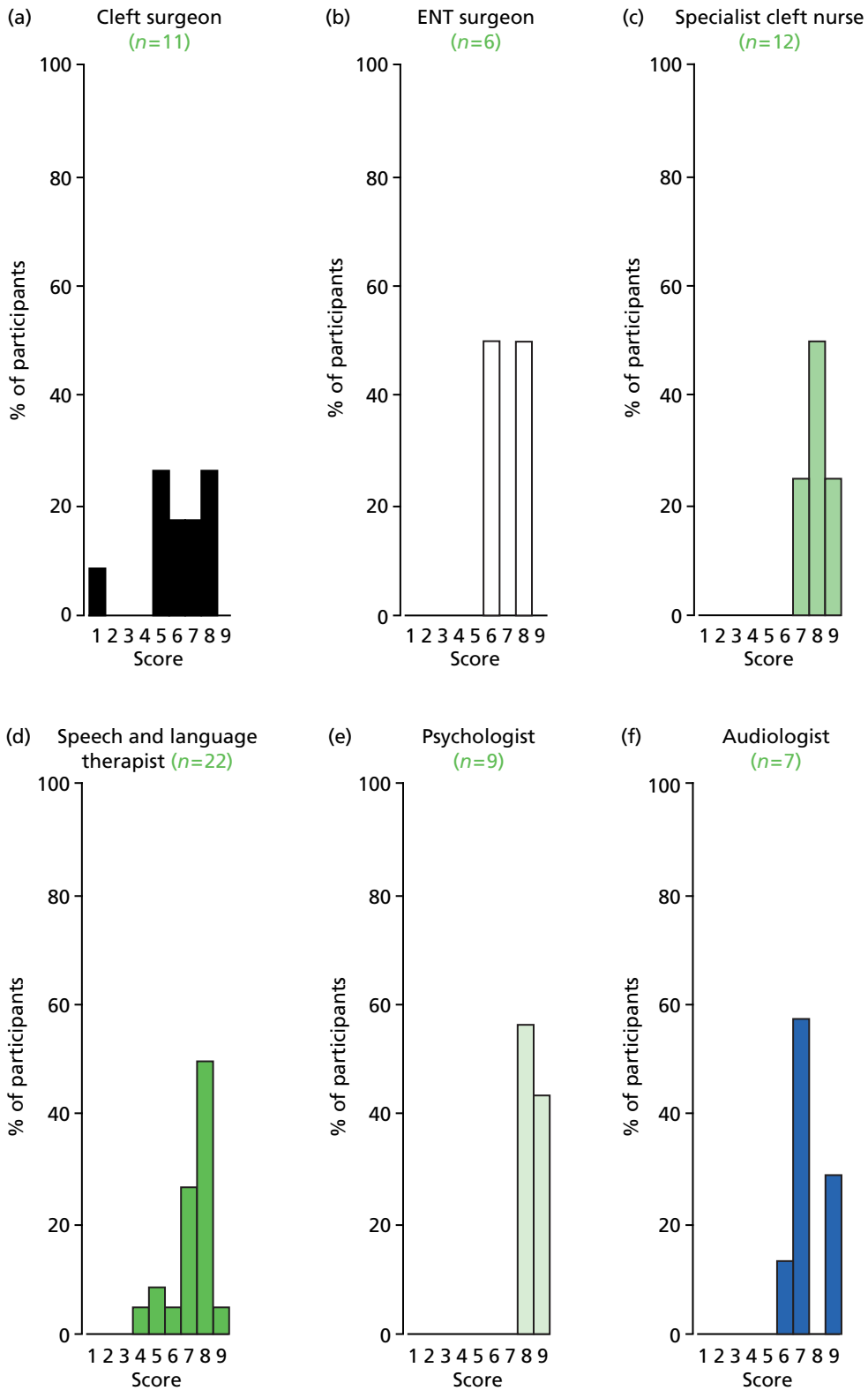


FIGURE 35 Outcome: child's satisfaction with treatment. (*continued*)

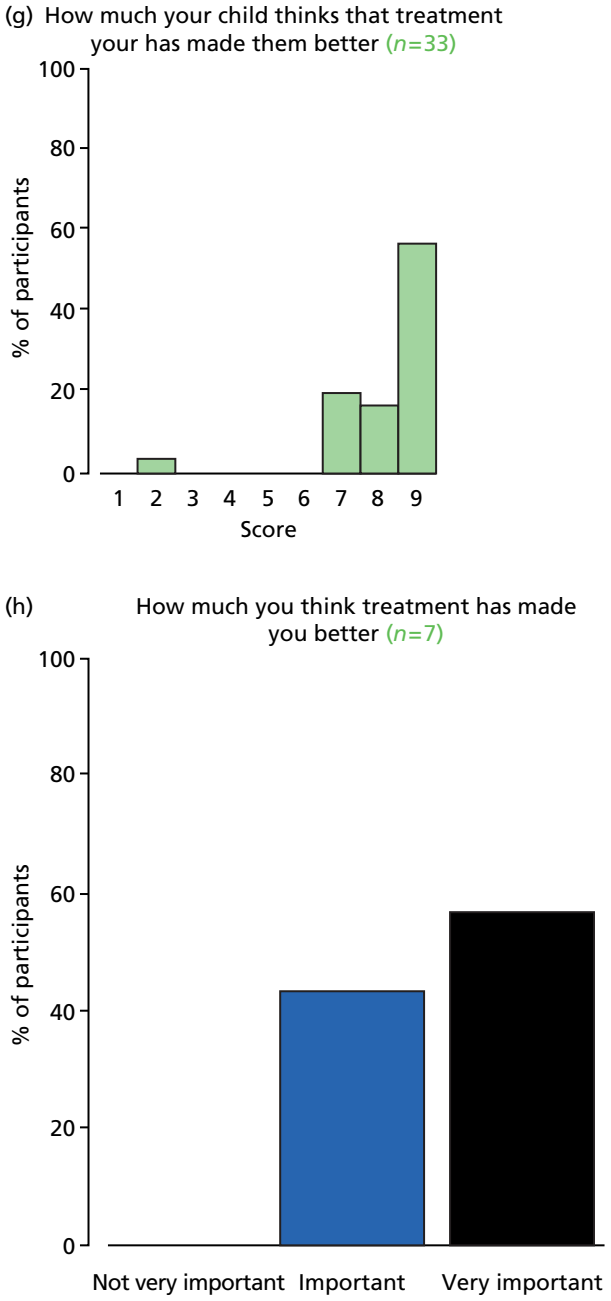


FIGURE 35 Outcome: child's satisfaction with treatment.

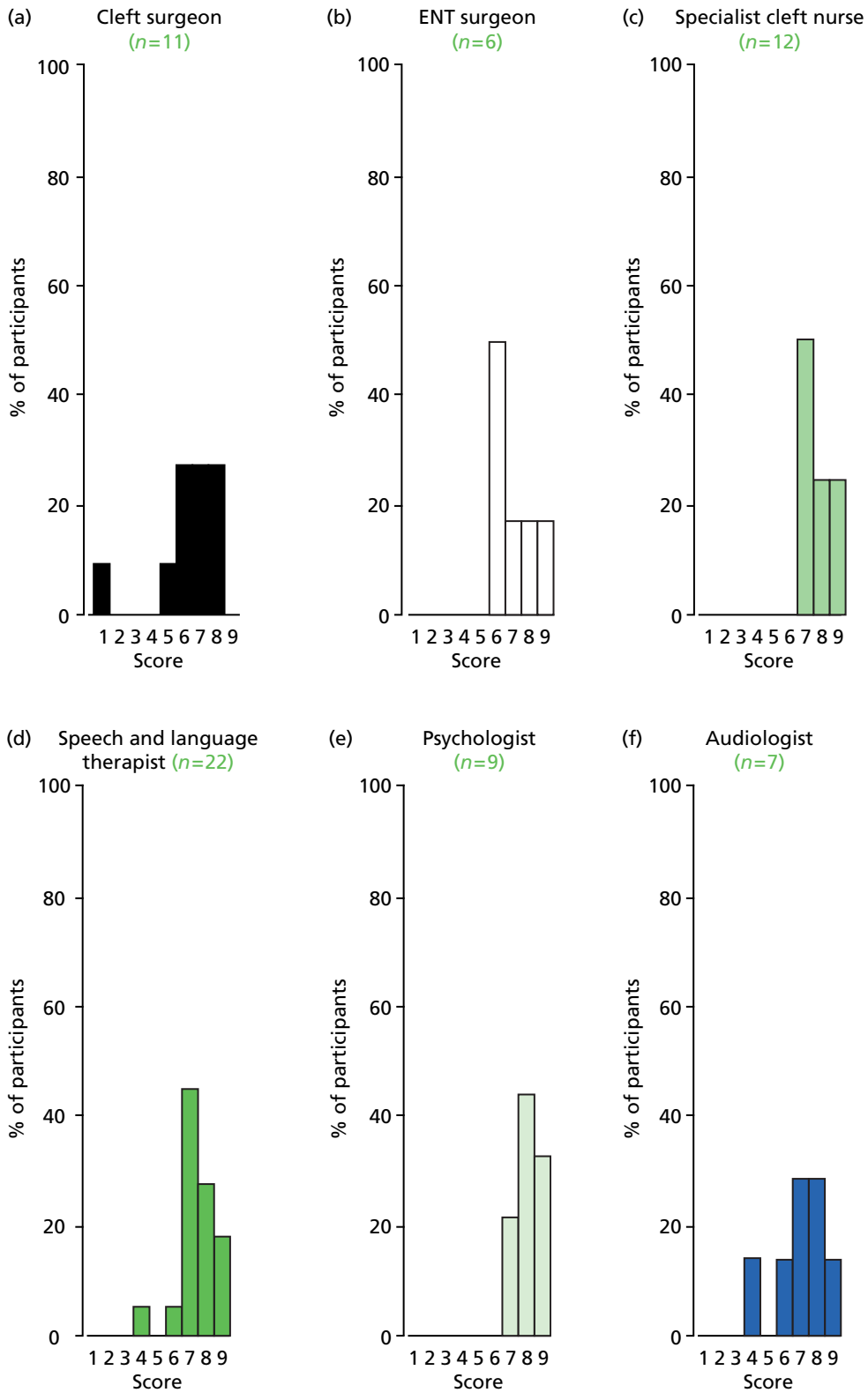


FIGURE 36 Outcome: child's perspective of speech. (*continued*)

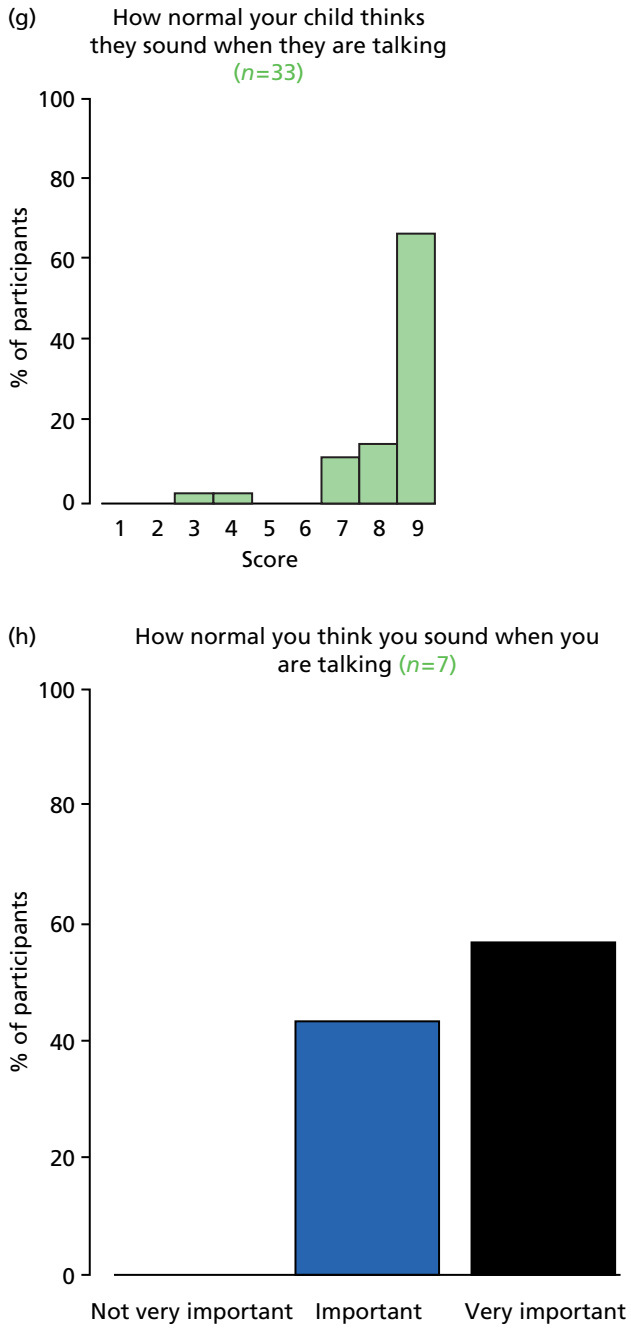


FIGURE 36 Outcome: child's perspective of speech.

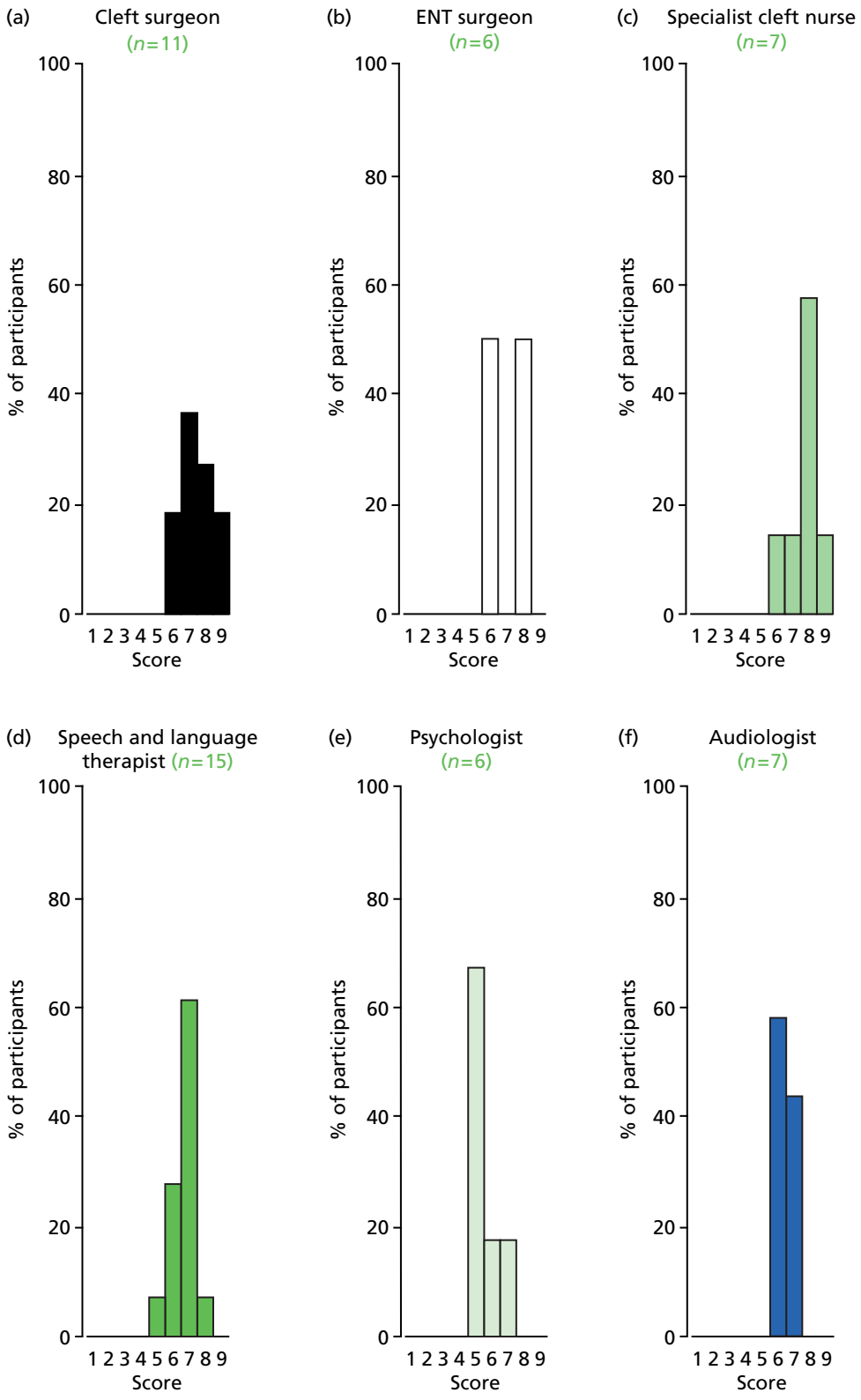


FIGURE 37 Outcome: persistent tympanic membrane retraction. (*continued*)

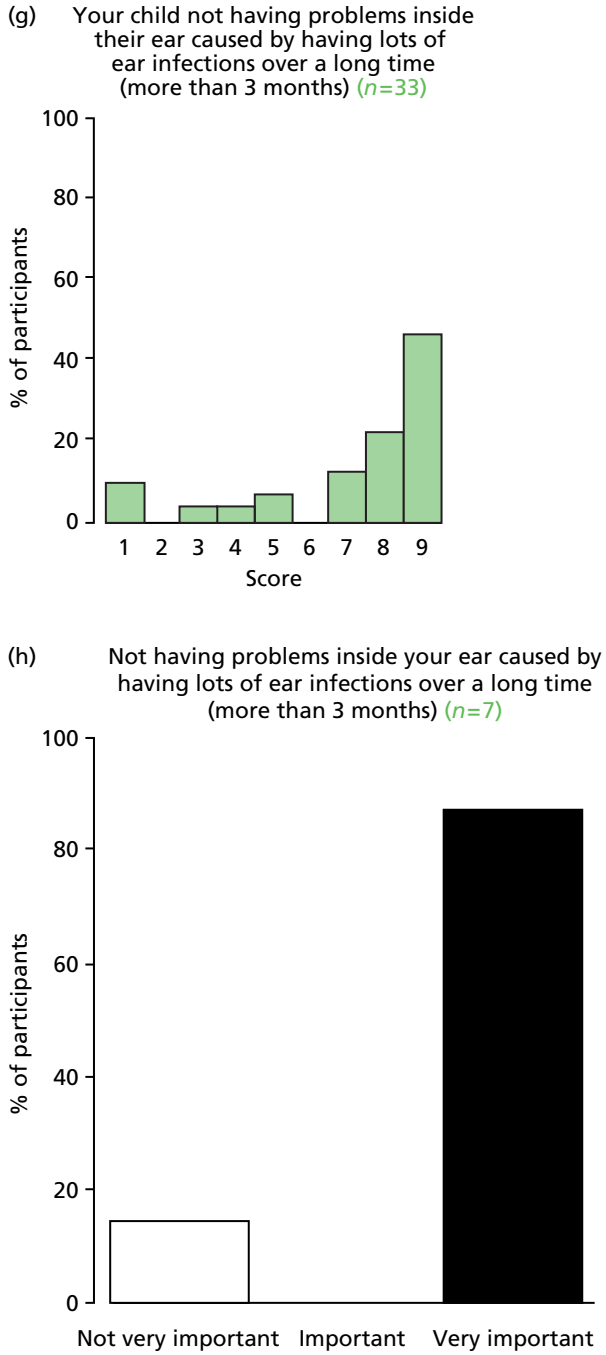


FIGURE 37 Outcome: persistent tympanic membrane retraction.

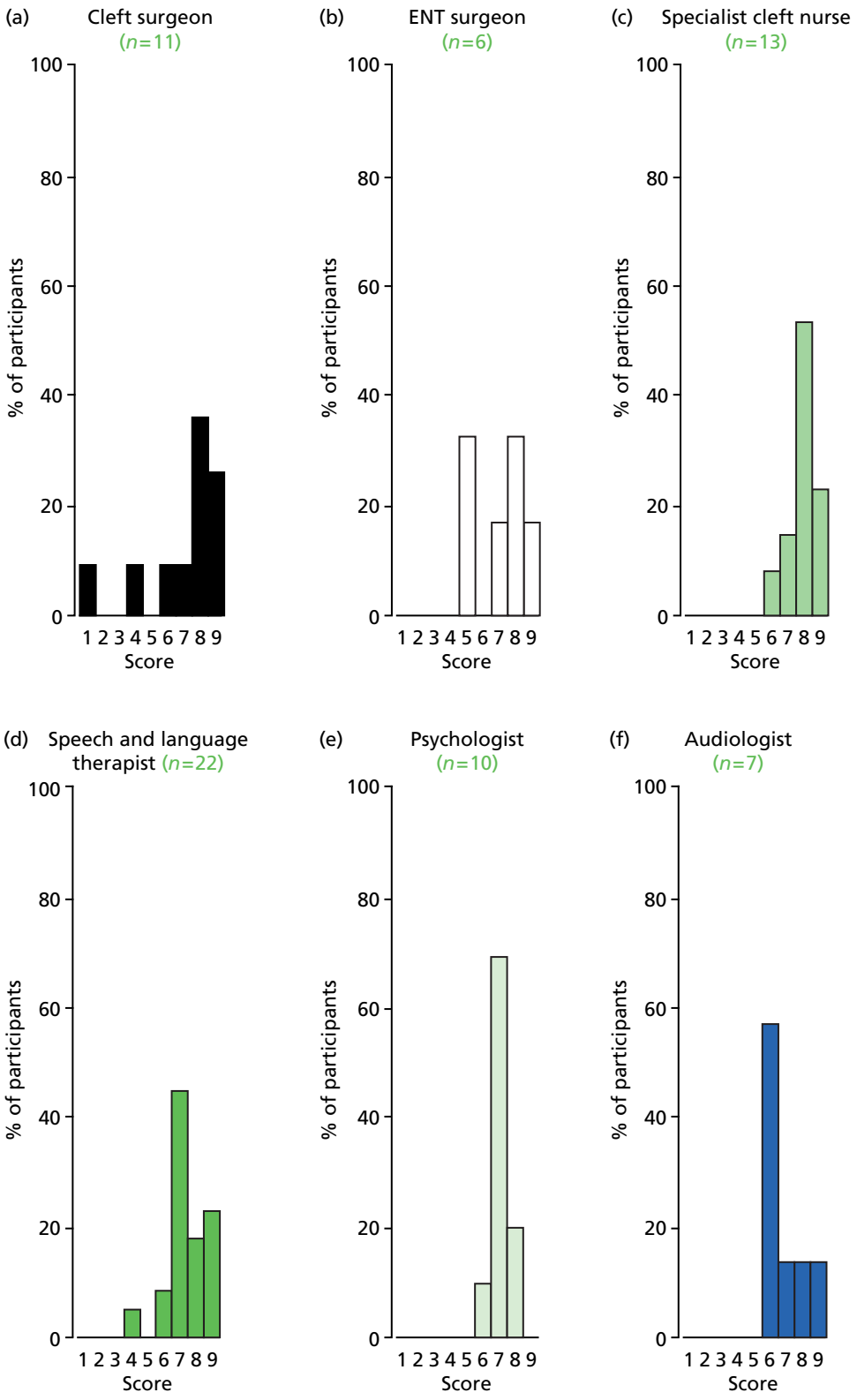


FIGURE 38 Outcome: cognitive development. (*continued*)

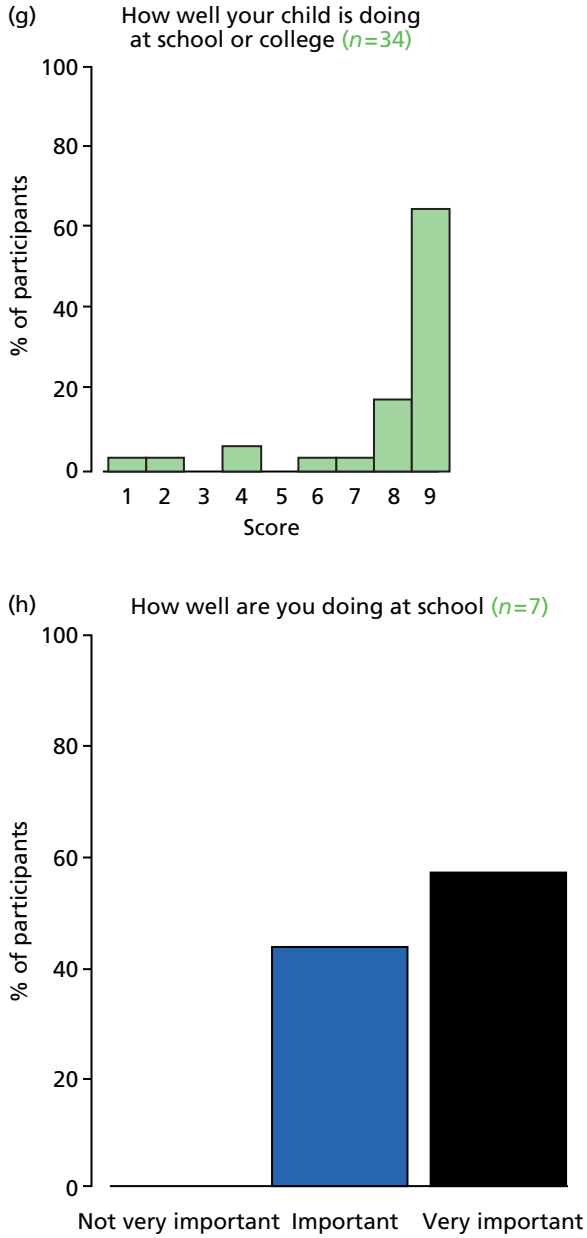


FIGURE 38 Outcome: cognitive development.

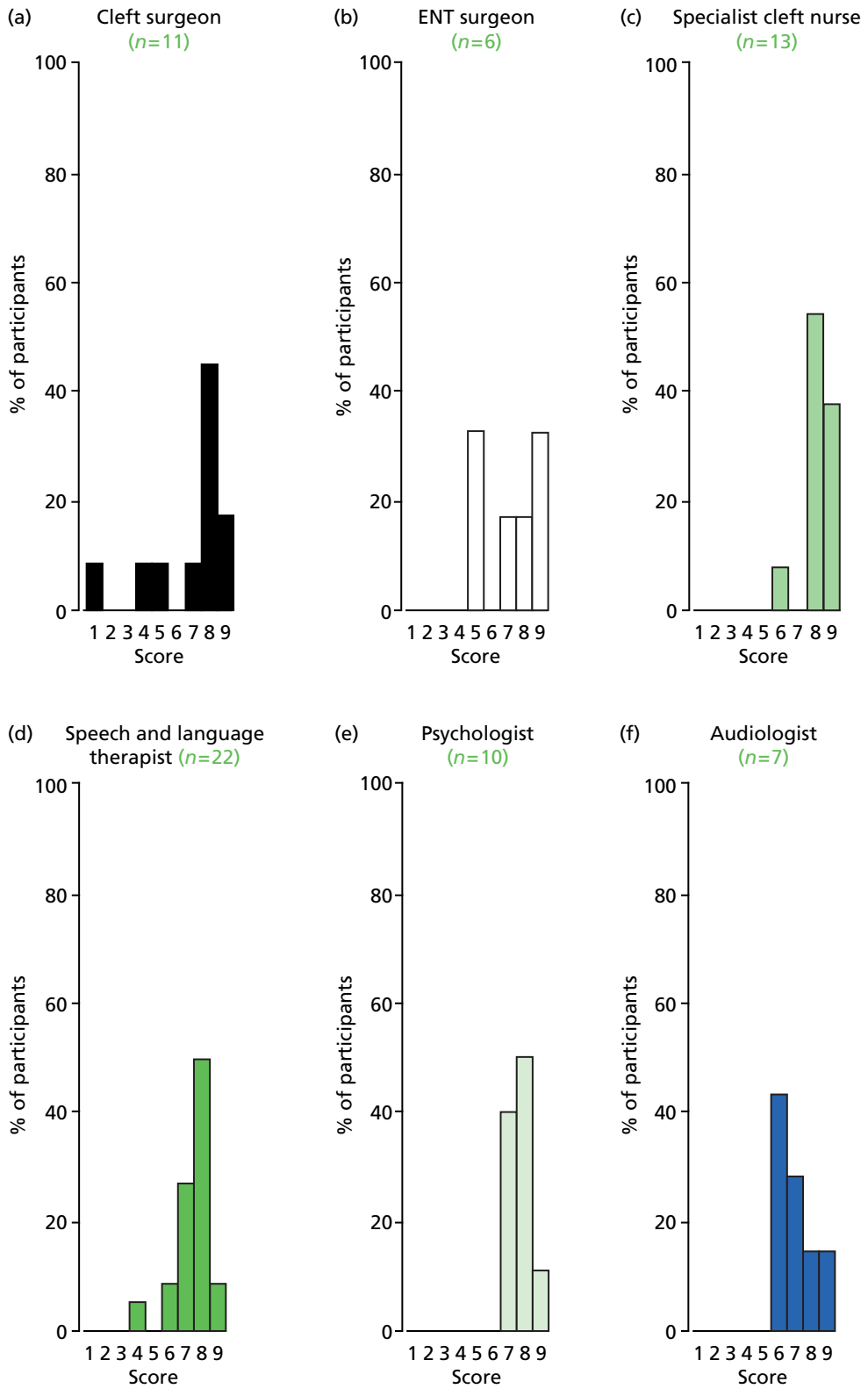


FIGURE 39 Outcome: developmental progress. (*continued*)

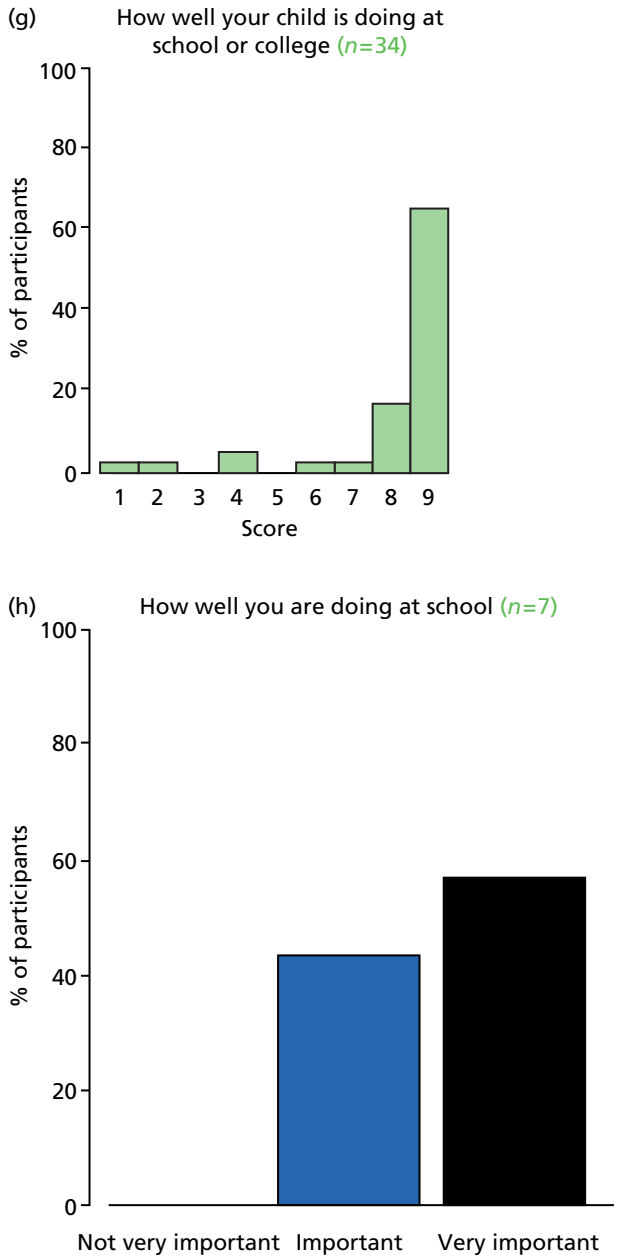


FIGURE 39 Outcome: developmental progress.

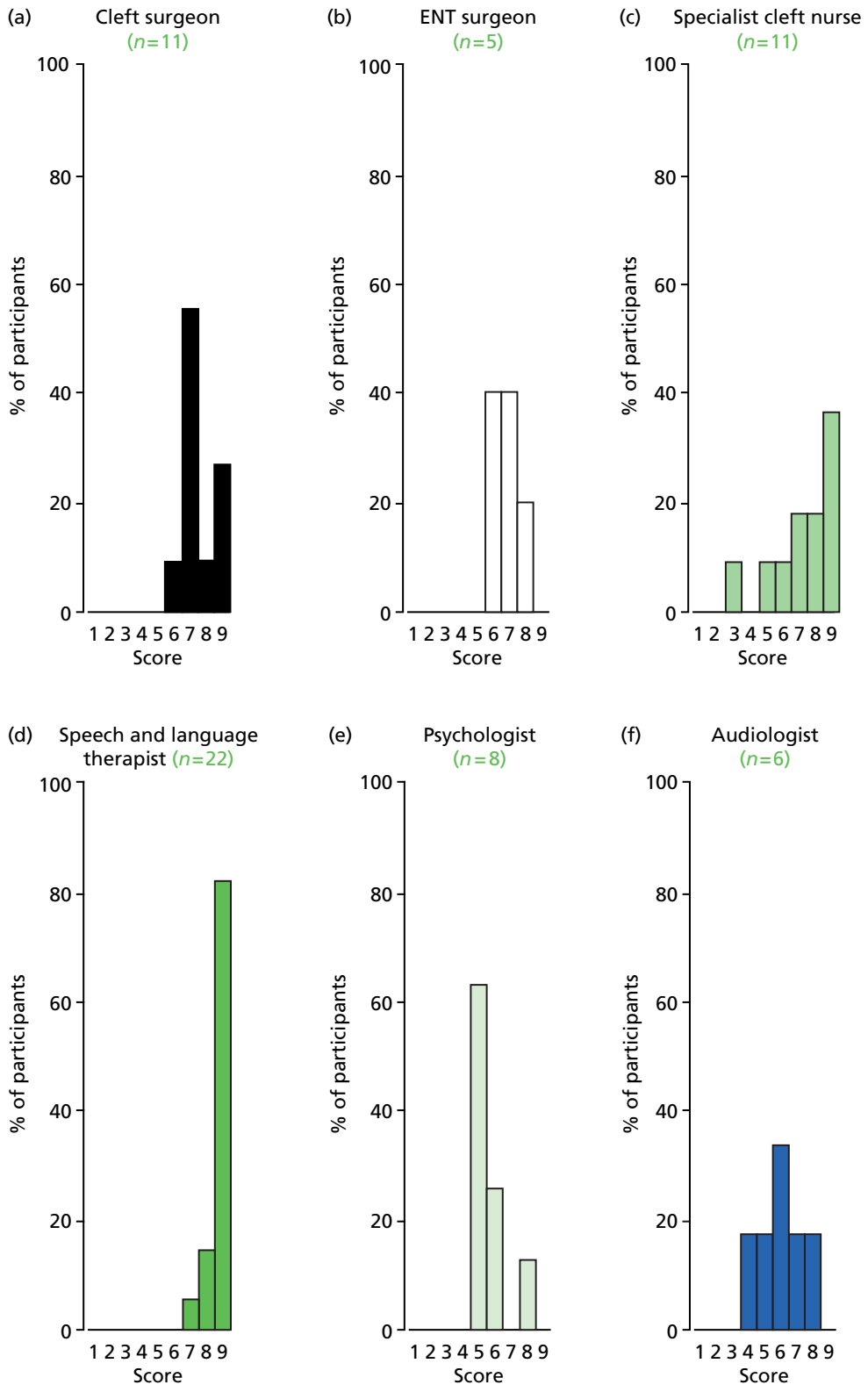


FIGURE 40 Outcome: consonant production – cleft-related speech patterns. (*continued*)

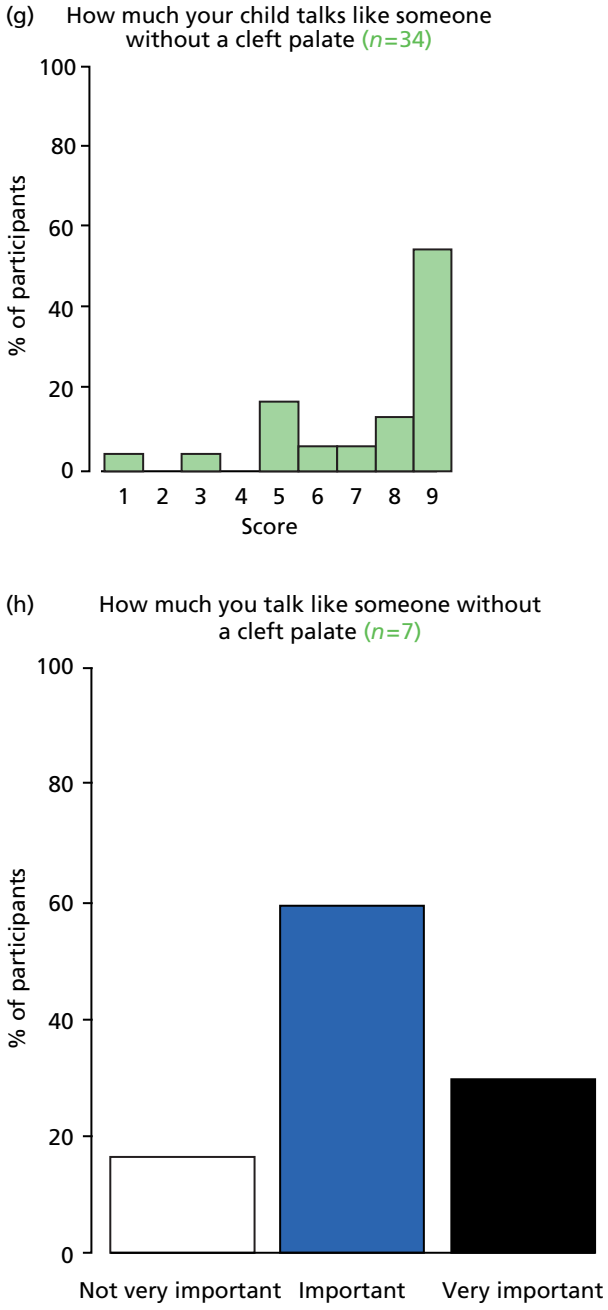


FIGURE 40 Outcome: consonant production – cleft-related speech patterns.

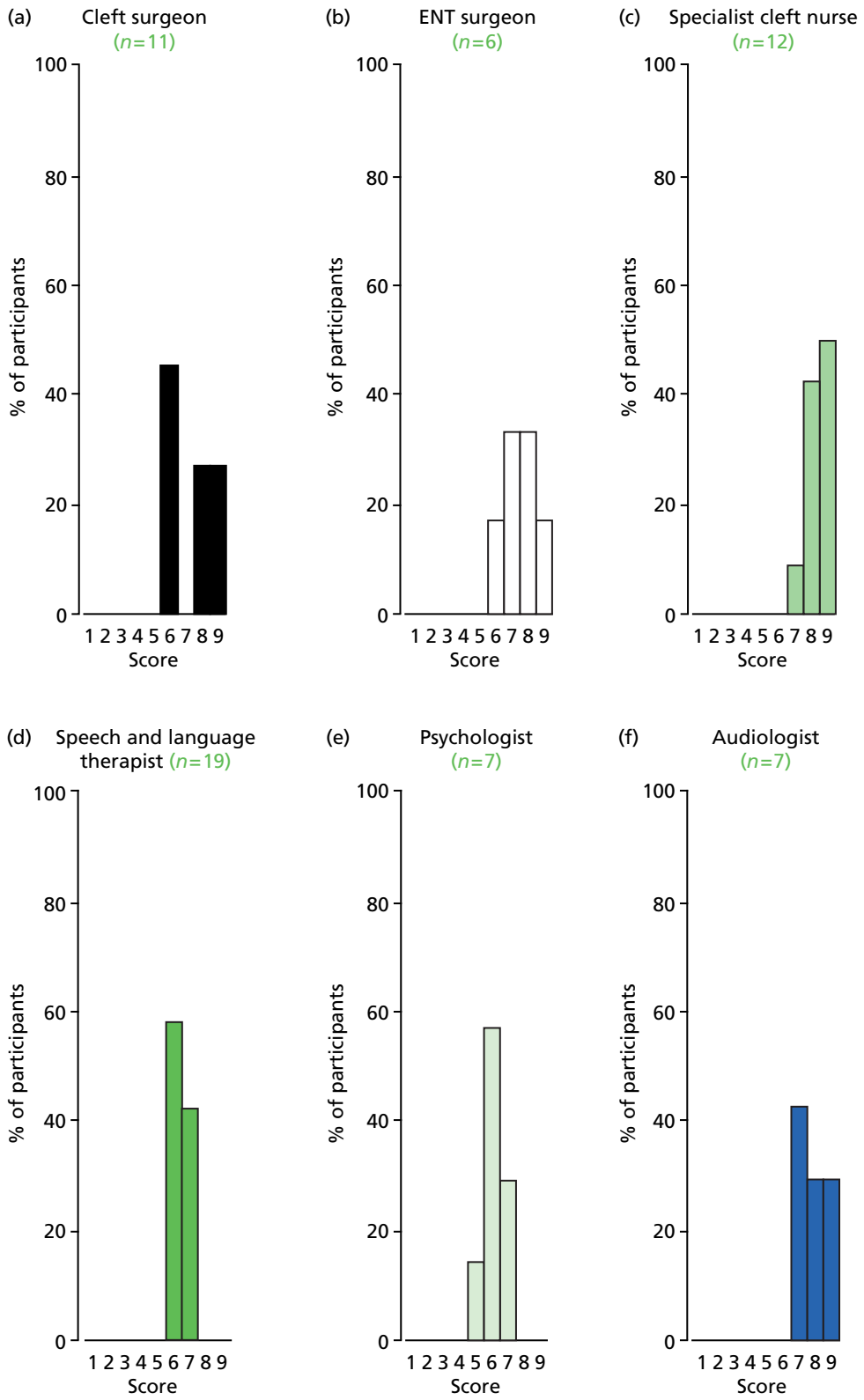


FIGURE 41 Outcome: side effects of treatment. (*continued*)

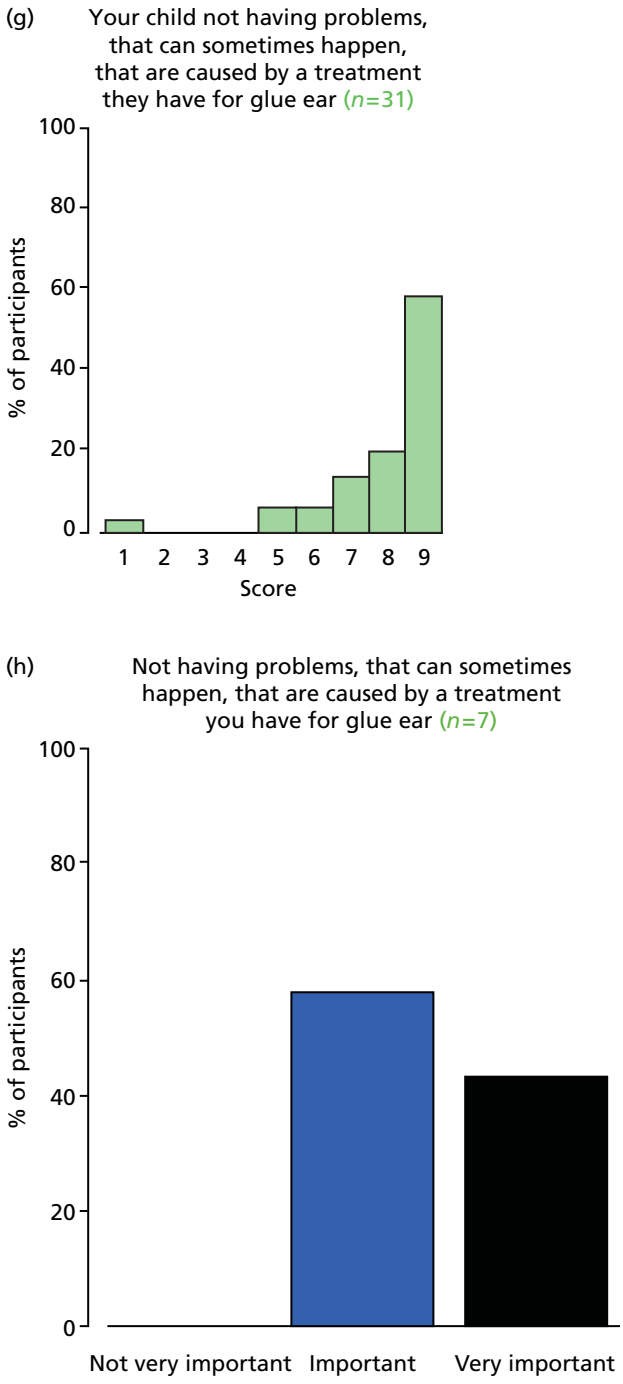


FIGURE 41 Outcome: side effects of treatment.

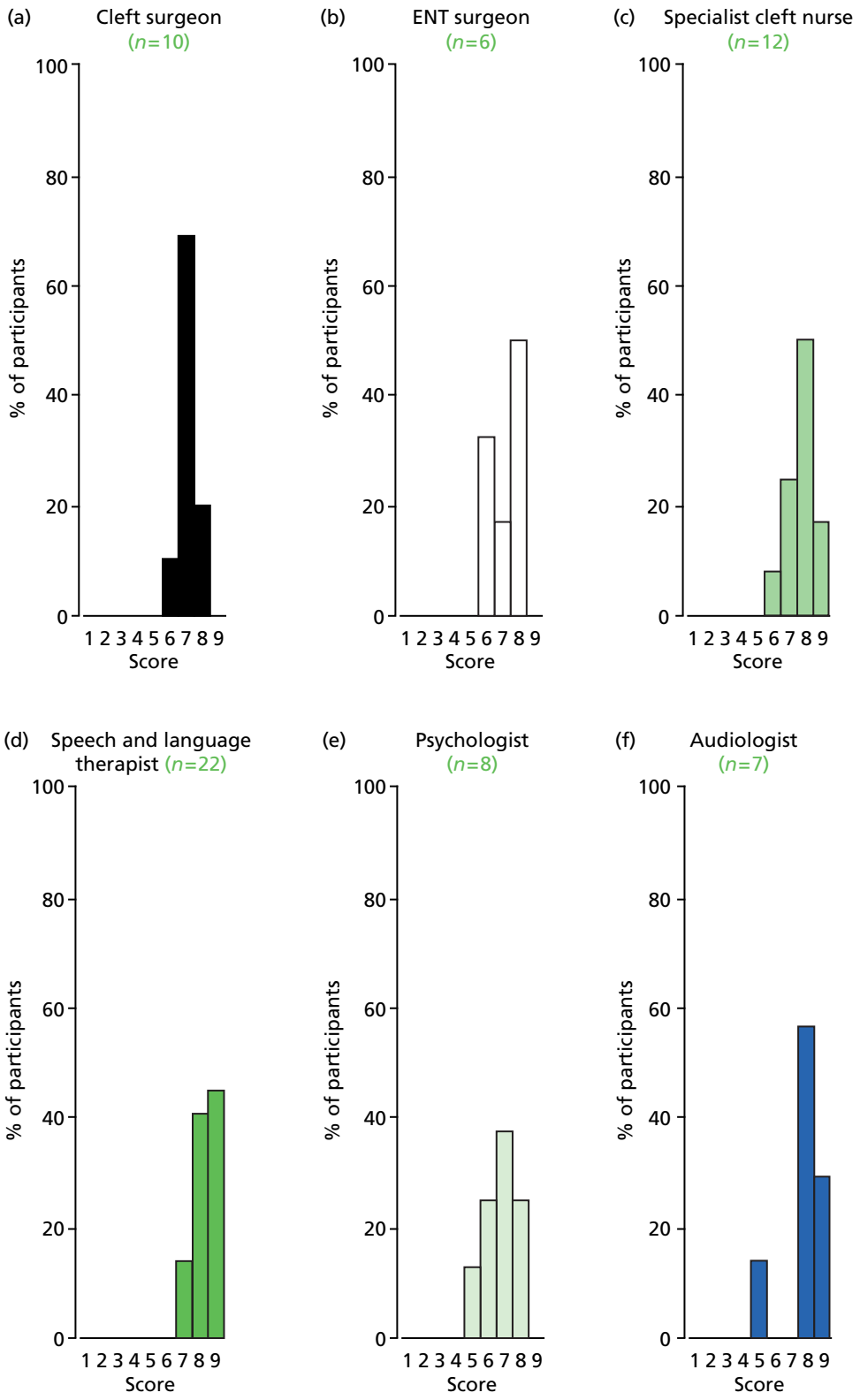


FIGURE 42 Outcome: listening skills.

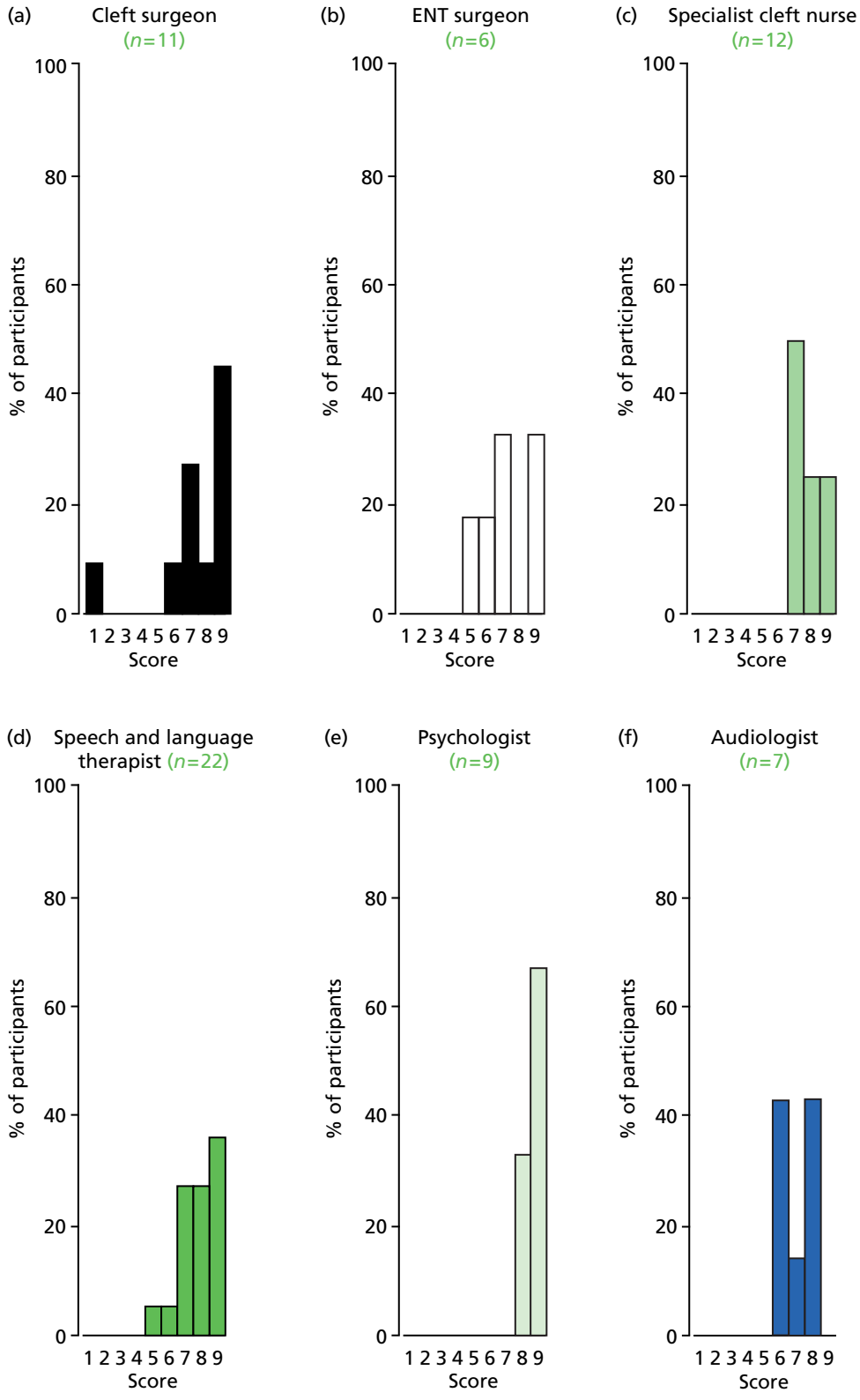


FIGURE 43 Outcome: psychosocial well-being.

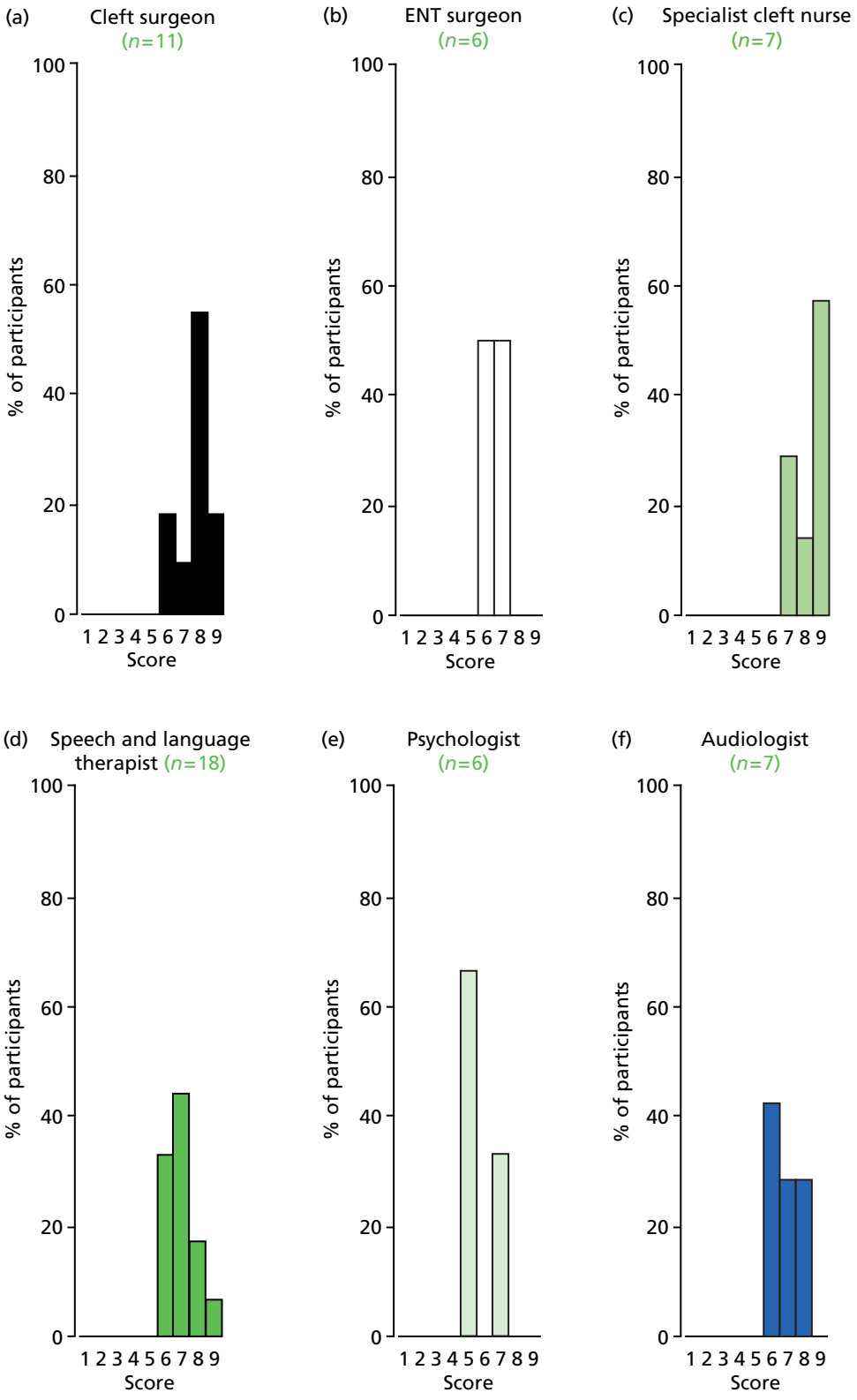


FIGURE 44 Outcome: persistent tympanic membrane perforation. (*continued*)

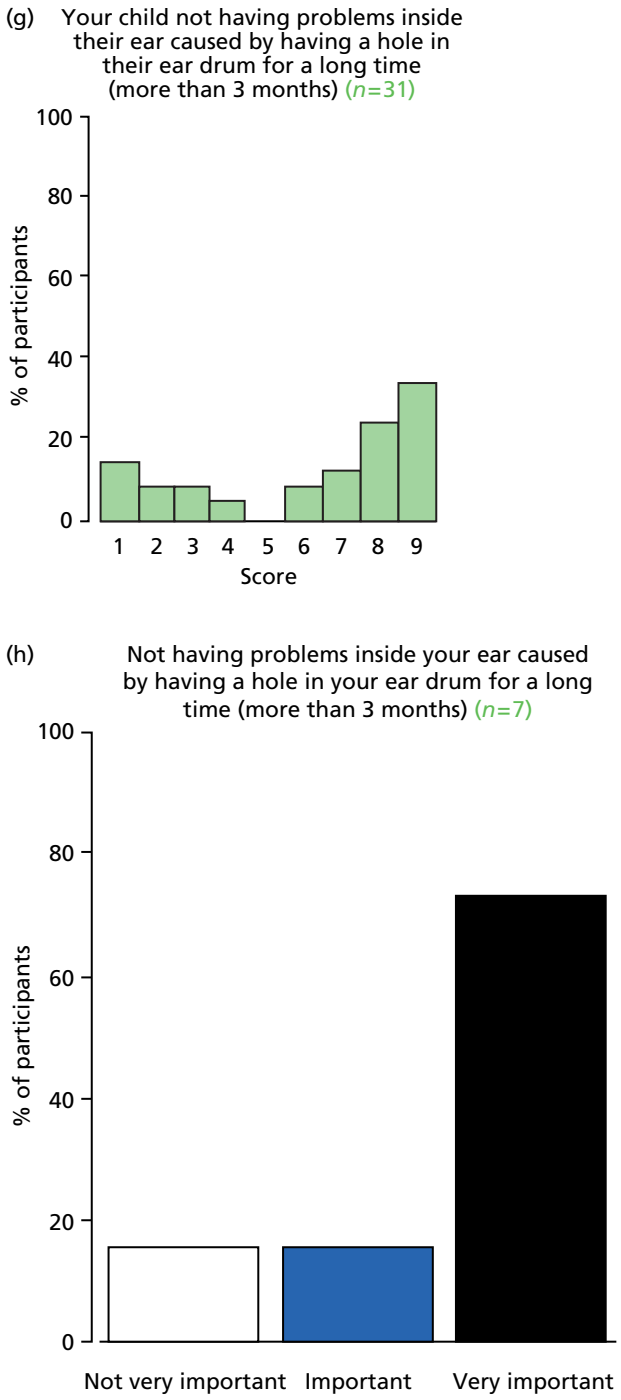


FIGURE 44 Outcome: persistent tympanic membrane perforation.

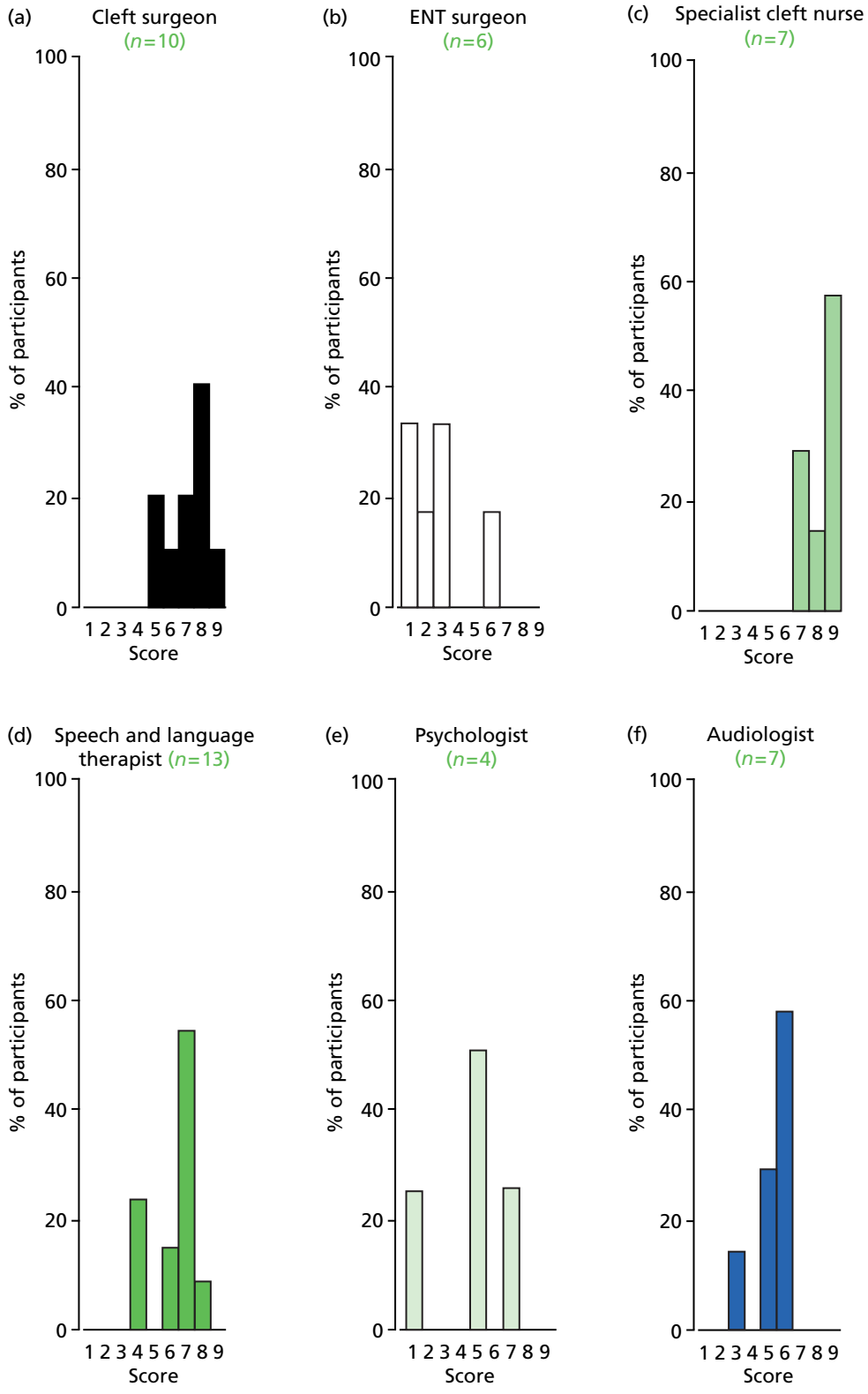


FIGURE 45 Outcome: tympanosclerosis. (*continued*)

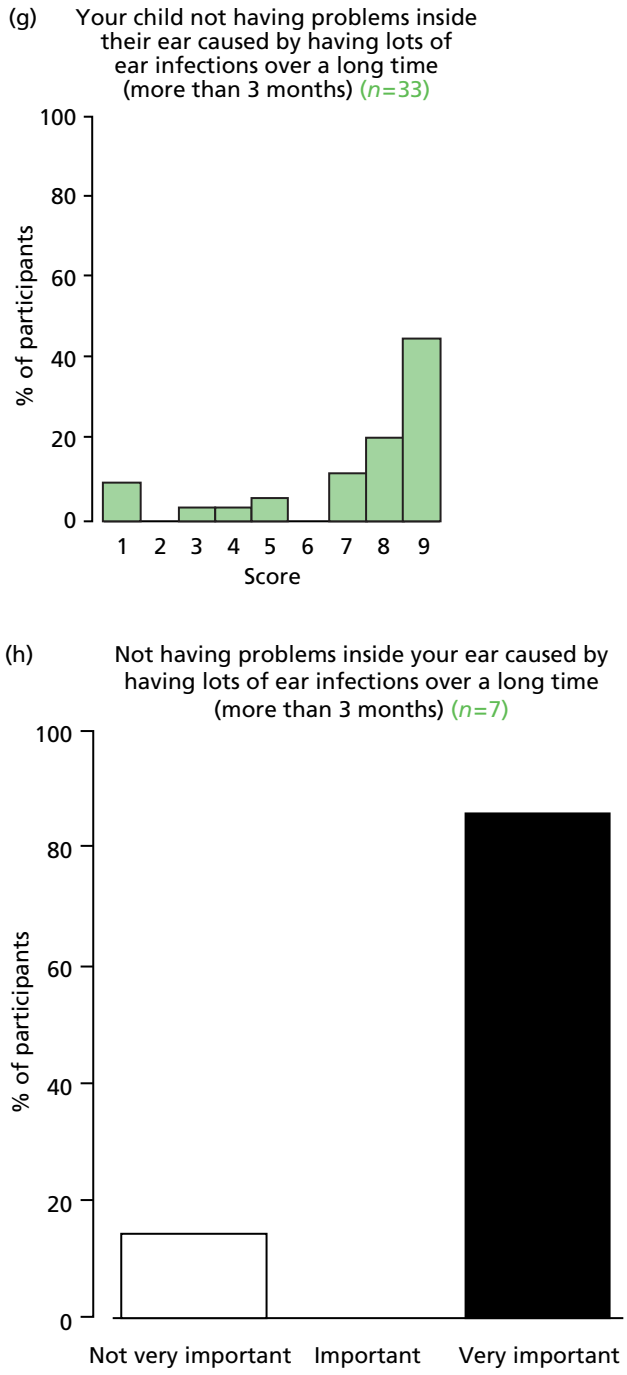


FIGURE 45 Outcome: tympanosclerosis.

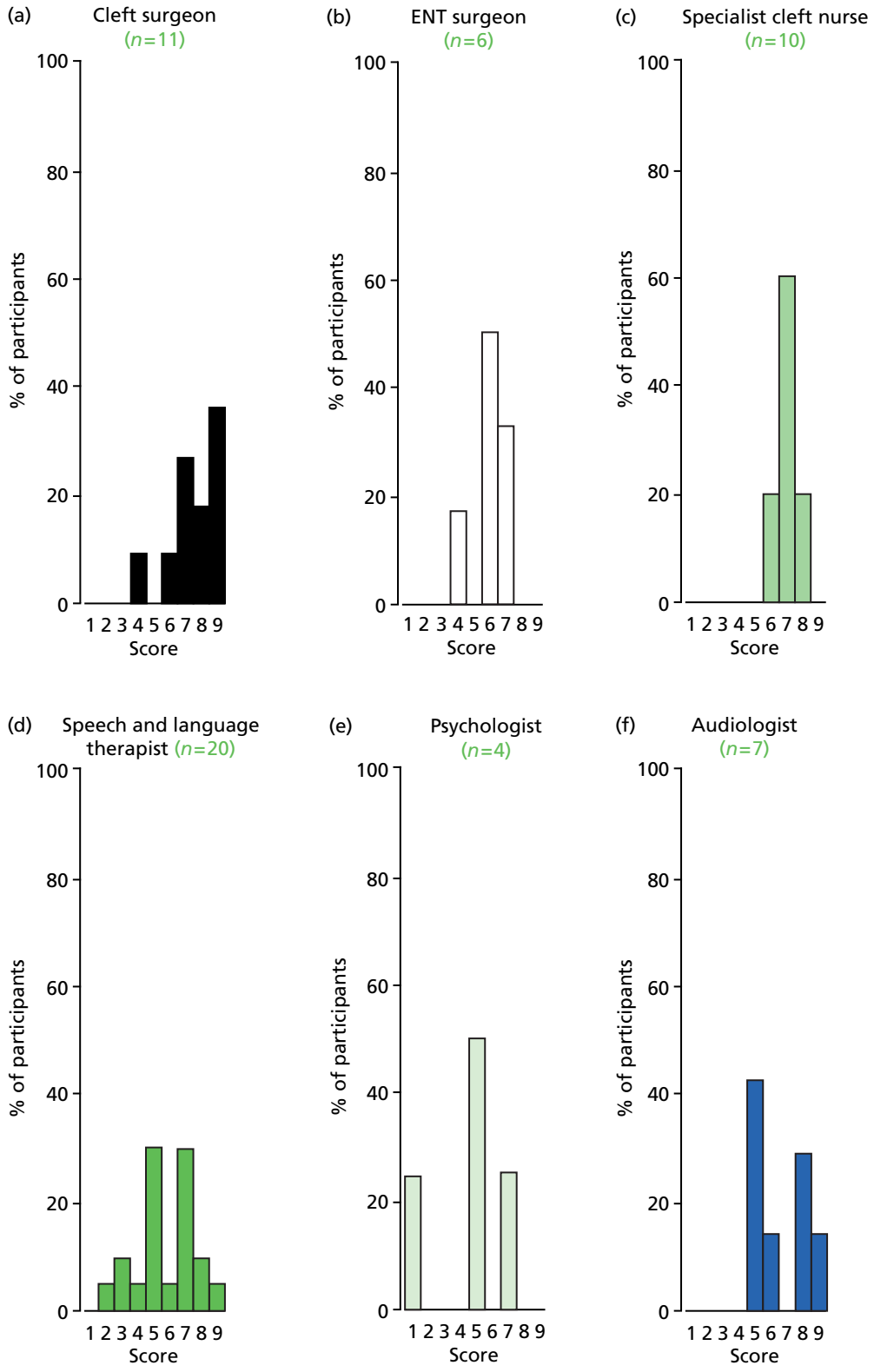


FIGURE 46 Outcome: otalgia. (*continued*)

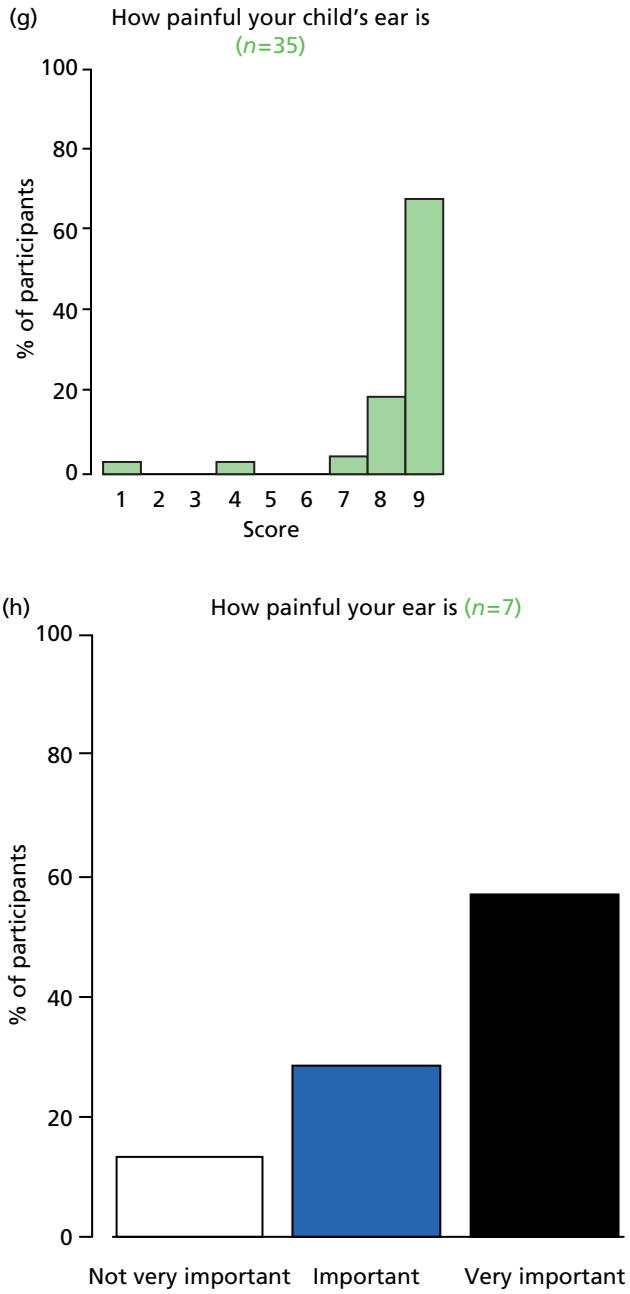


FIGURE 46 Outcome: otalgia.

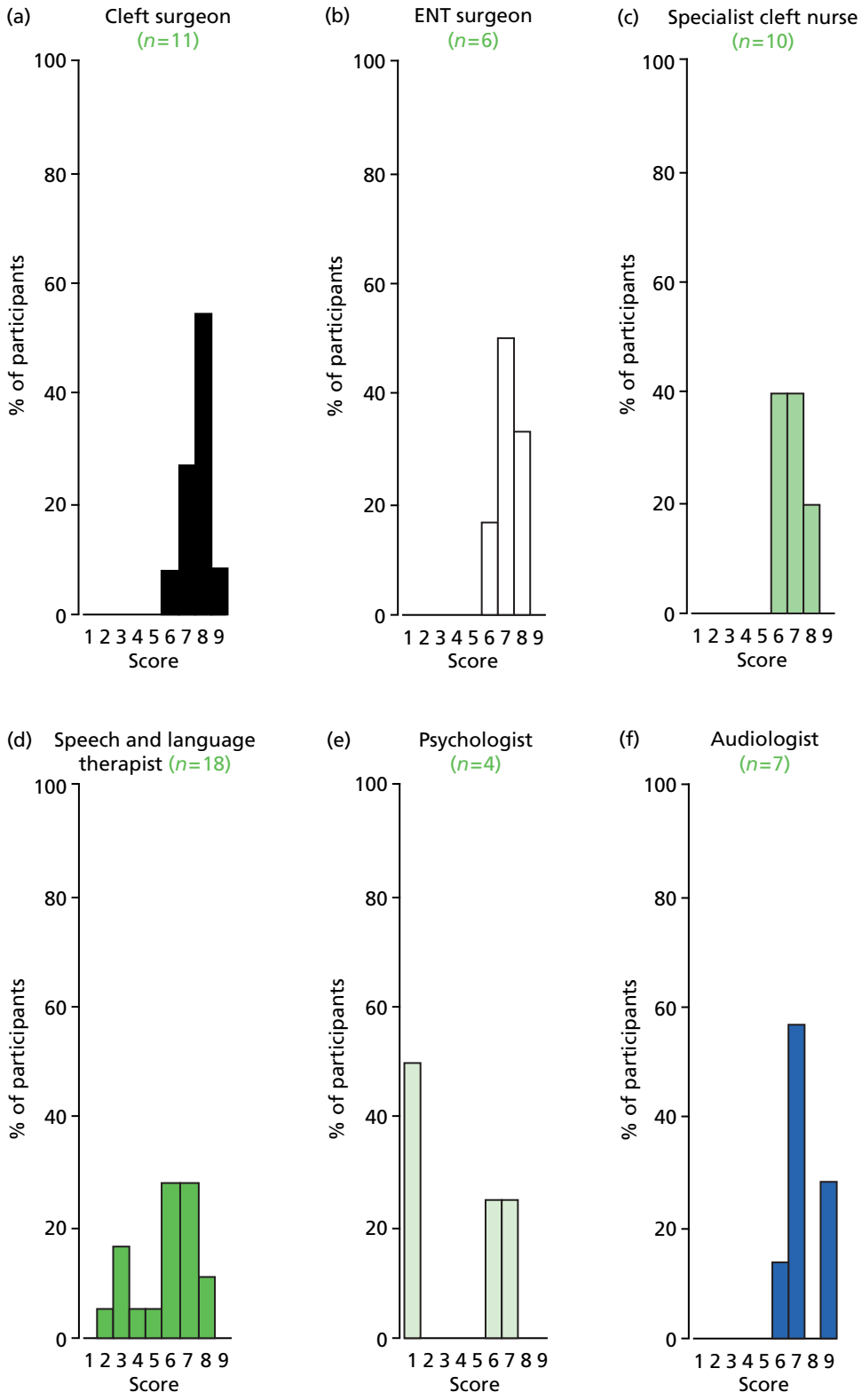


FIGURE 47 Outcome: otorrhoea. (*continued*)

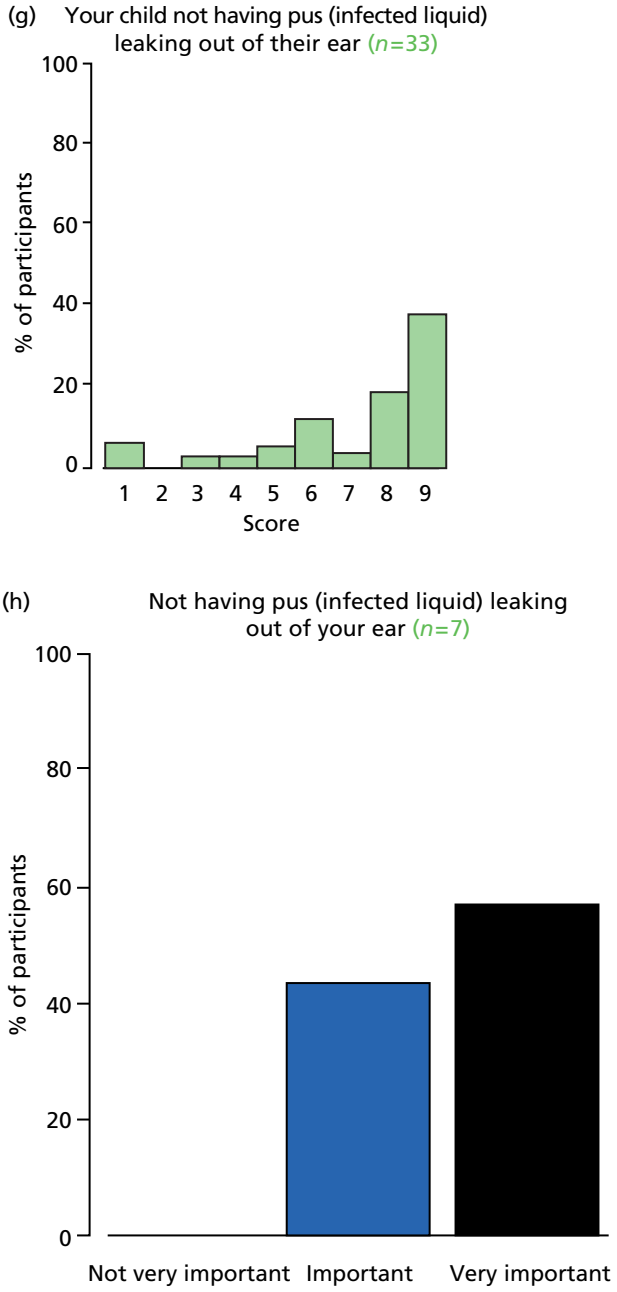


FIGURE 47 Outcome: otorrhoea.

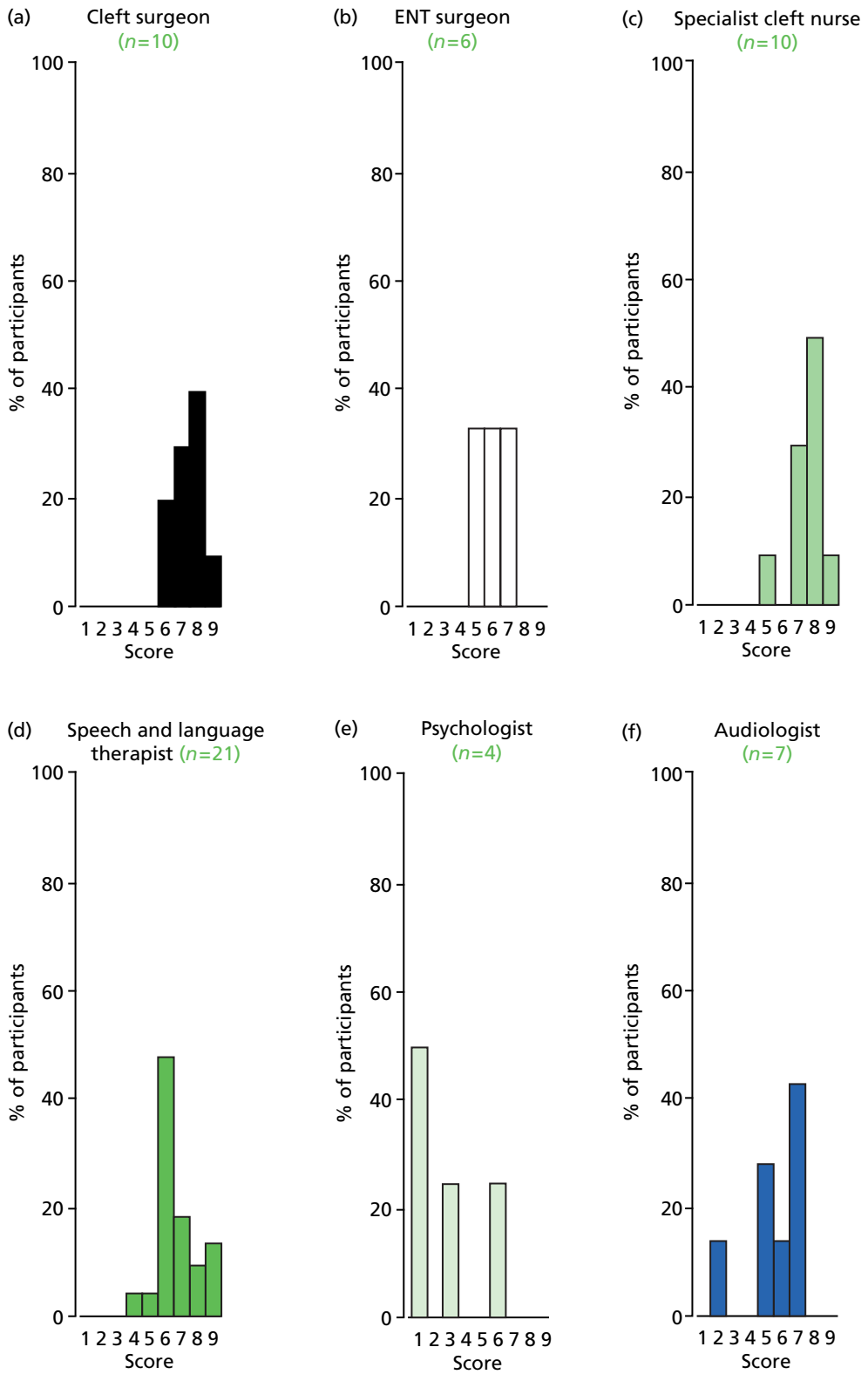


FIGURE 48 Outcome: eustachian tube function. (*continued*)

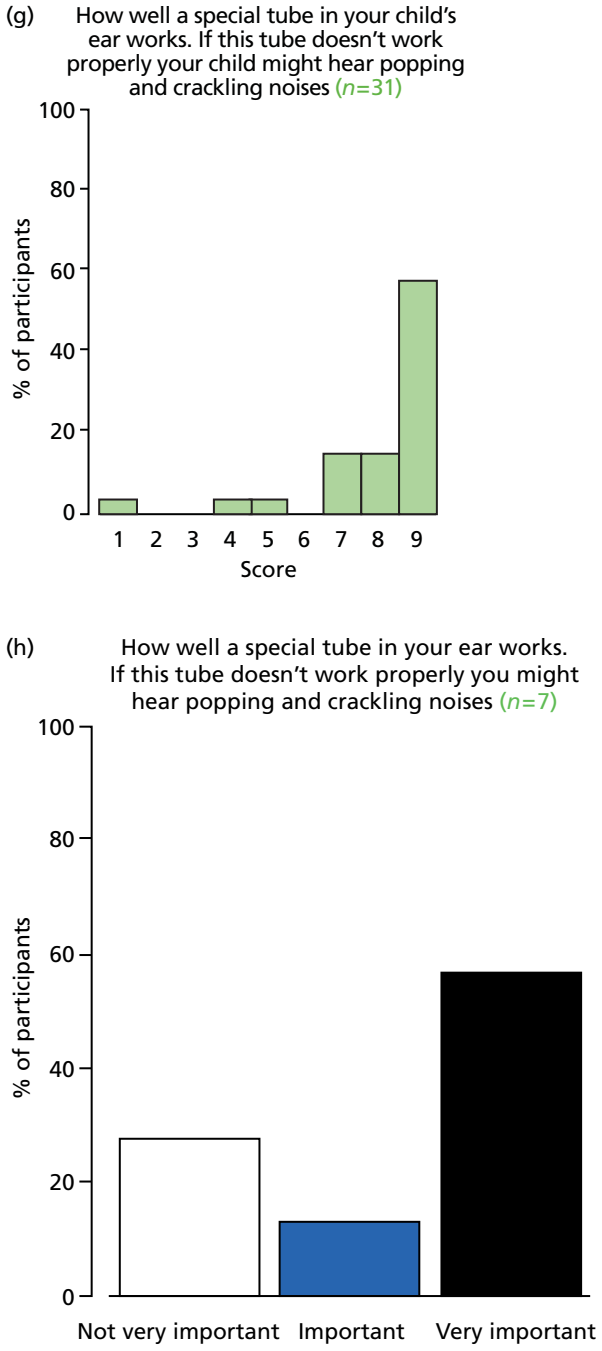


FIGURE 48 Outcome: eustachian tube function.

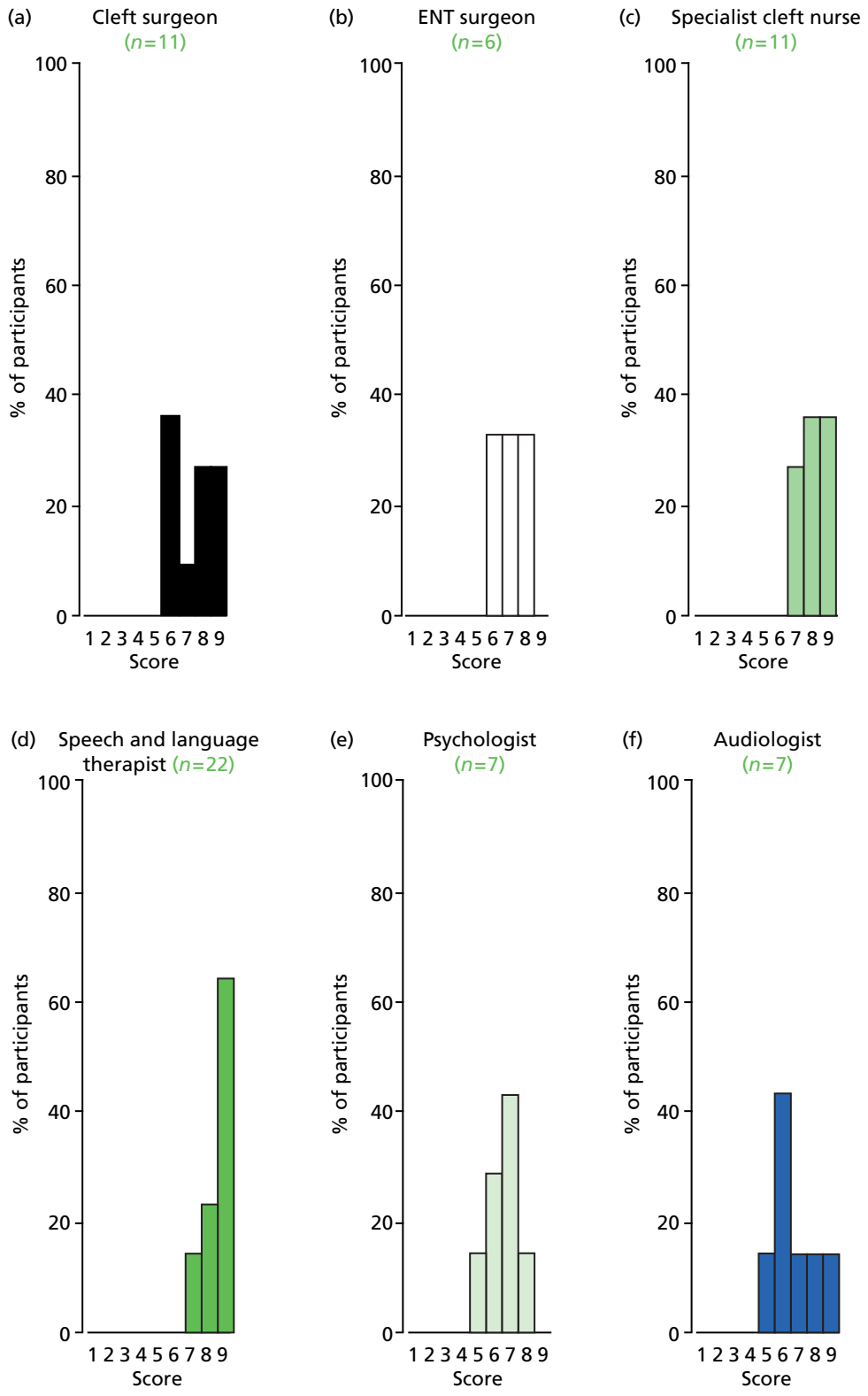


FIGURE 49 Outcome: expressive language skills. (*continued*)

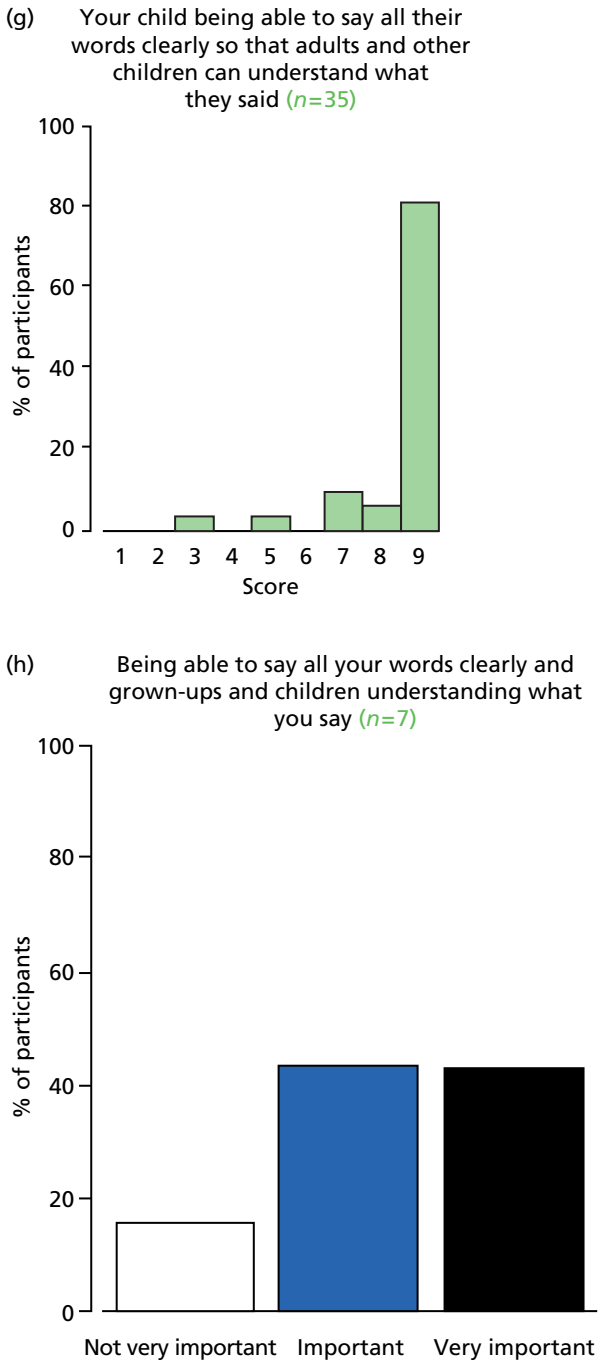


FIGURE 49 Outcome: expressive language skills.

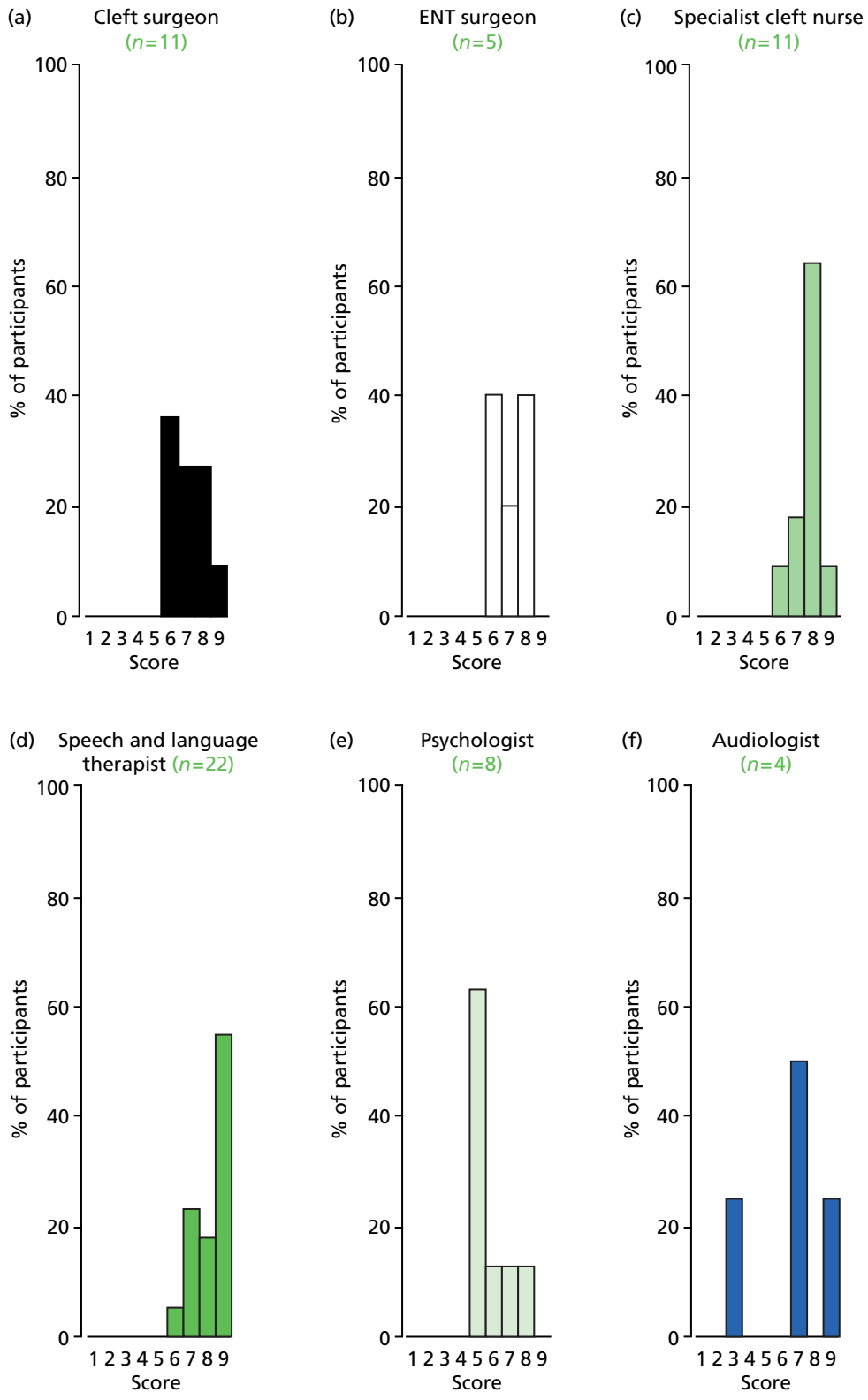


FIGURE 50 Outcome: speech signs of velopharyngeal insufficiency. (*continued*)

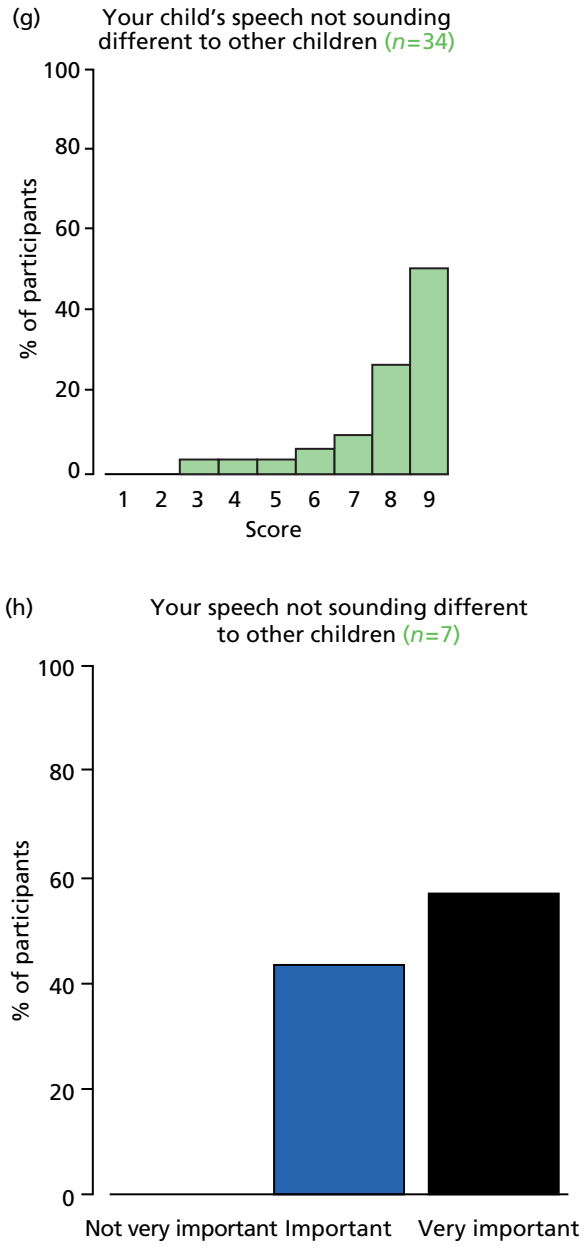


FIGURE 50 Outcome: speech signs of velopharyngeal insufficiency.

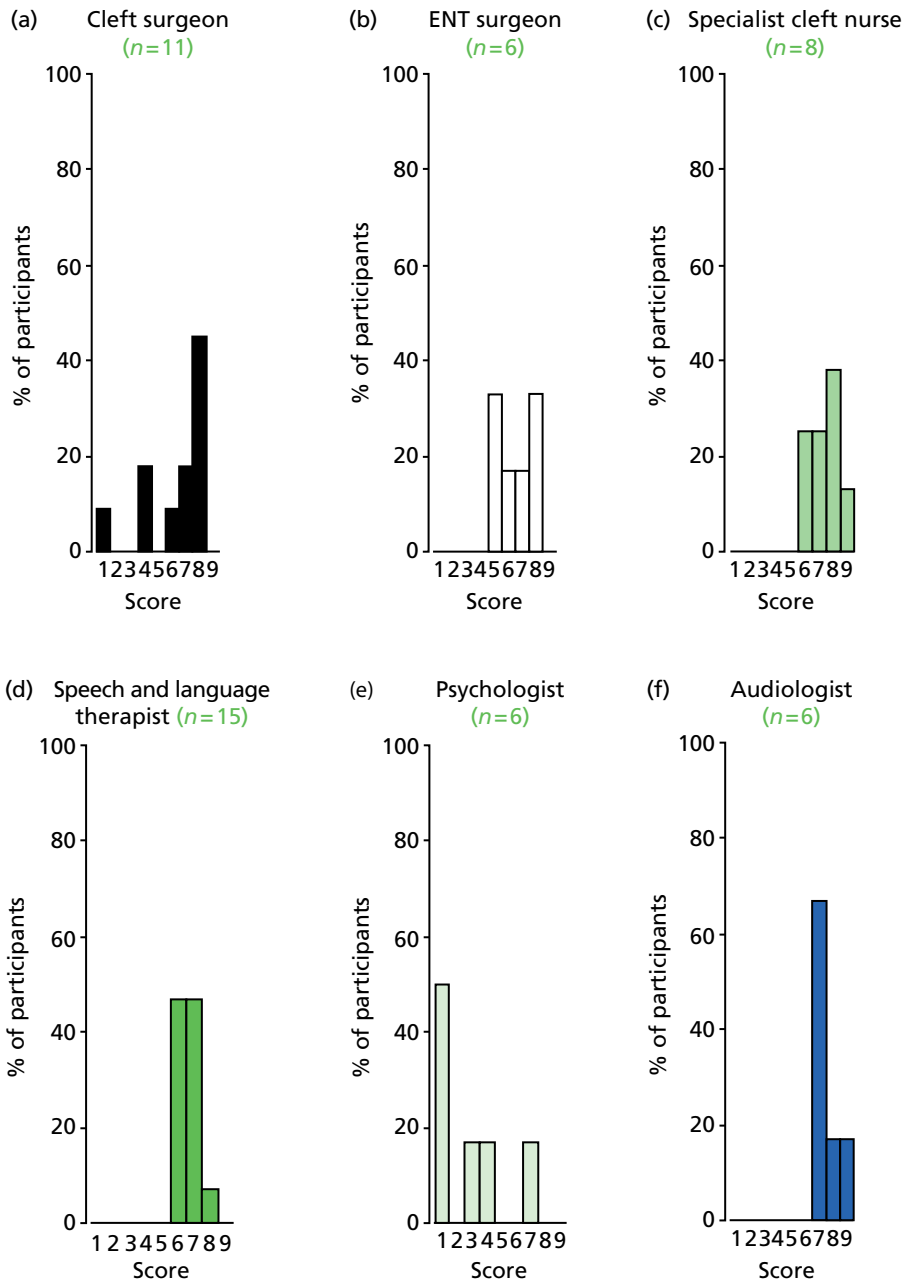


FIGURE 51 Outcome: early extrusion or blockage of VTs. (*continued*)

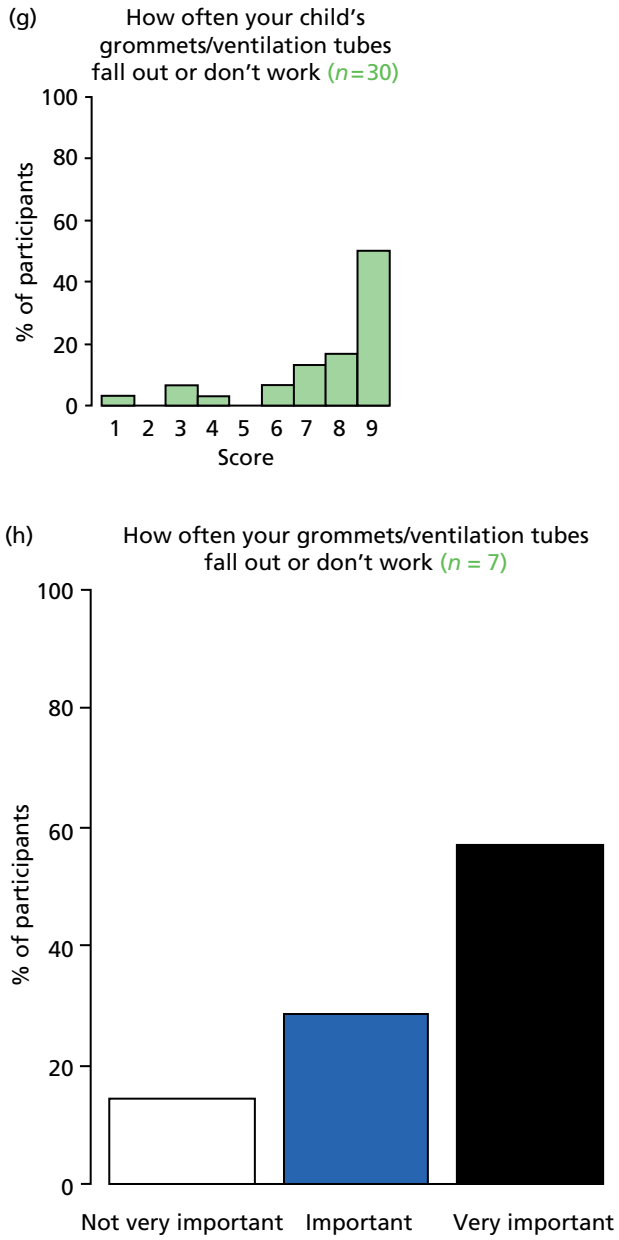


FIGURE 51 Outcome: early extrusion or blockage of VTs.

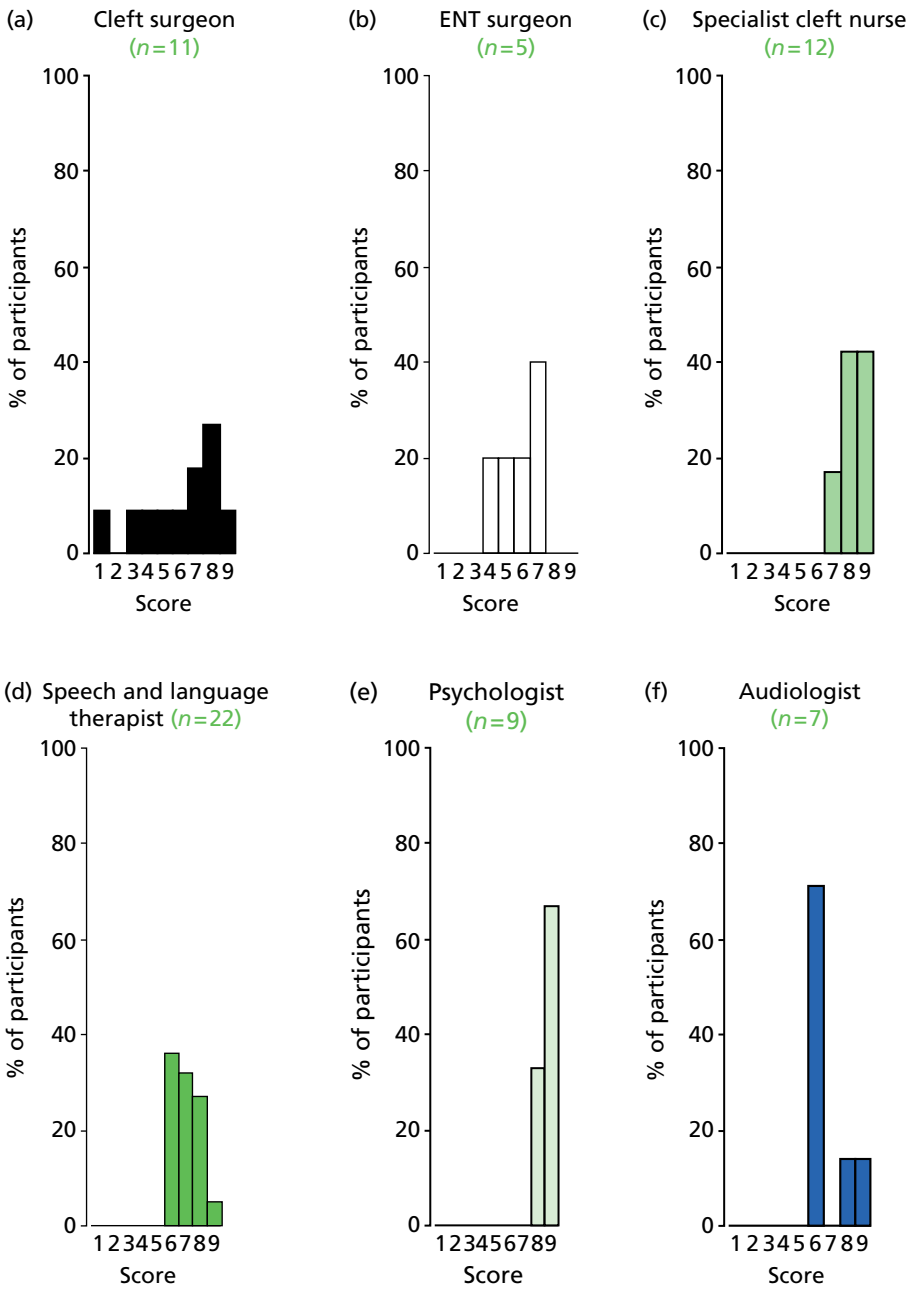


FIGURE 52 Outcome: child stress. (*continued*)

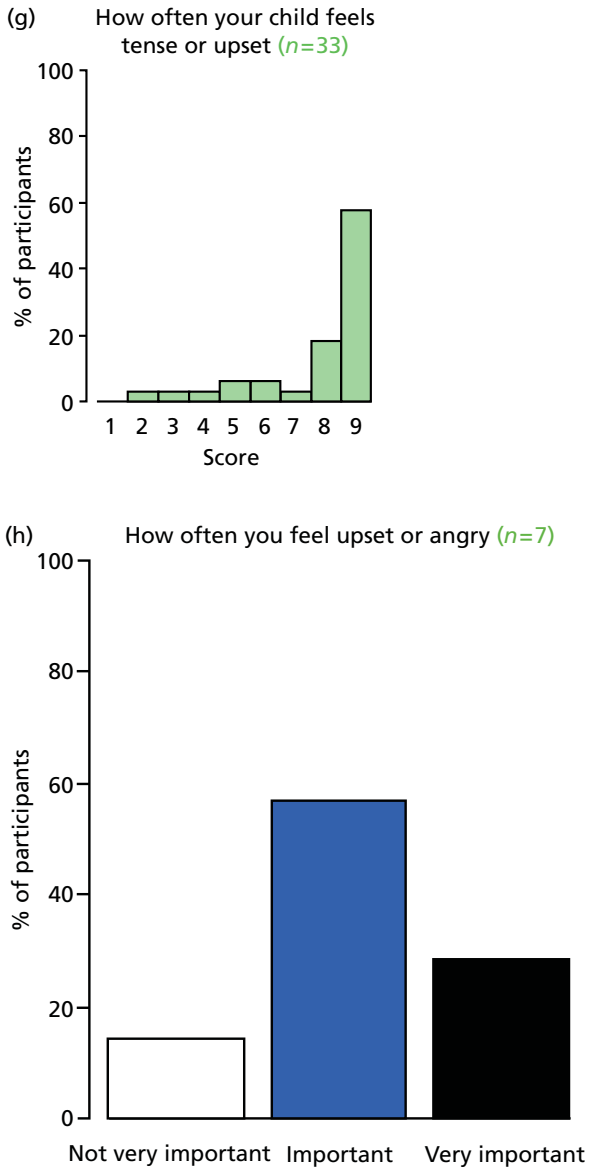


FIGURE 52 Outcome: child stress.

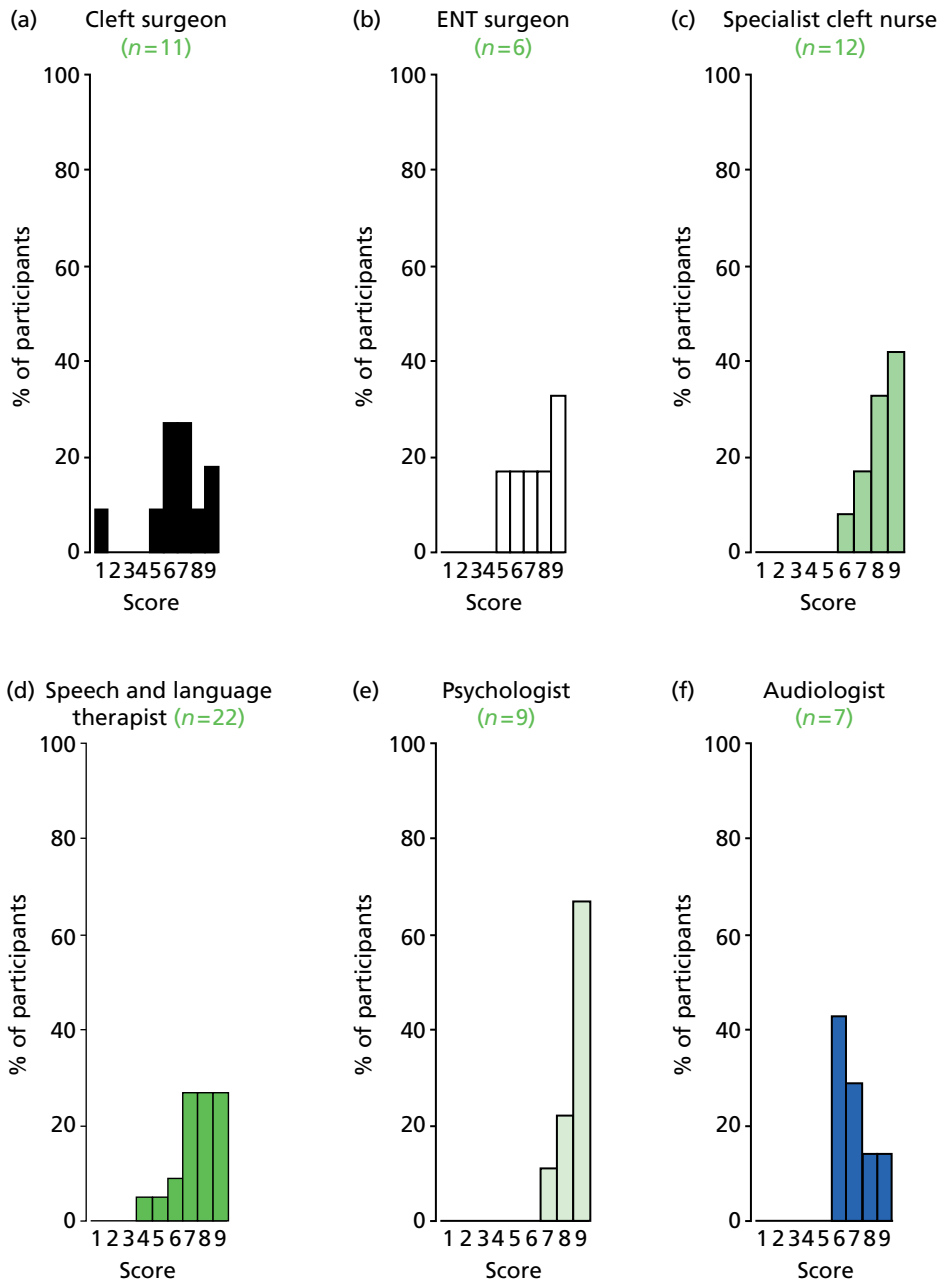


FIGURE 53 Outcome: psychological well-being.

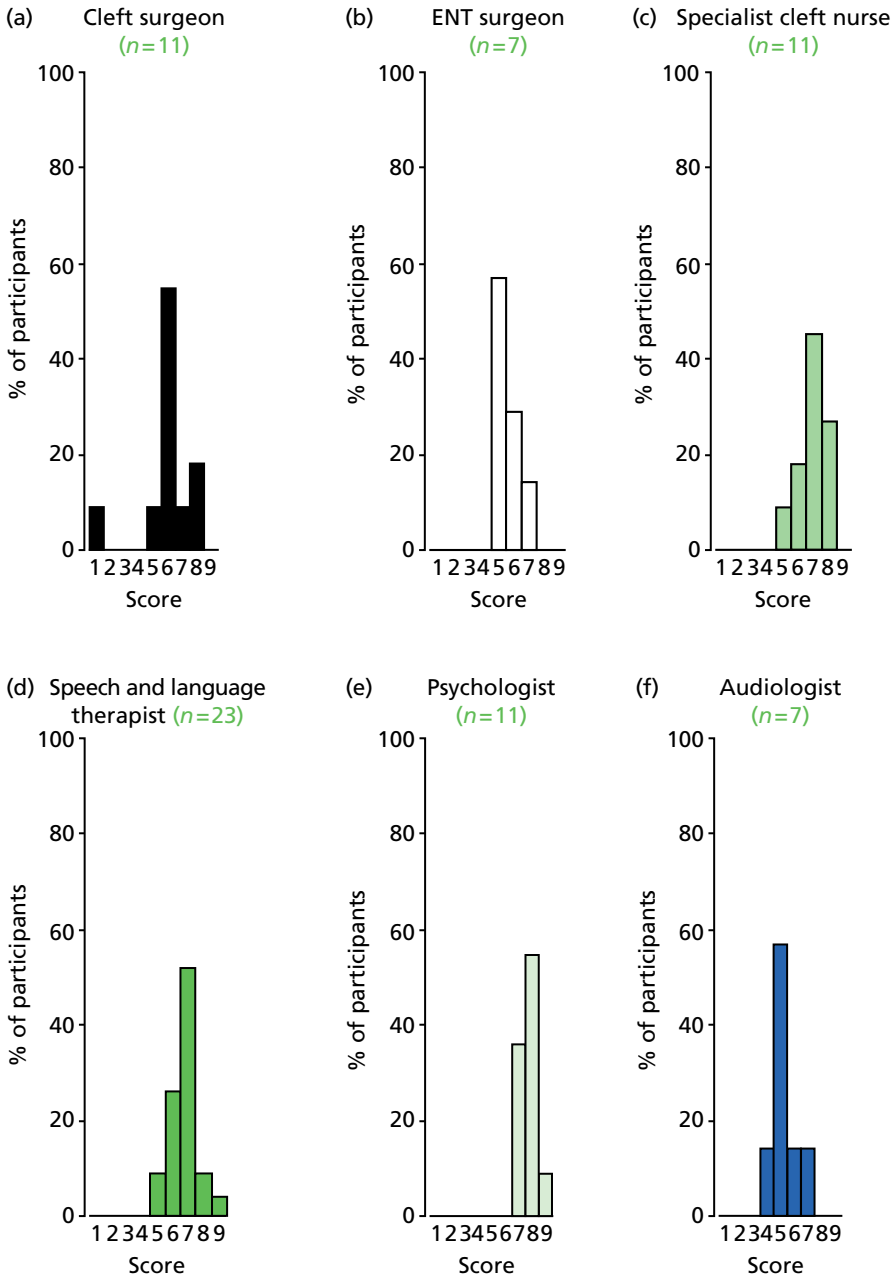


FIGURE 54 Outcome: internalising behaviour. (*continued*)

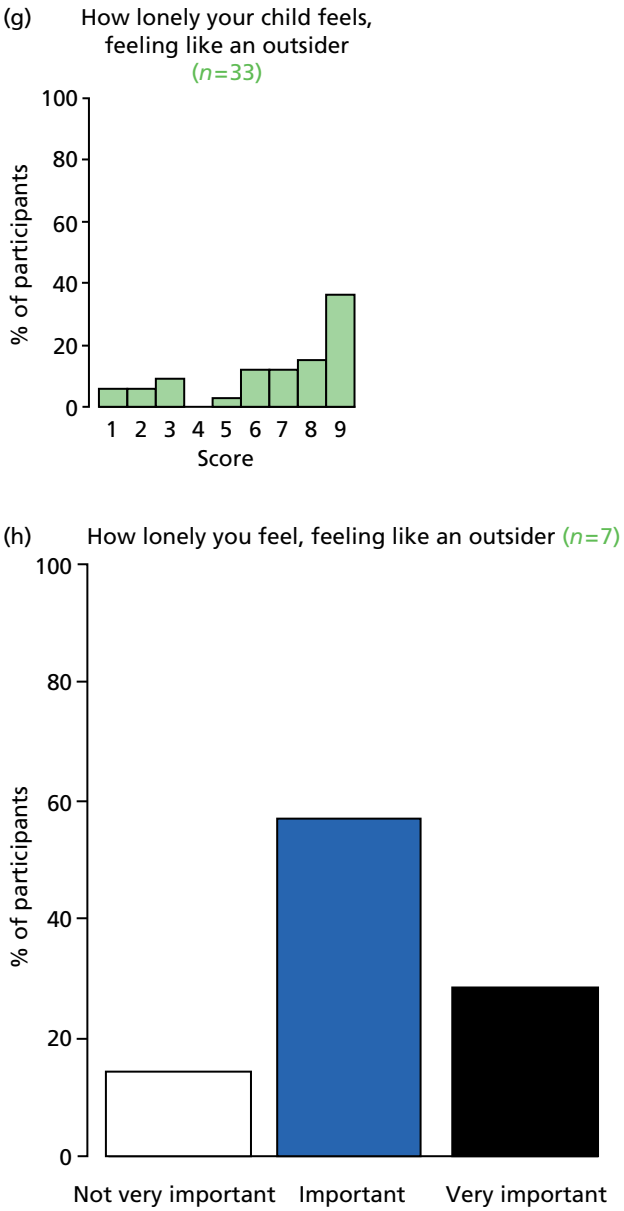


FIGURE 54 Outcome: internalising behaviour.

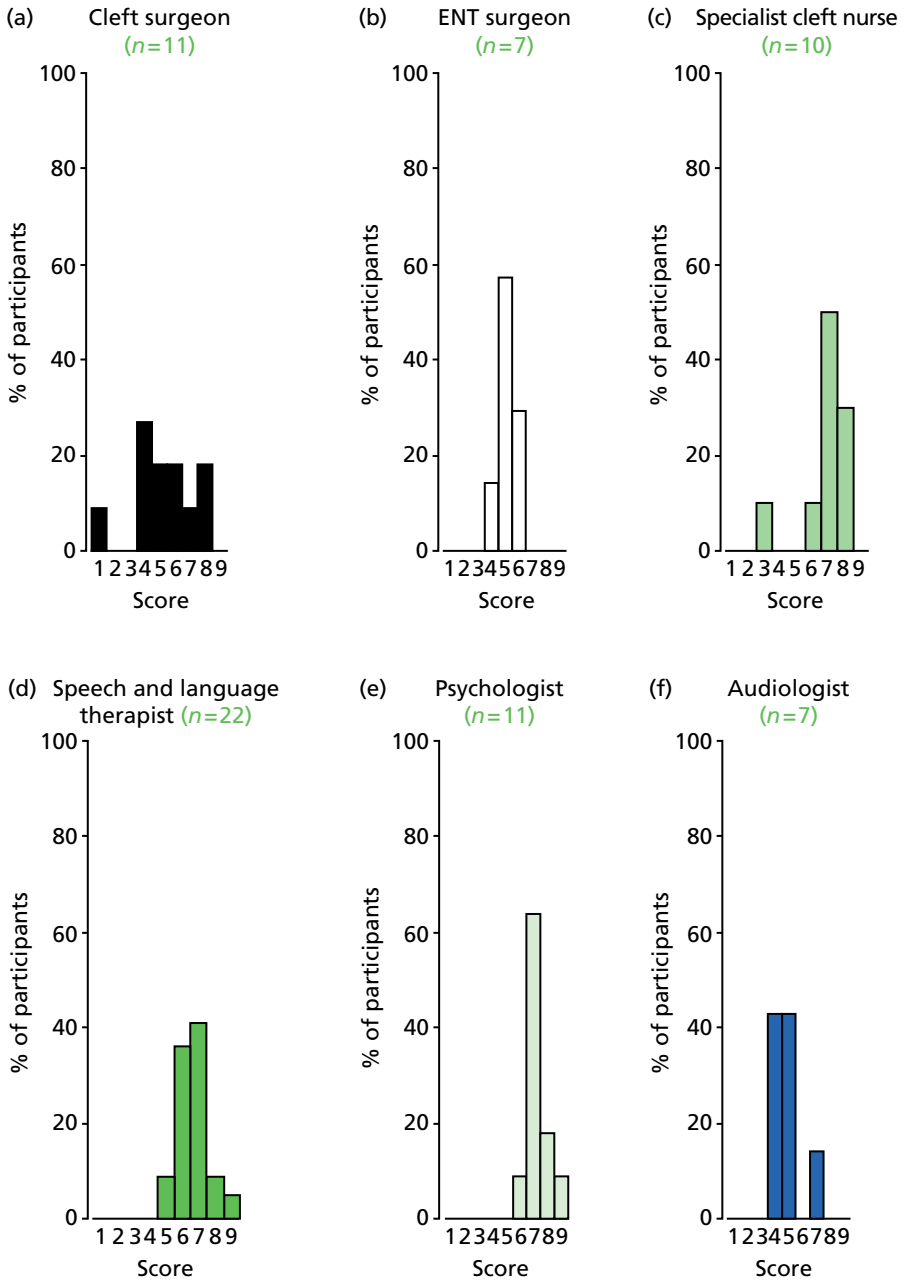


FIGURE 55 Outcome: externalising behaviour. (*continued*)

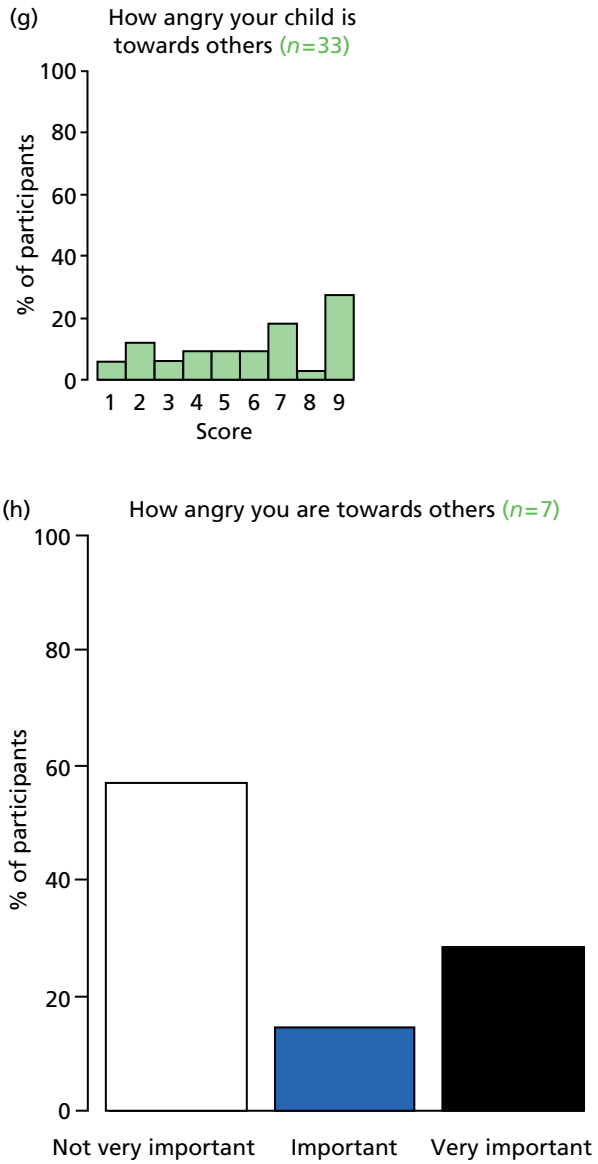


FIGURE 55 Outcome: externalising behaviour.

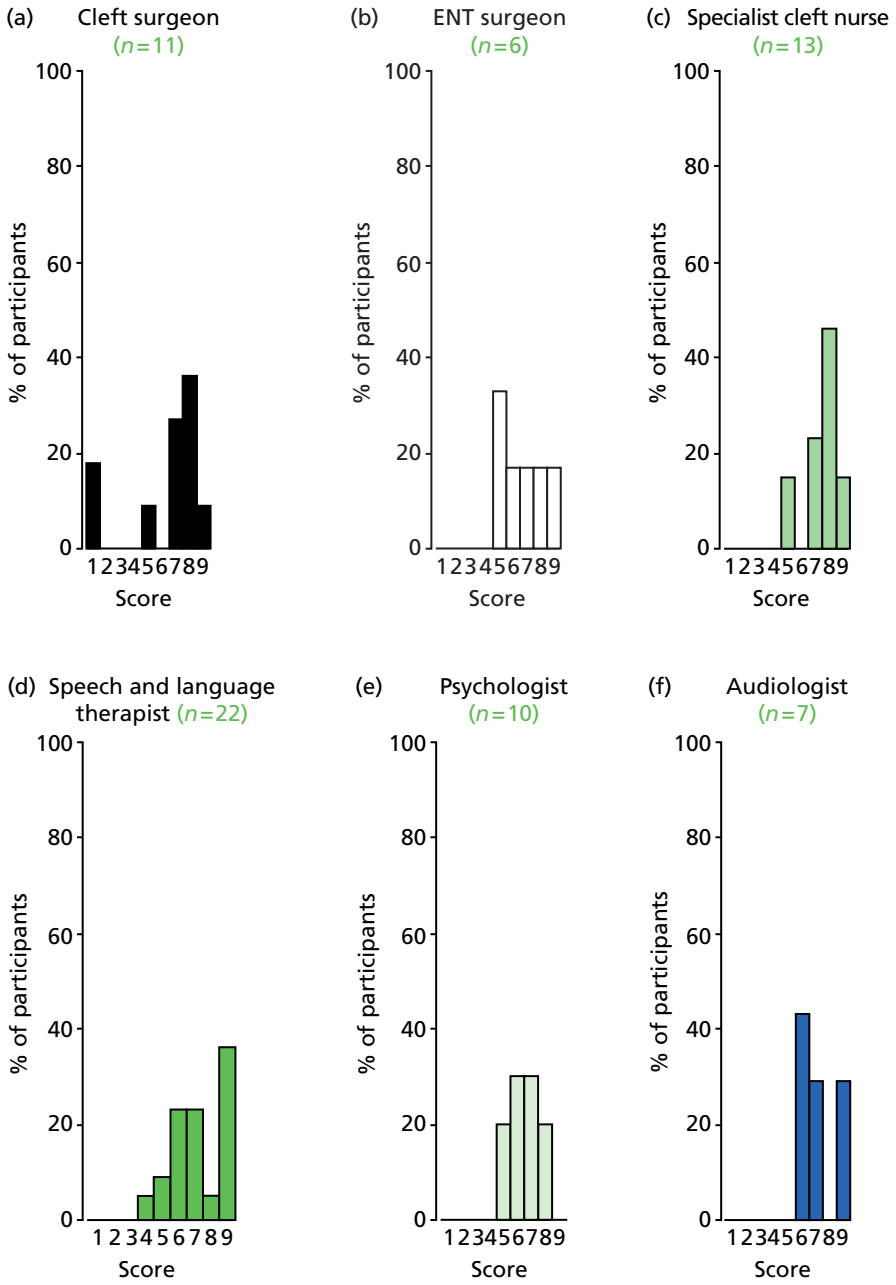


FIGURE 56 Outcome: academic achievement. (*continued*)

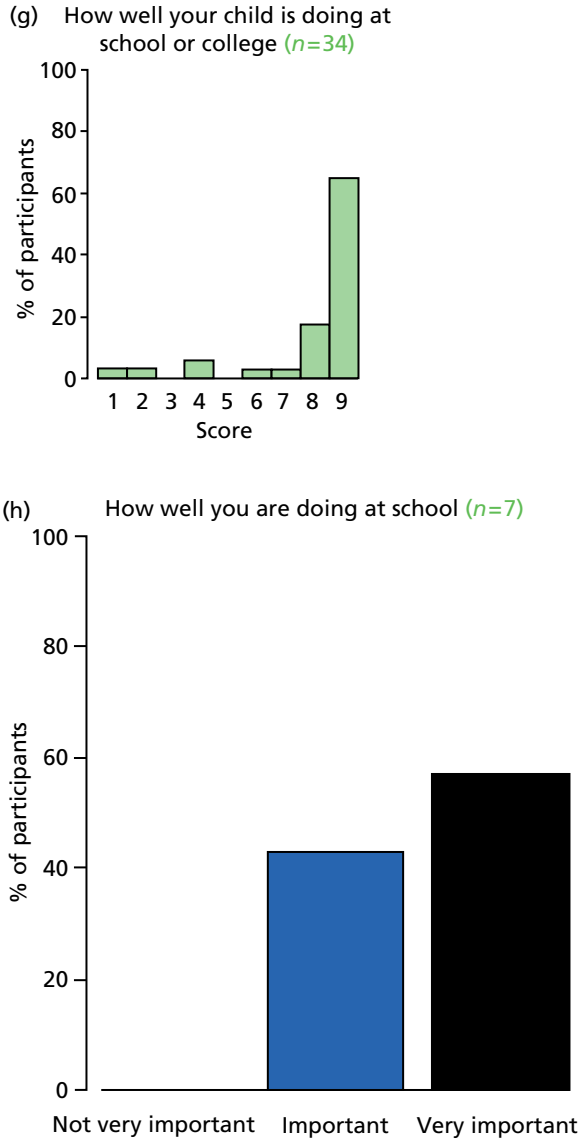


FIGURE 56 Outcome: academic achievement.

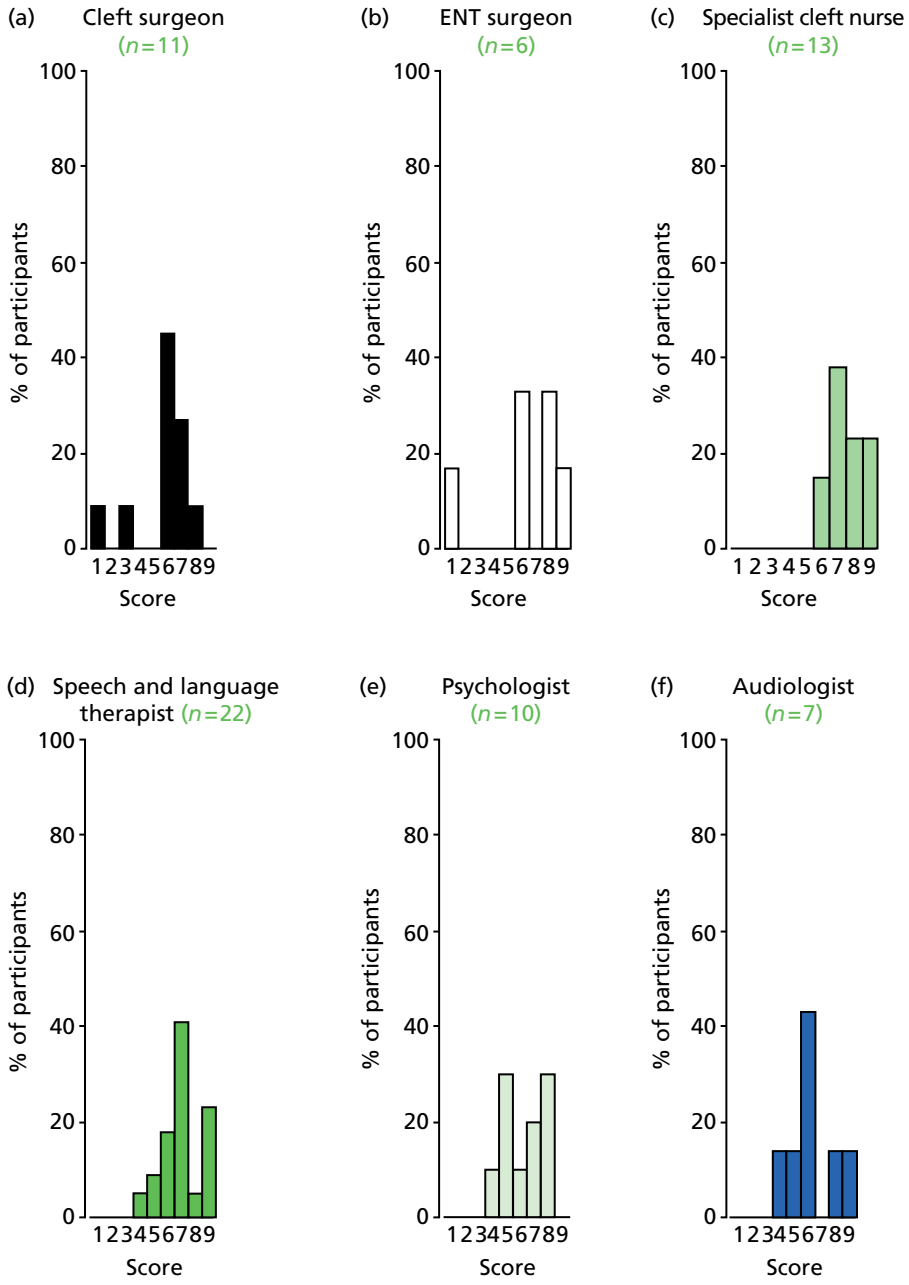


FIGURE 57 Outcome: literacy. (*continued*)

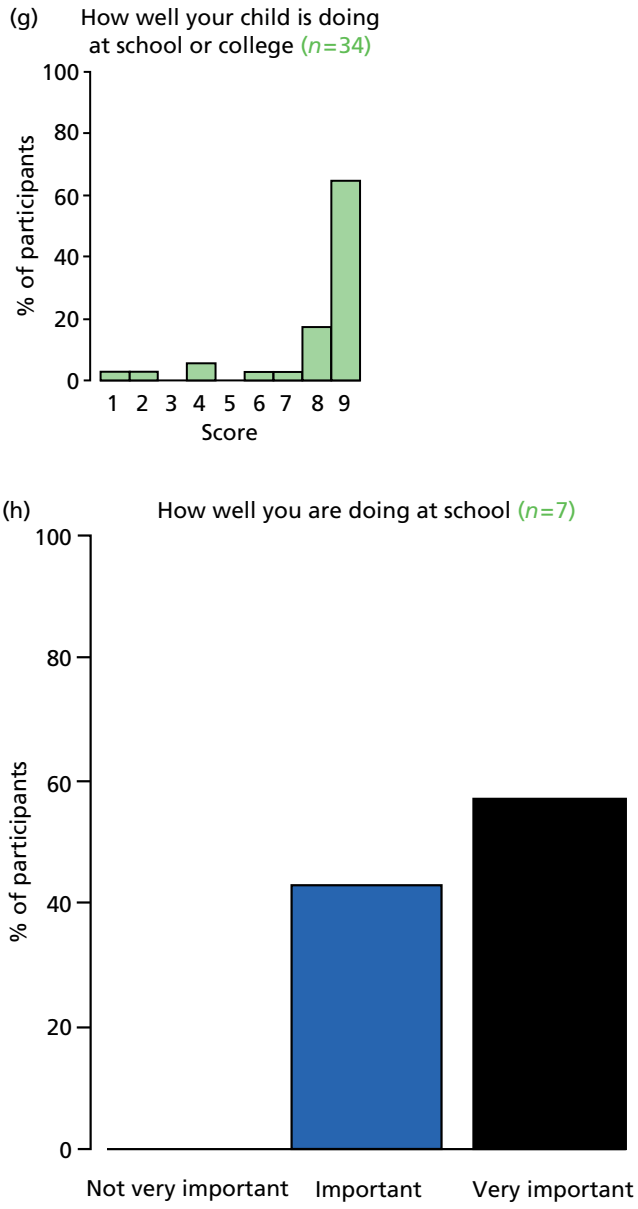


FIGURE 57 Outcome: literacy.

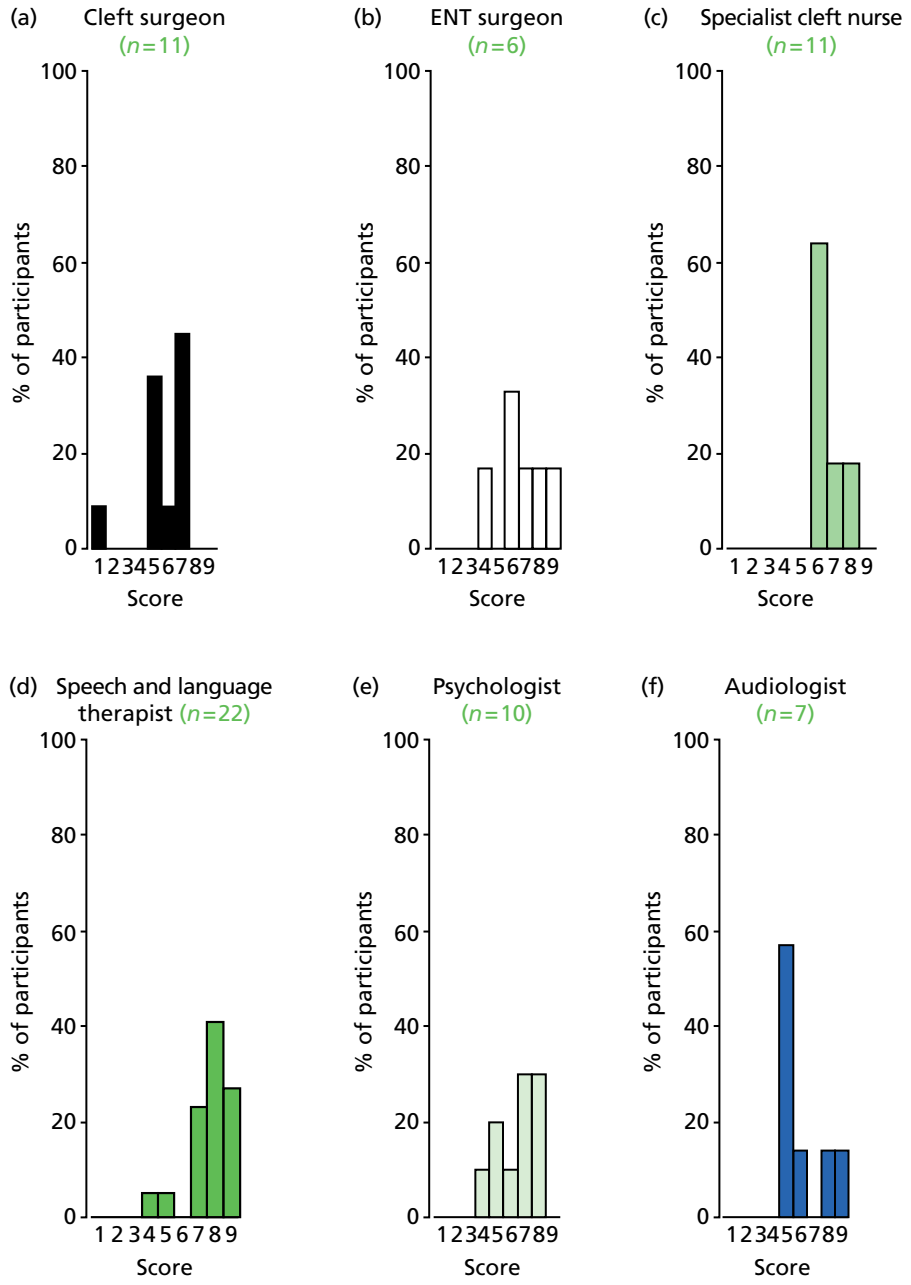


FIGURE 58 Outcome: phonological memory. (*continued*)

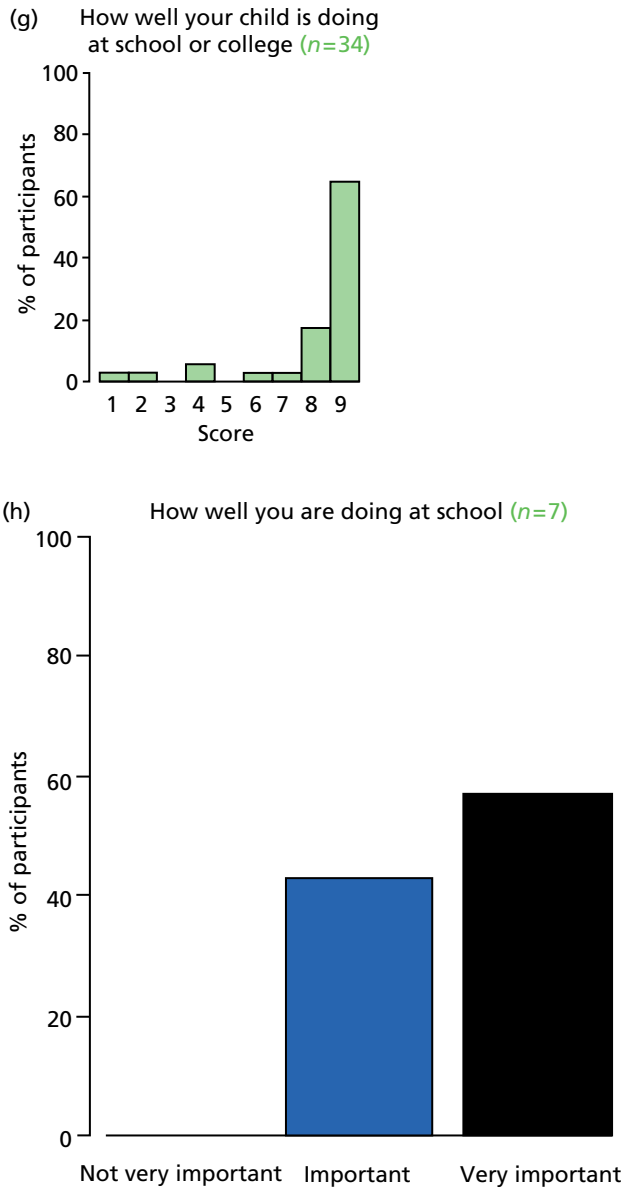


FIGURE 58 Outcome: phonological memory.

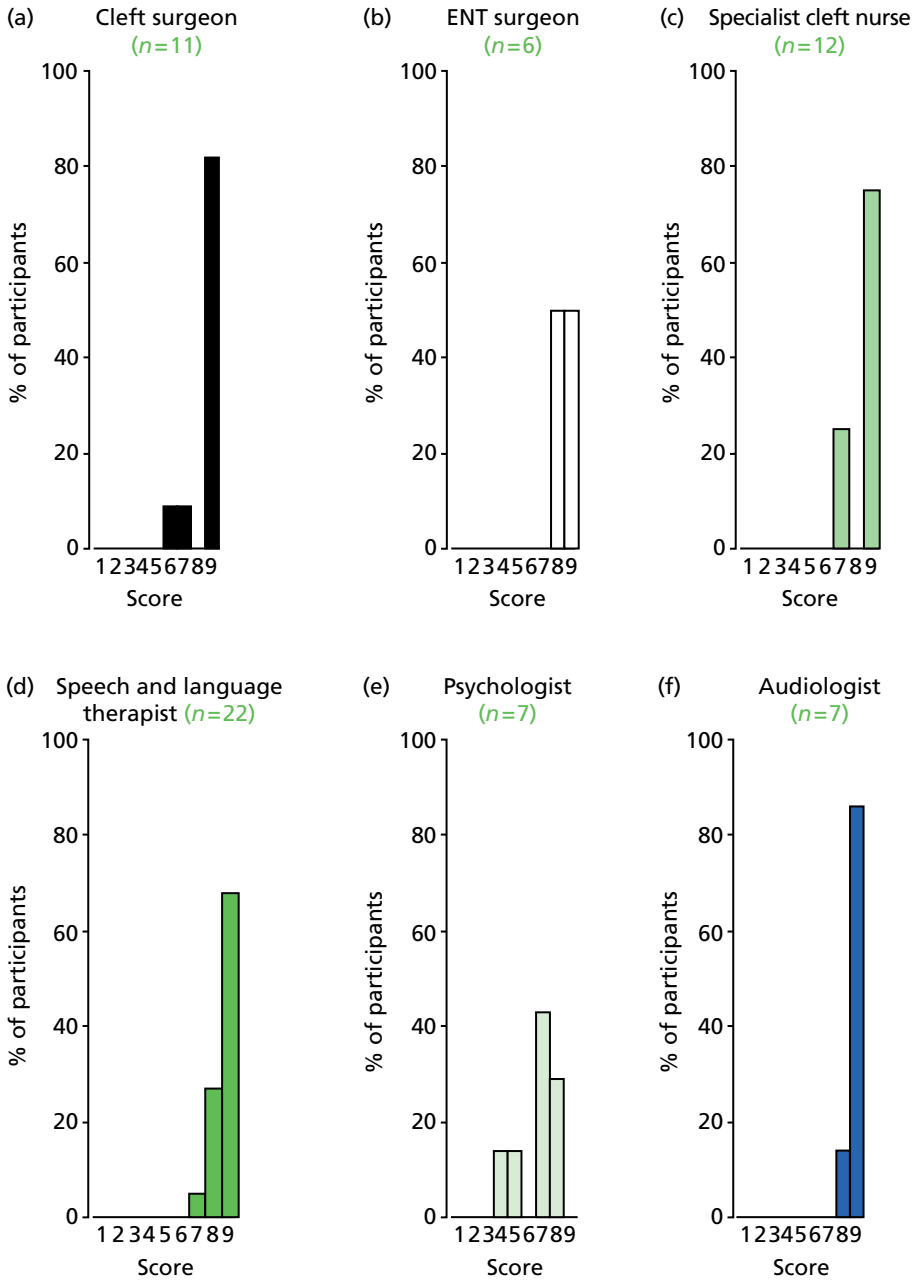


FIGURE 59 Outcome: hearing. (*continued*)

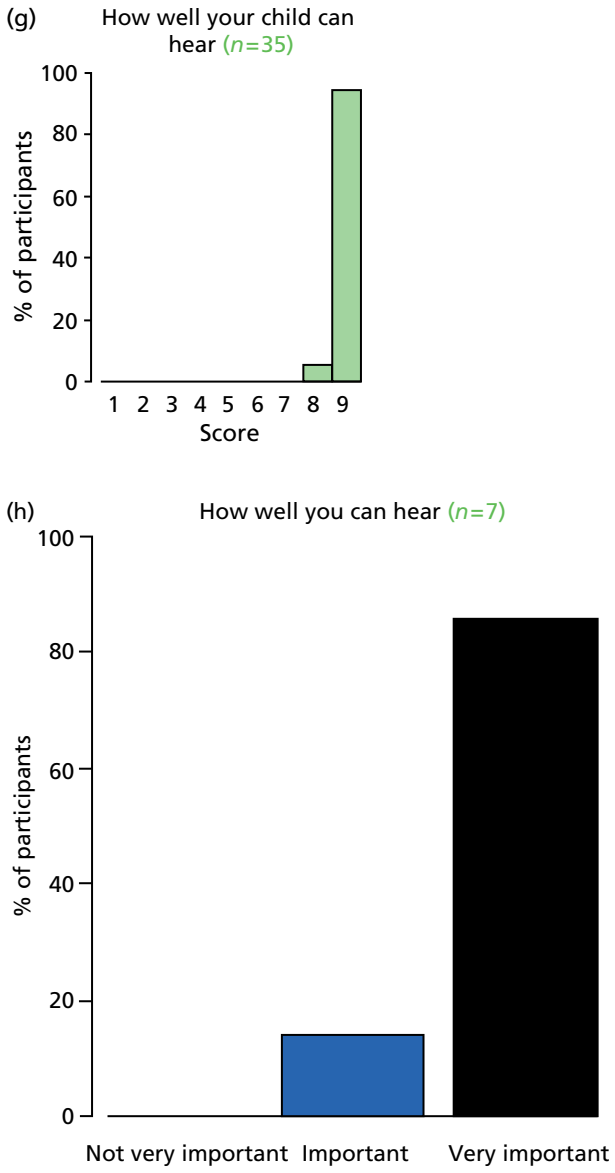


FIGURE 59 Outcome: hearing.

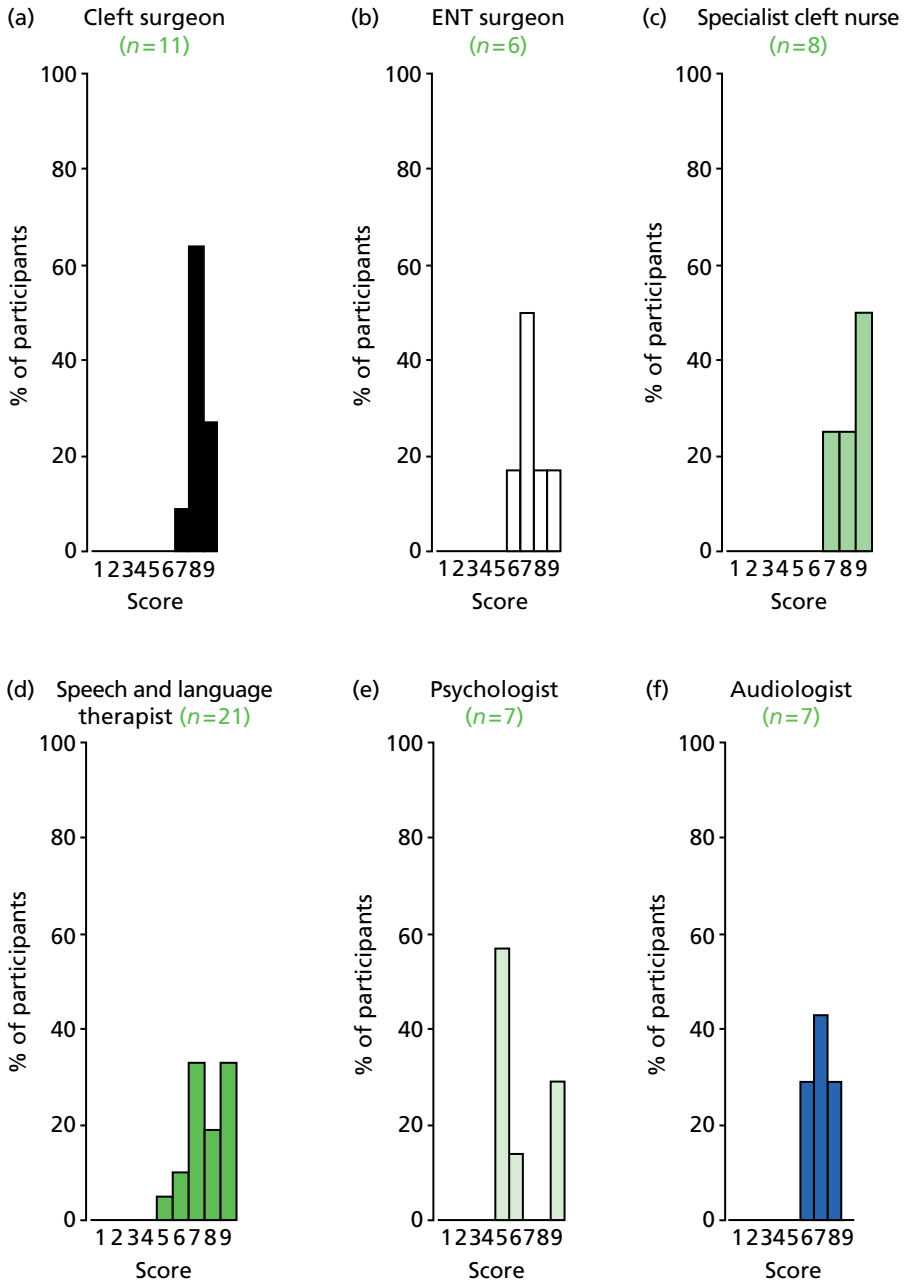


FIGURE 60 Outcome: chronic otitis media. (*continued*)

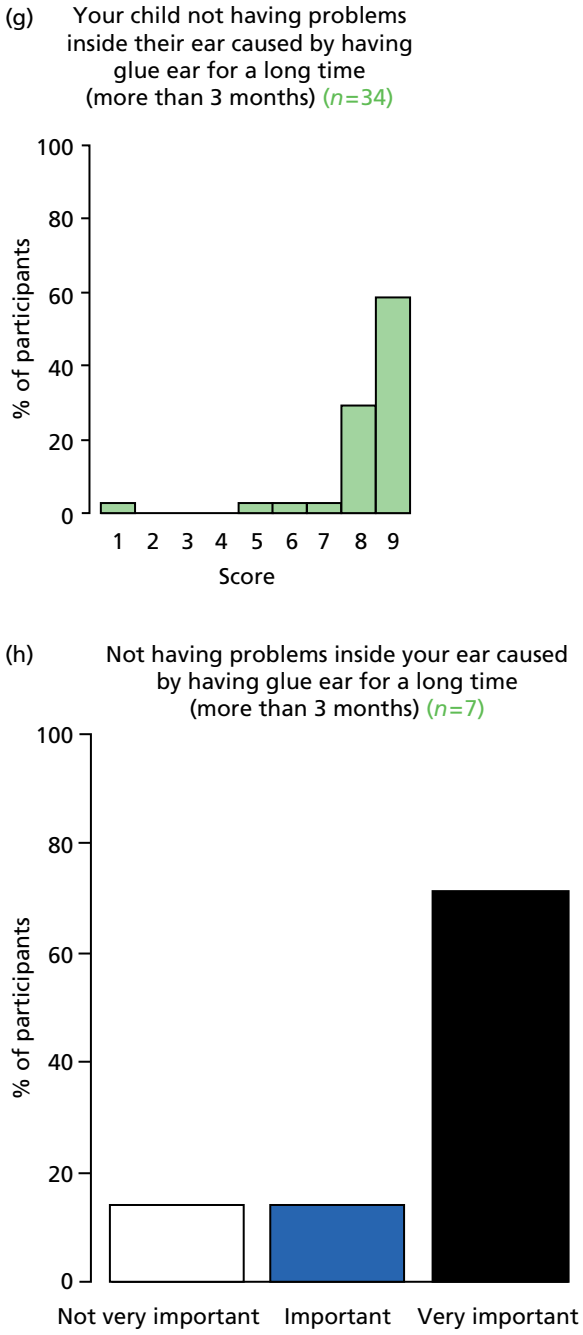


FIGURE 60 Outcome: chronic otitis media.

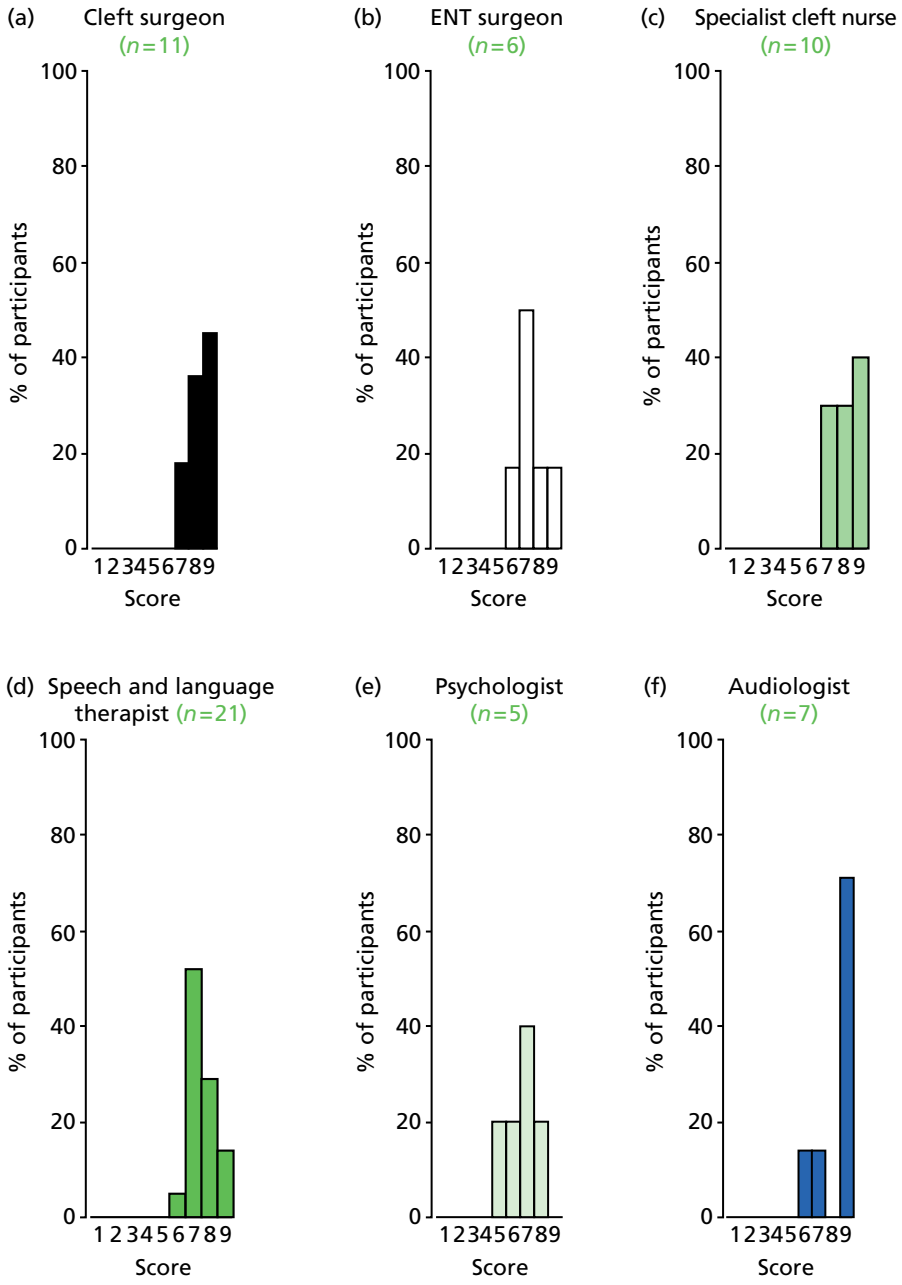


FIGURE 61 Outcome: OME. (*continued*)

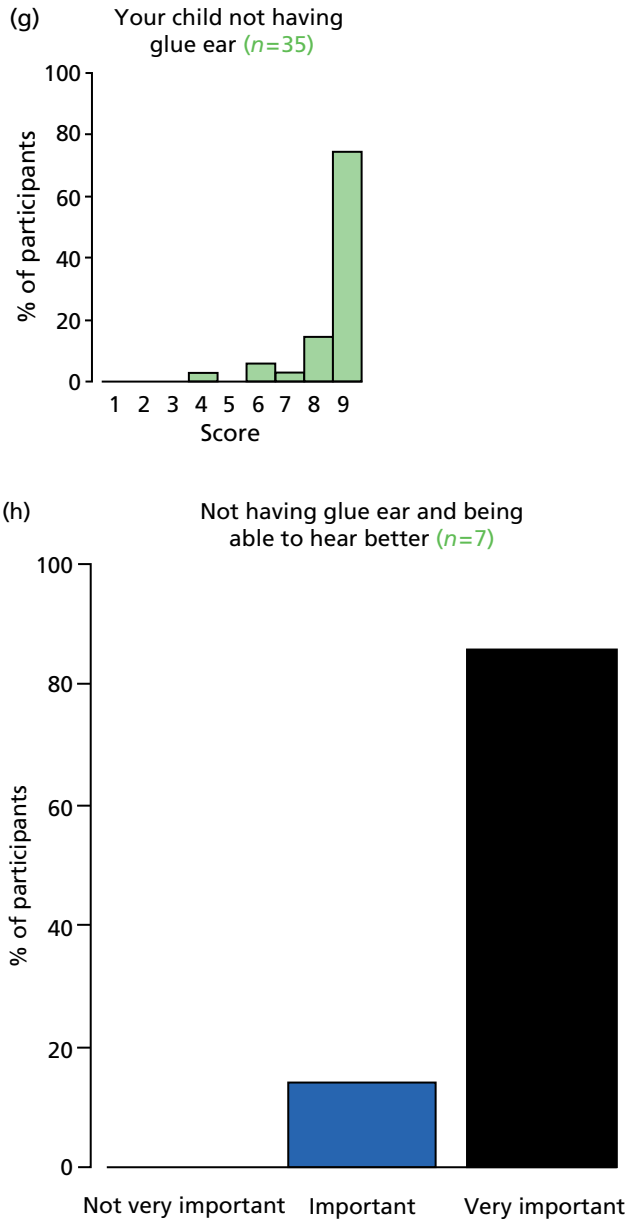


FIGURE 61 Outcome: OME.

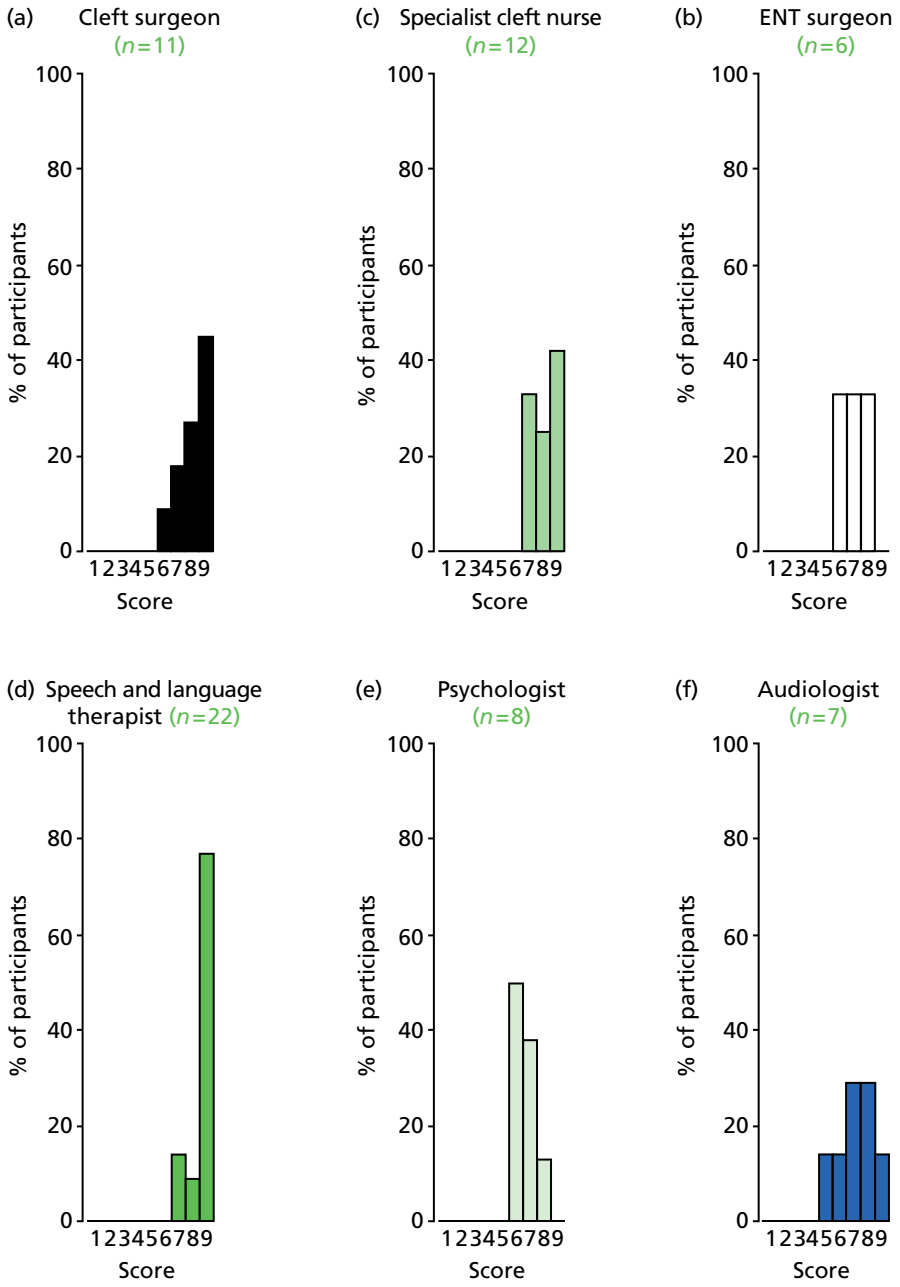


FIGURE 62 Outcome: receptive language skills. (*continued*)

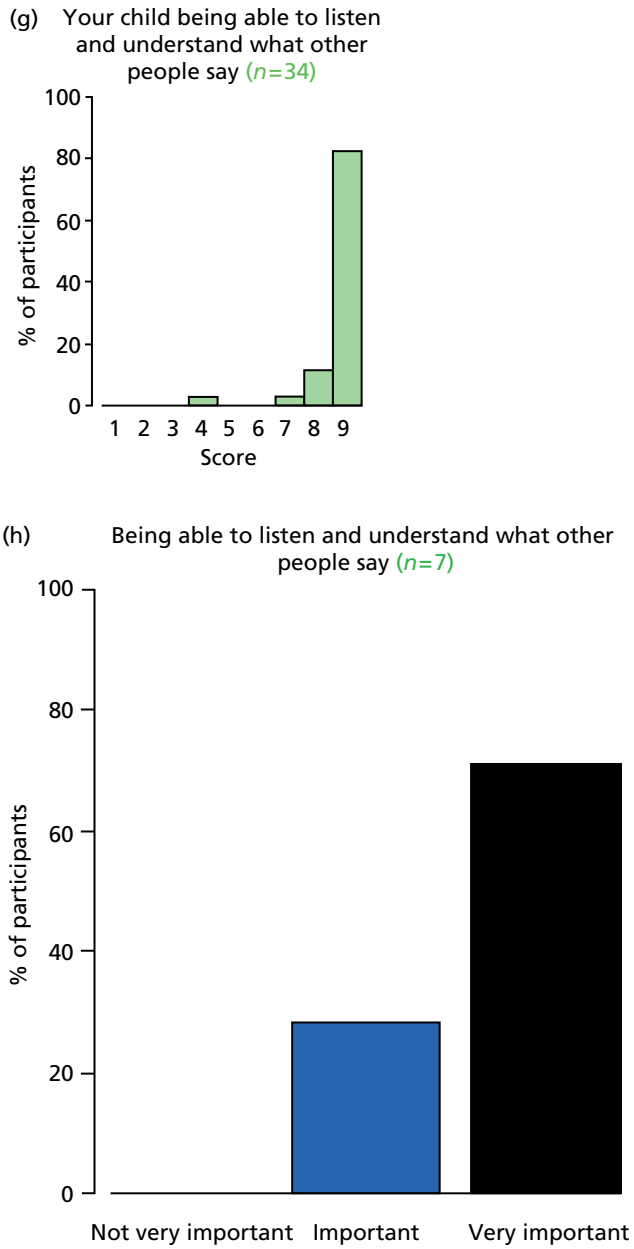


FIGURE 62 Outcome: receptive language skills.

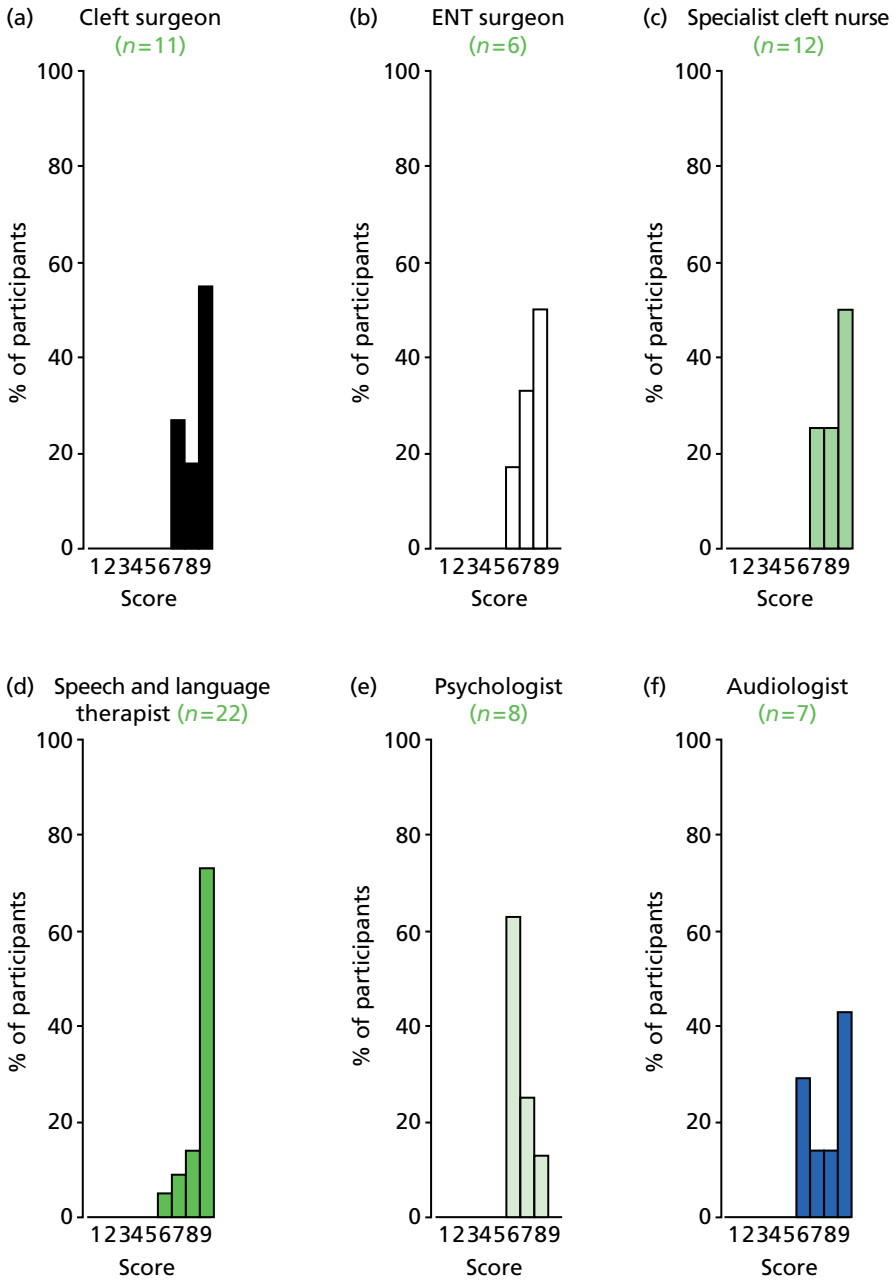


FIGURE 63 Outcome: speech development. (*continued*)

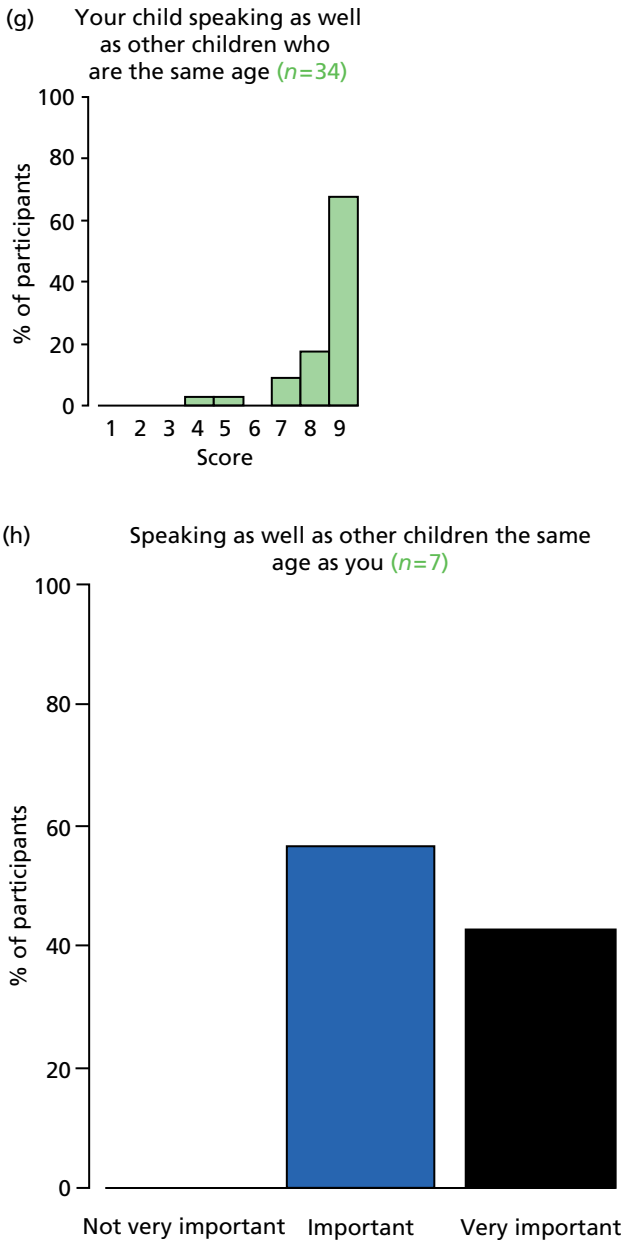


FIGURE 63 Outcome: speech development.

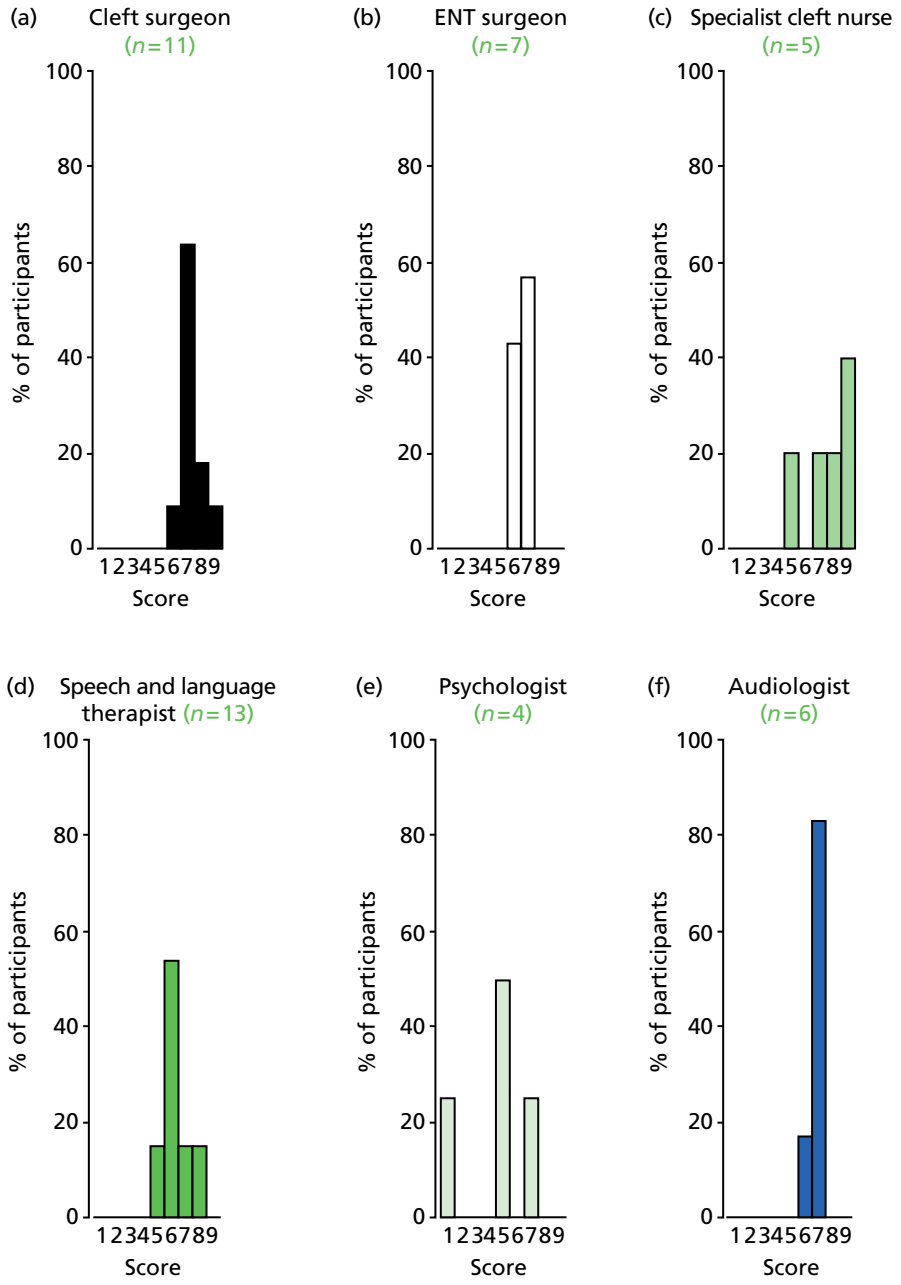


FIGURE 64 Outcome: atelectasis. (*continued*)

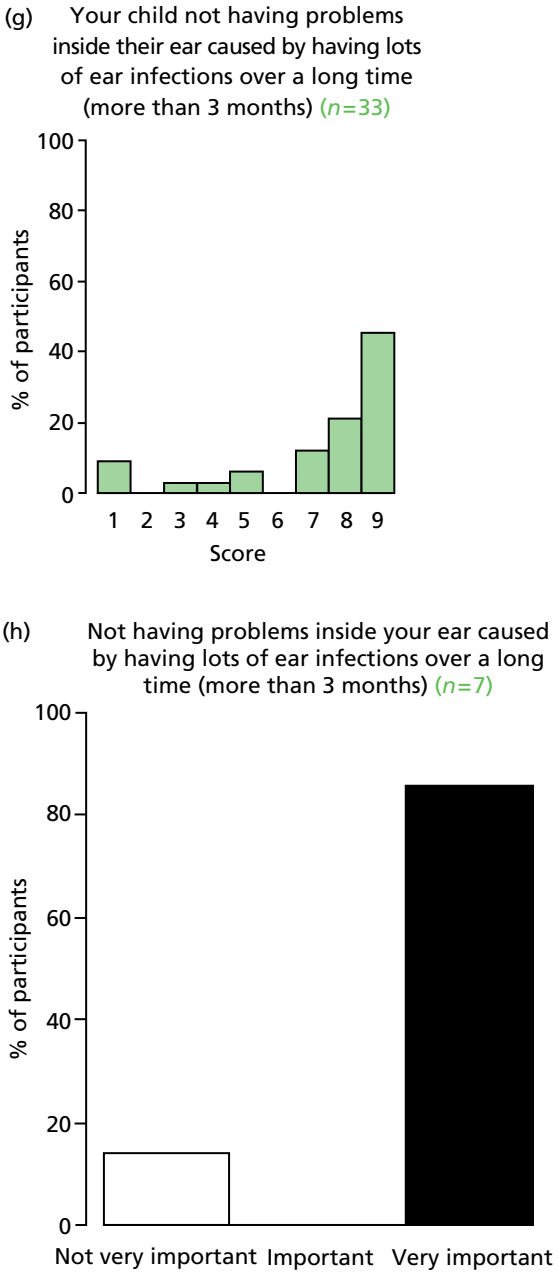


FIGURE 64 Outcome: atelectasis.

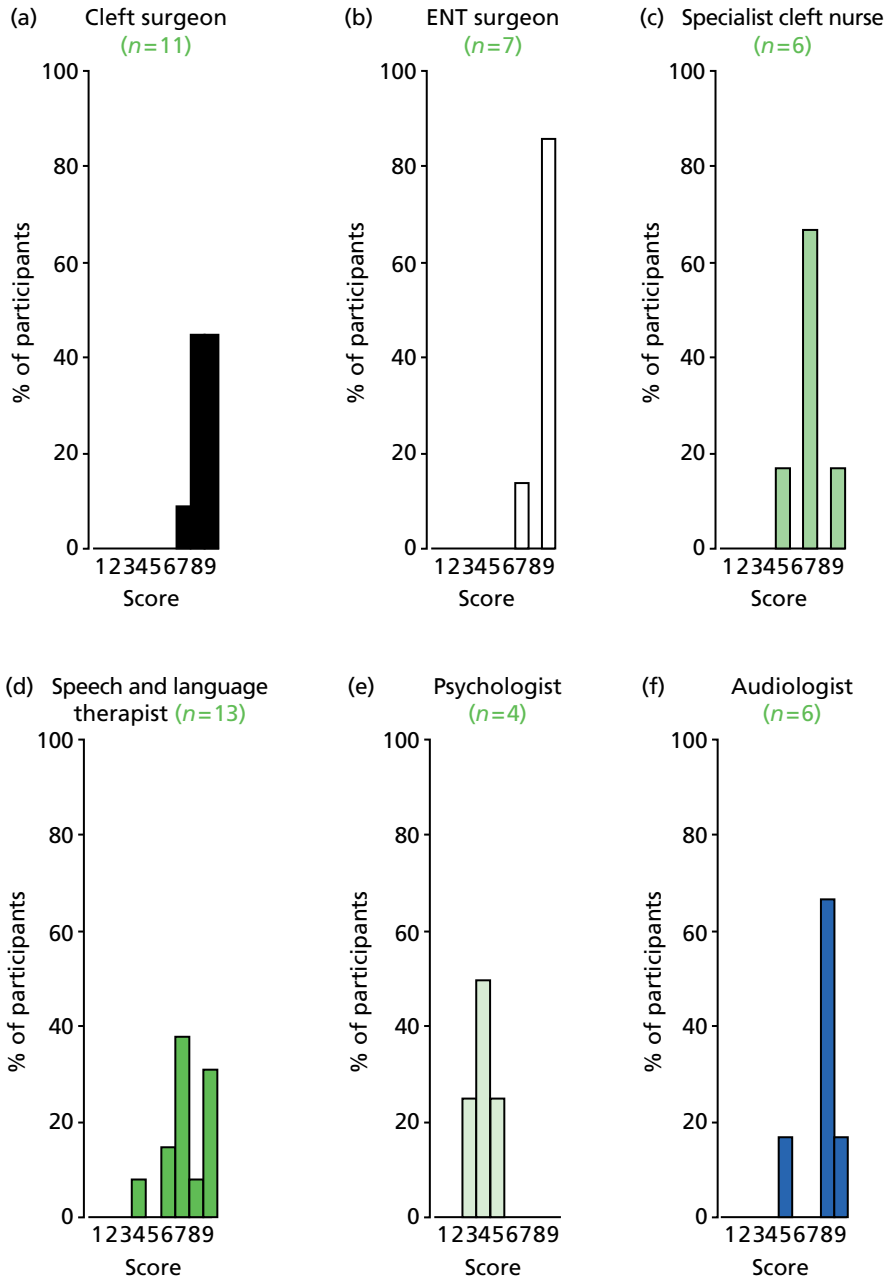


FIGURE 65 Outcome: cholesteatoma. (*continued*)

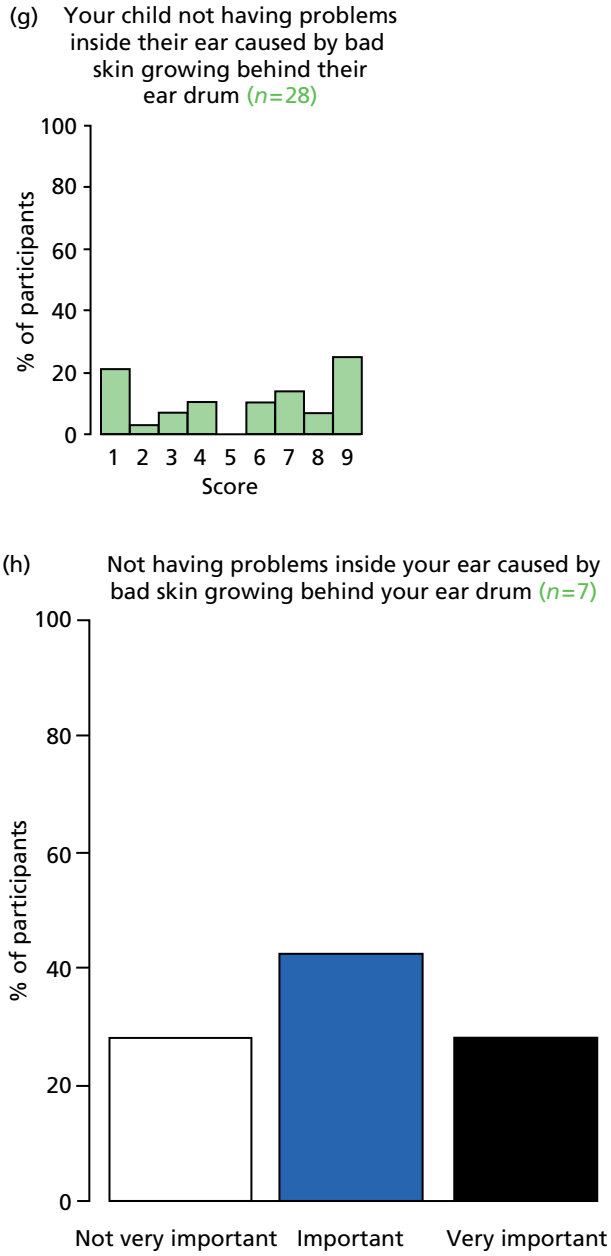


FIGURE 65 Outcome: cholesteatoma.

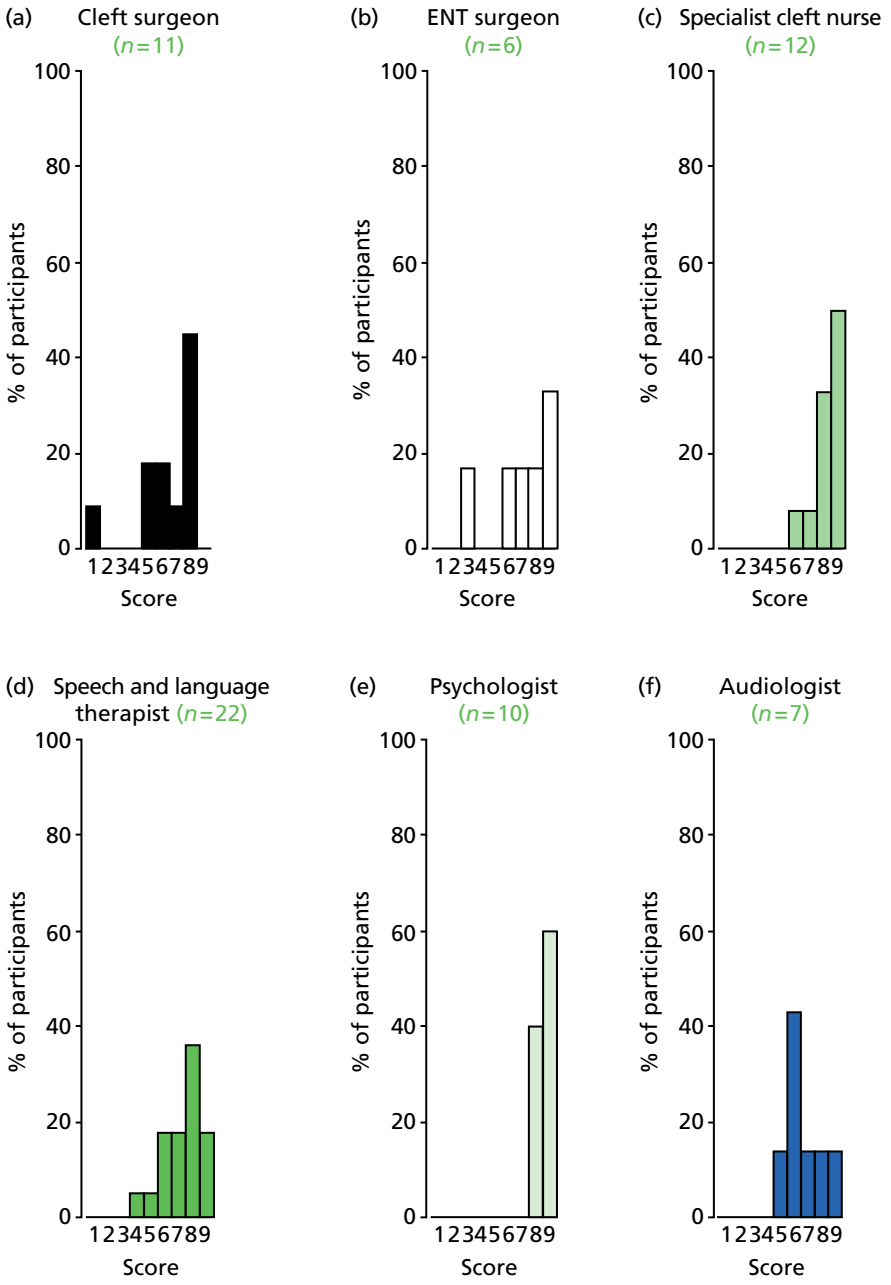


FIGURE 66 Outcome: psychosocial development. (*continued*)

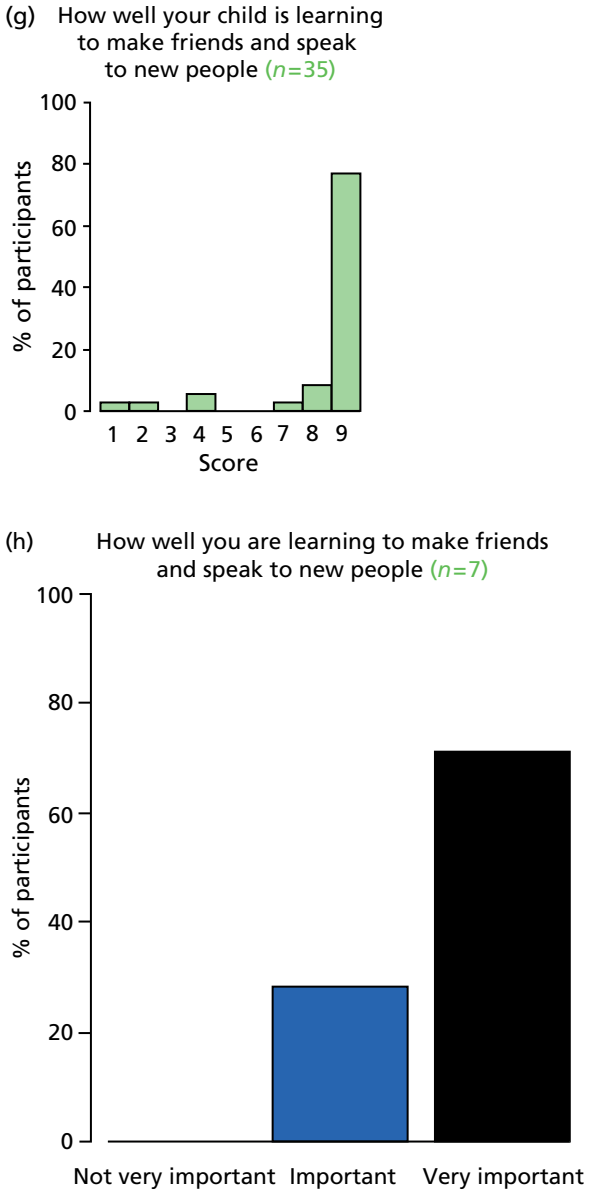


FIGURE 66 Outcome: psychosocial development.

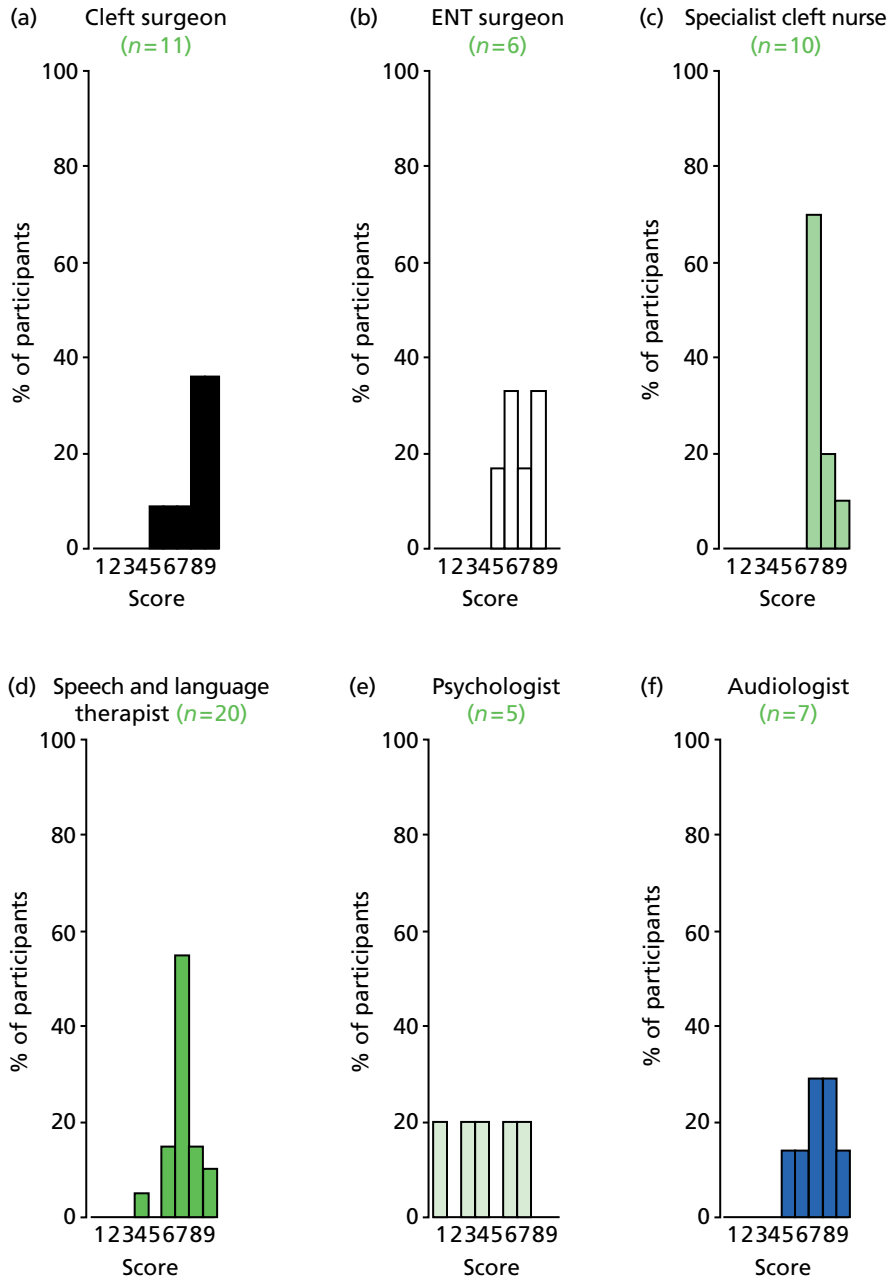


FIGURE 67 Outcome: AOM. (*continued*)

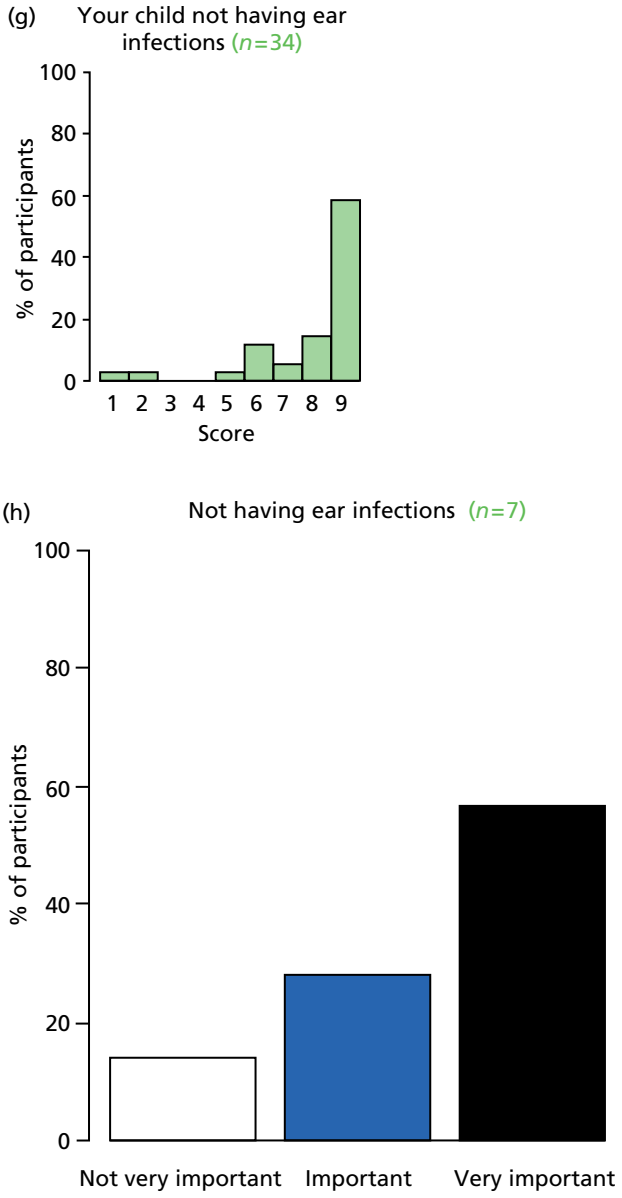


FIGURE 67 Outcome: AOM.

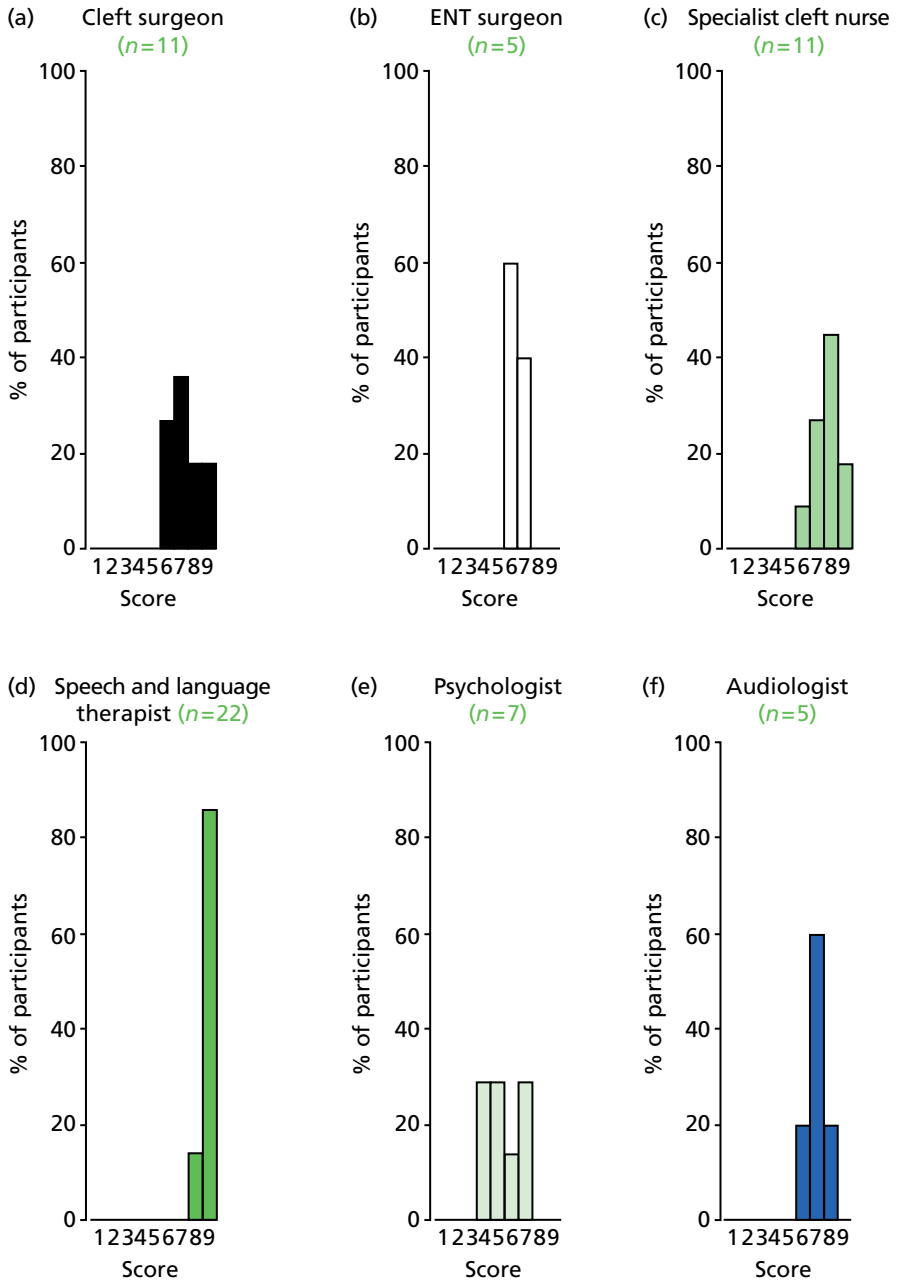


FIGURE 68 Outcome: consonant production. (*continued*)

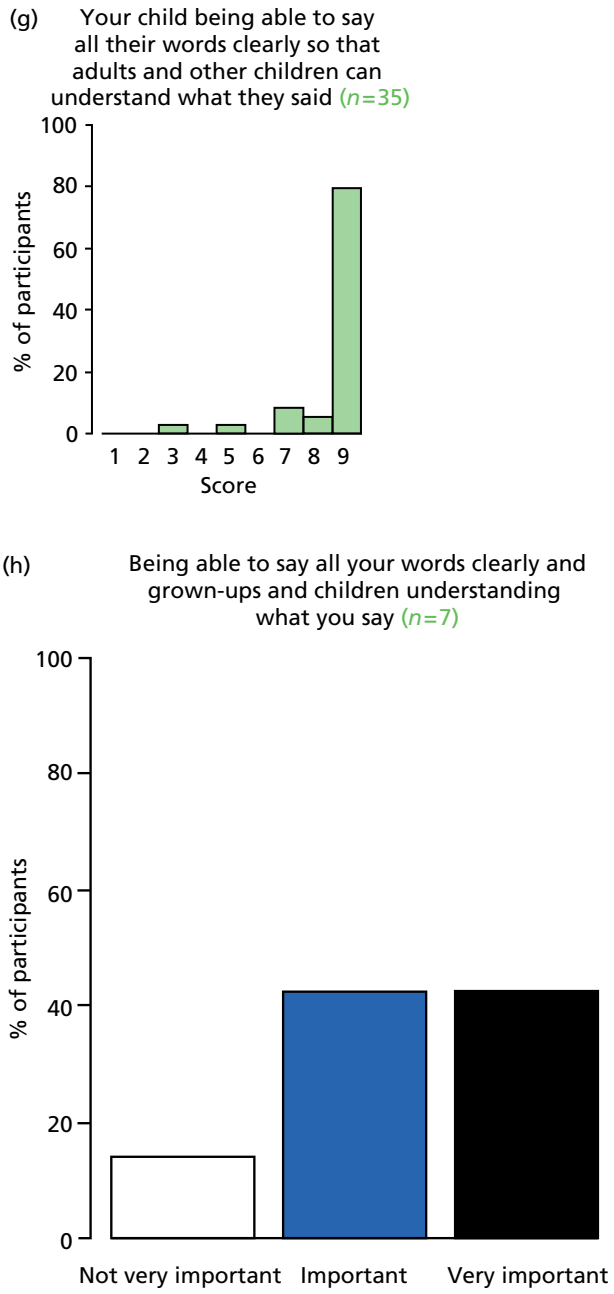


FIGURE 68 Outcome: consonant production.

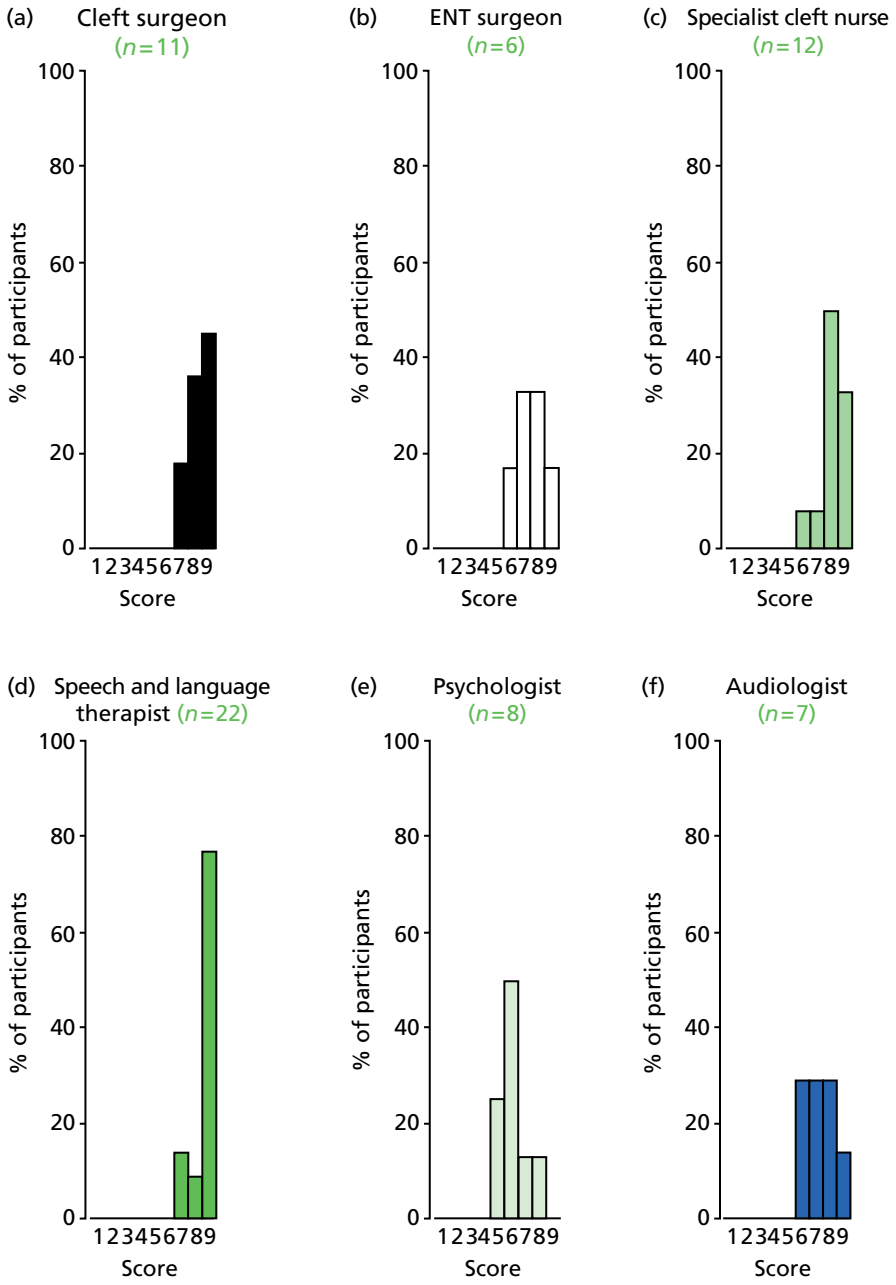


FIGURE 69 Outcome: speech intelligibility. (*continued*)

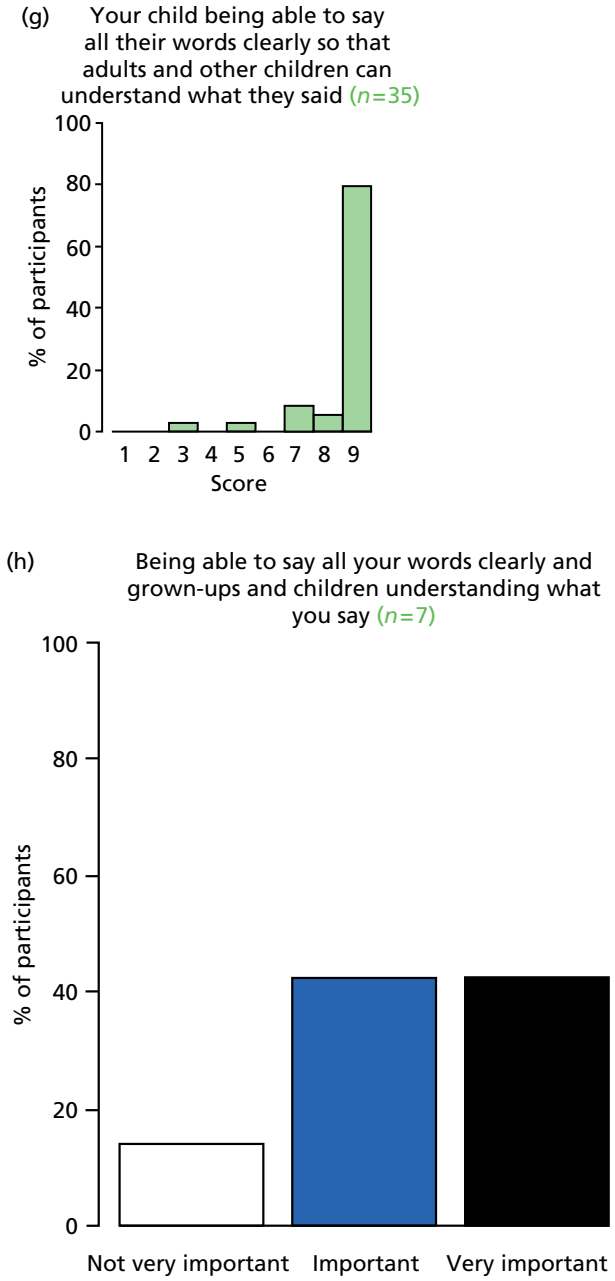


FIGURE 69 Outcome: speech intelligibility.

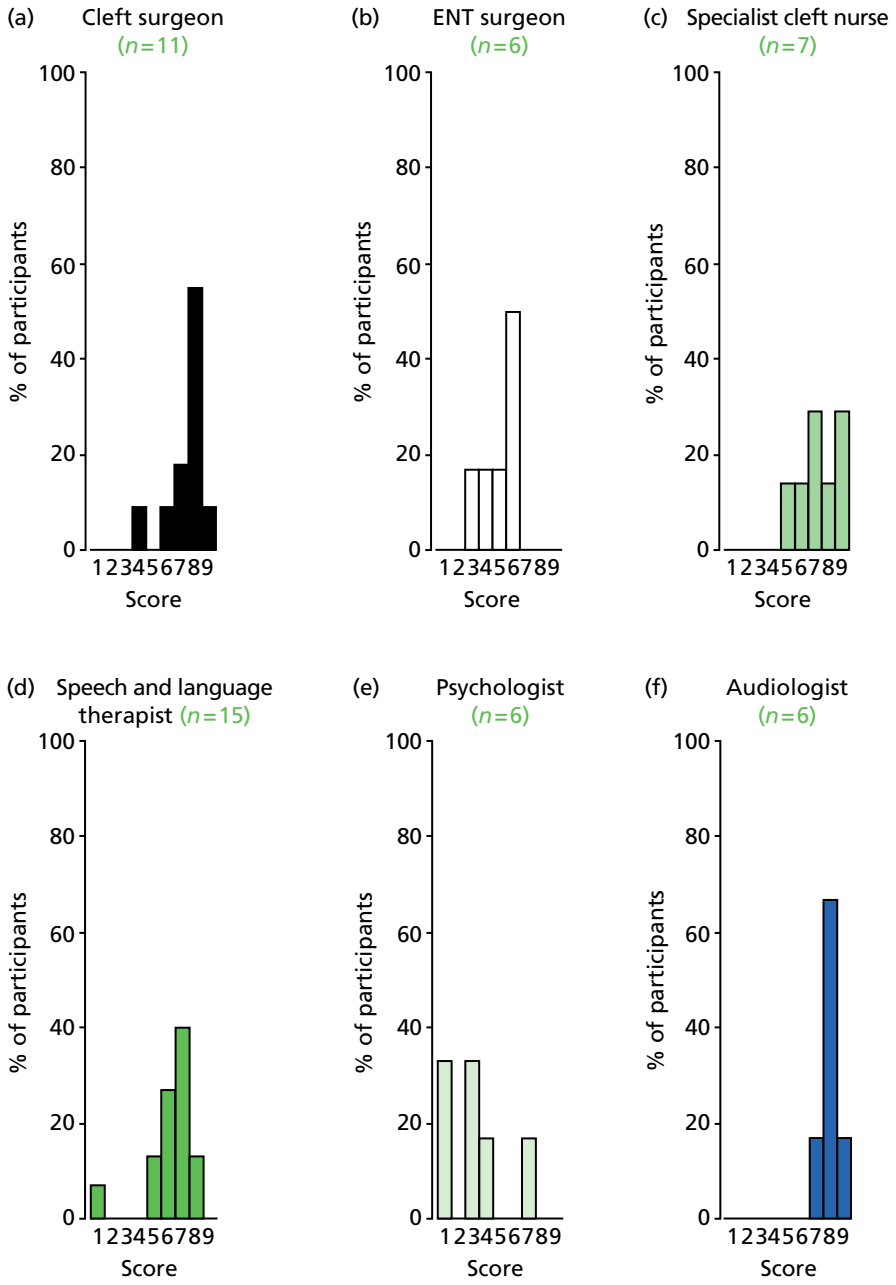


FIGURE 70 Outcome: necessity to remove VTs. (*continued*)

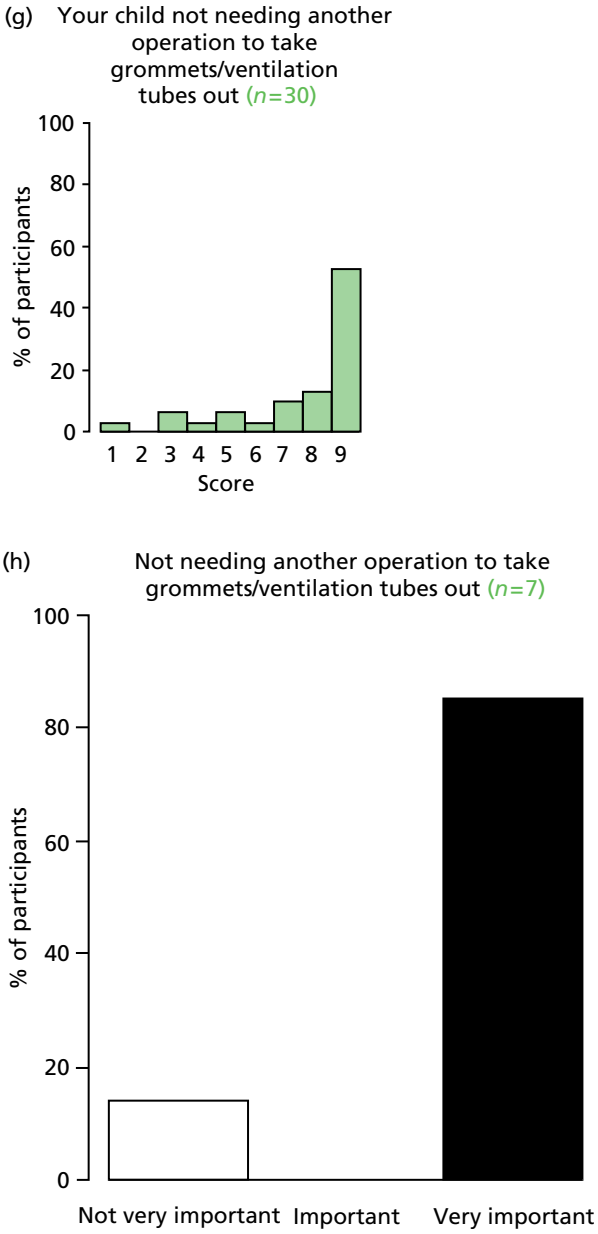


FIGURE 70 Outcome: necessity to remove VTs.

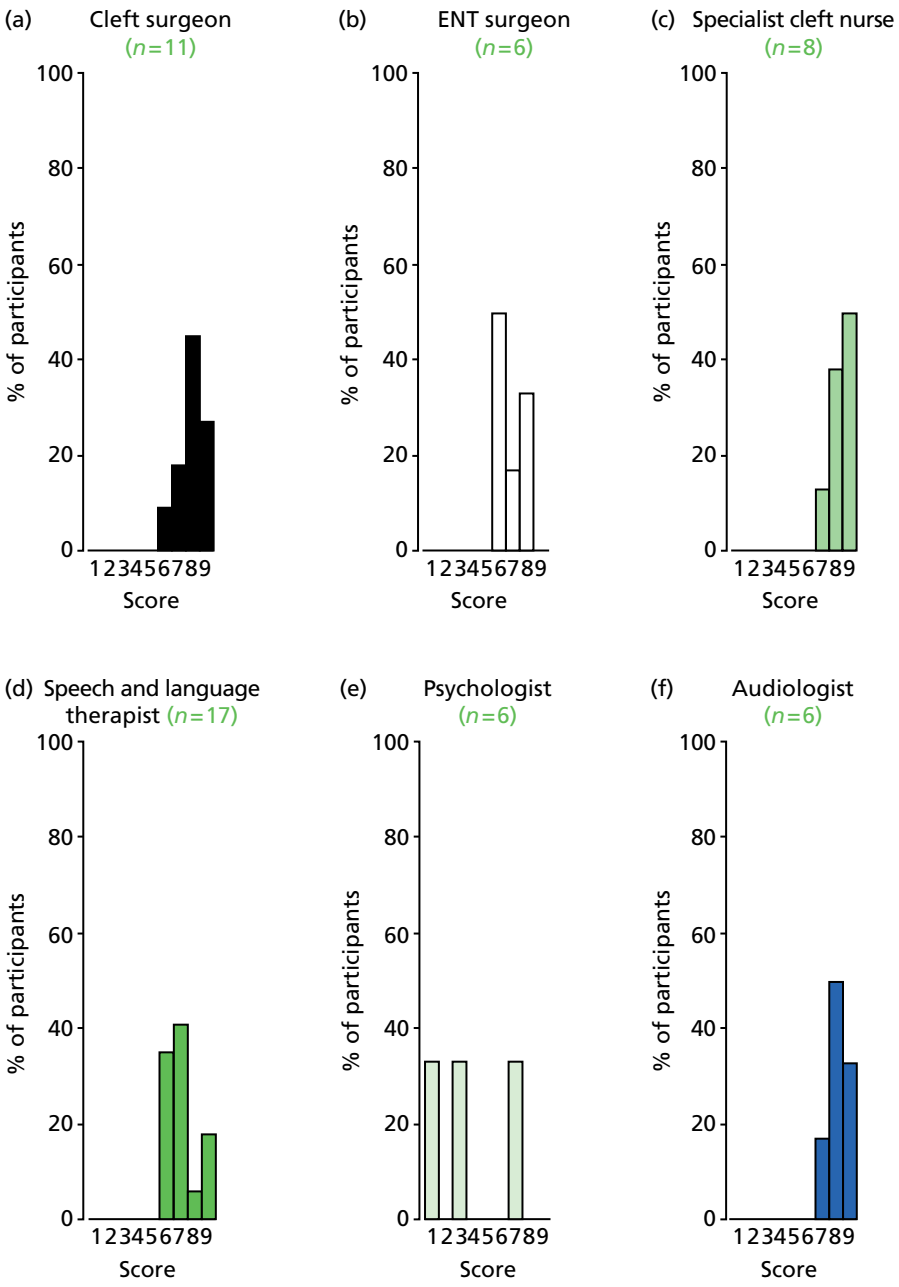


FIGURE 71 Outcome: requirement for repeated VTs. (*continued*)

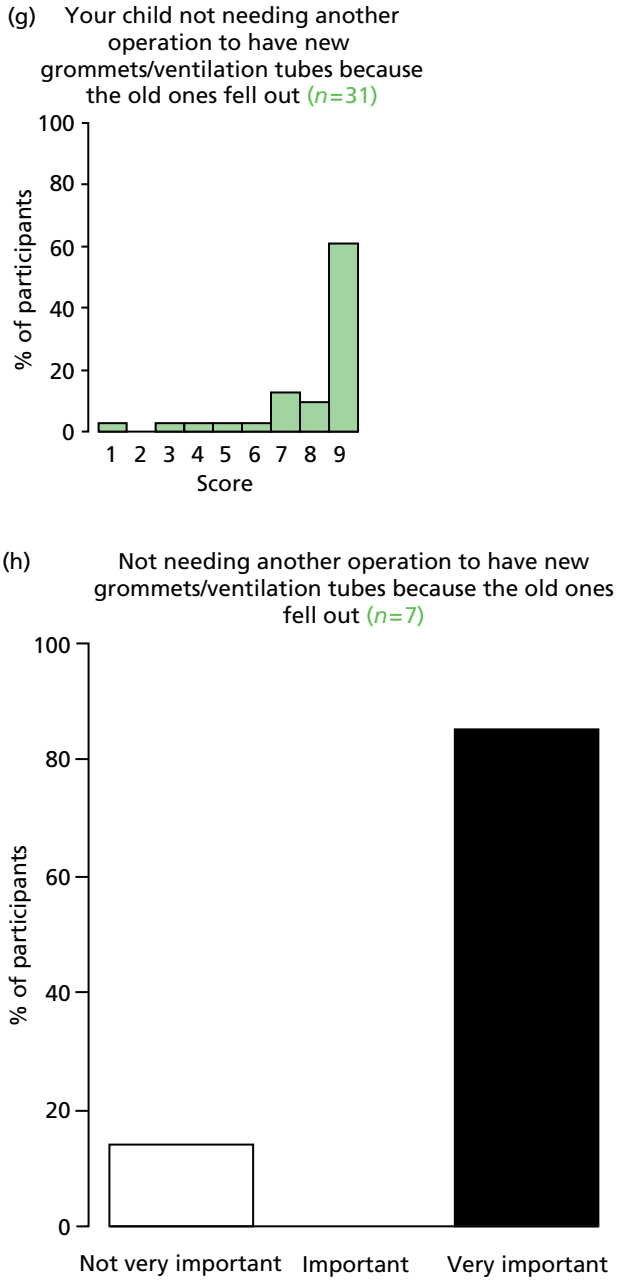


FIGURE 71 Outcome: requirement for repeated VTs.

Appendix 9 Individual outcome discussions and results of scoring at consensus meeting

Note: the scores of participants scoring 1–9 only have been included. The figure for the number of participants scoring 1–9 has been given together with the number who answered 'unable to score'. Some meeting participants left the meeting early and the outcomes affected by this are marked with an asterisk.

Outcome number	16
Outcome name	Hearing
Number of participants scoring 1–9	14
Number of participants unable to score	0

Notes
All eight of the stakeholder groups had reached consensus that hearing was an important outcome prior to the meeting. Consistency of hearing was raised as being important by JH in reference to the requirement to adjust HAs based on fluctuations in hearing level. TB supported this but all agreed that this is a component of the outcome 'hearing' and would be addressed in discussions on 'how' this outcome should be measured.

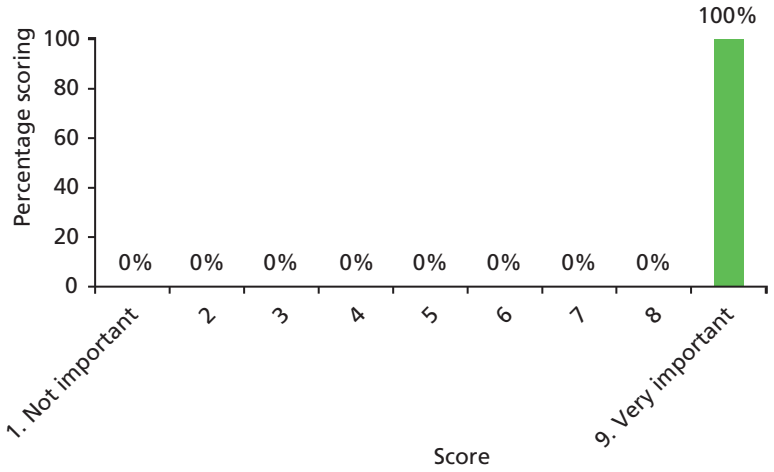


FIGURE 72 Results of scoring: hearing – how well your child can hear.

Outcome number	7
Outcome name	COM
Number of participants scoring 1–9	14
Number of participants unable to score	0

COM, chronic otitis media.
Notes
Nothing raised about COM.

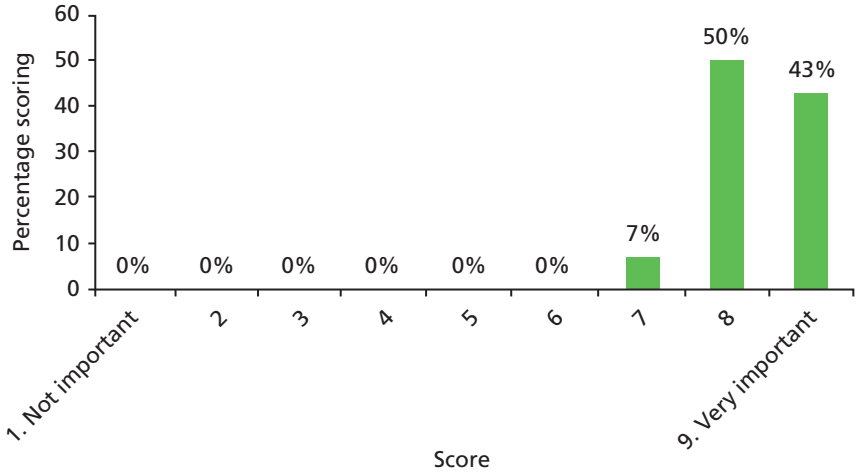


FIGURE 73 Results of scoring: chronic otitis media – your child not having problems inside their ear that can be caused by having glue ear for a long time (more than 3 months).

Outcome number	26
Outcome name	OME
Number of participants scoring 1–9	14
Number of participants unable to score	0

Notes
AHB raised that the frequency of OME was important. All agreed that this should be addressed in how the outcome is measured.

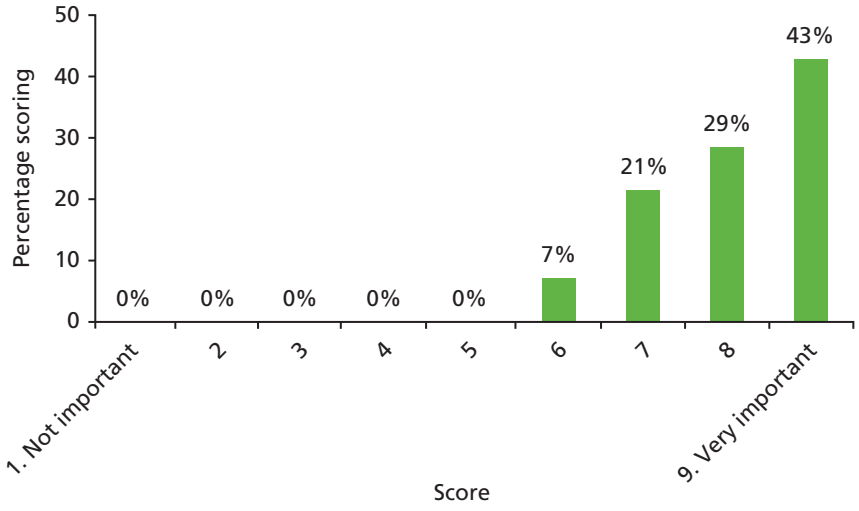


FIGURE 74 Results of scoring: OME – your child not having glue ear.

Outcome number	30
Outcome name	Speech intelligibility
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
No discussion. 'Speech intelligibility', 'consonant production' and 'expressive language skills' were scored by parents and children as one outcome: 'your child being able to say all of their words clearly so that adults and other children can understand what they said'. Agreed that the outcomes scored as one 'your child being able to say all of their words clearly so that adults and other children can understand what they said' should be considered part of the 'how' of speech development and this will be discussed further with the SAG.

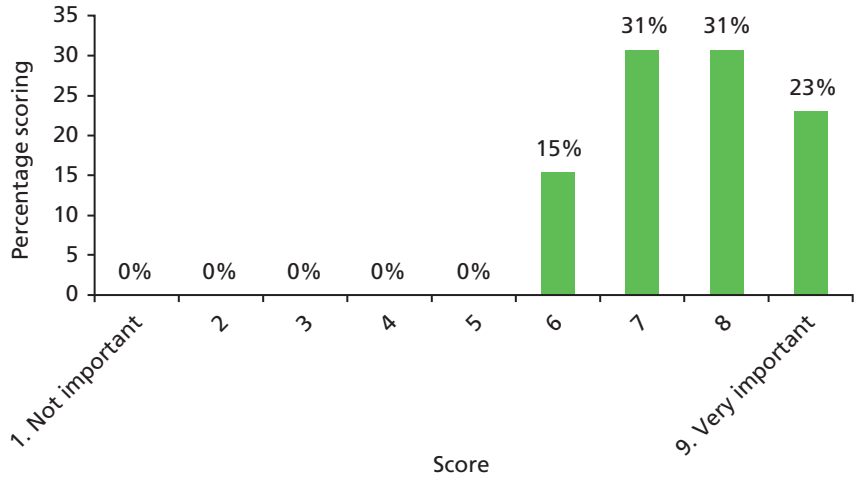


FIGURE 75 Results of scoring: speech intelligibility – your child being able to say all of their words clearly so that adults and other children can understand what they said.

Outcome number	33
Outcome name	Receptive language skills
Number of participants scoring 1–9	14
Number of participants unable to score	0

Notes
FJ highlighted that this would be used as part of the decision-making process for provision of treatment. CH commented that this is an easily measurable outcome.

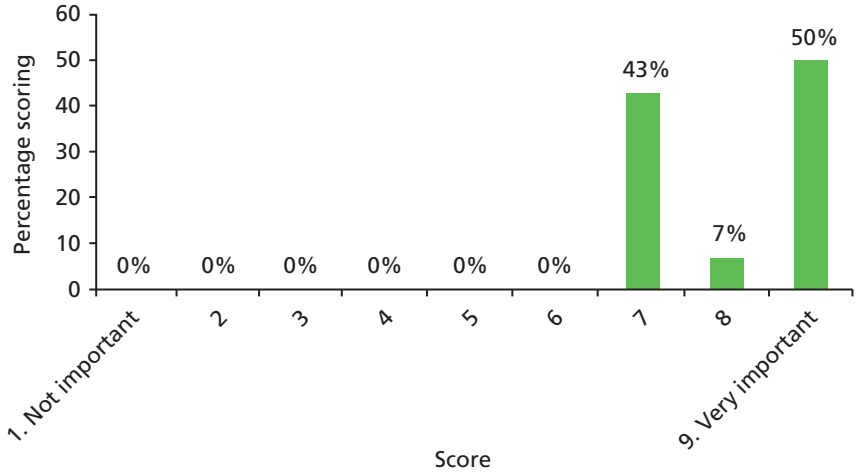


FIGURE 76 Results of scoring: receptive language skills – your child being able to listen and understand what other people say.

Outcome number	34
Outcome name	Speech development
Number of stakeholder groups achieving consensus	6
Number of participants scoring 1–9	14
Number of participants unable to score	0

Notes
JH commented that if this is based on how a child is developing compared with other children, then the age of the child is important, as concerns of parents might be greater at some ages than others. 'When' this outcome is measured might be important.
AH commented that in children with CP there are other factors, which might affect speech other than hearing. IAB and AHB commented that the way in which this outcome is measured, i.e. the 'how' is going to be important to determine whether changes in speech are due to changes in hearing or other aspects.

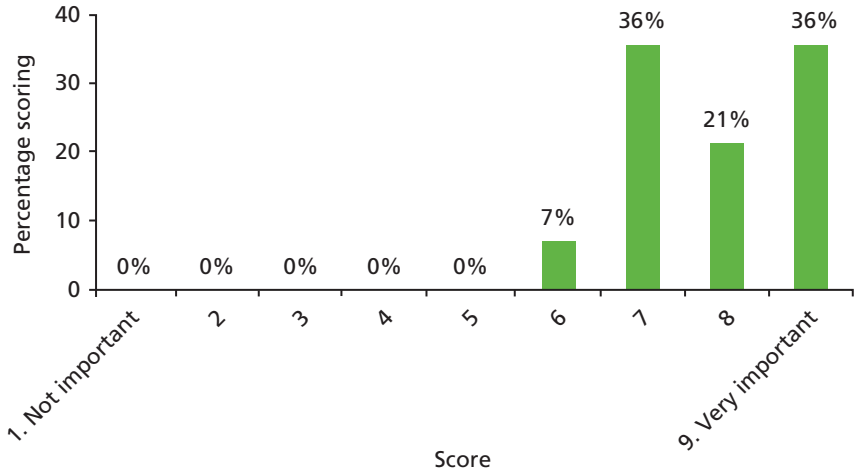


FIGURE 77 Results of scoring: speech development – your child speaking as well as other children who are the same age.

Outcome number	3
Outcome name	Atelectasis
Number of participants scoring 1–9	11
Number of participants unable to score	5

Notes
In the parent survey atelectasis, persistent tympanic membrane retraction and tympanosclerosis were scored as one outcome 'your child not having problems inside their ear caused by having lots of ear infections over a long time (more than 3 months)'.
TB asked if there could be one outcome about infections. AH and IAB agreed that it was not suitable to combine atelectasis in this way.
There was discussion around the symptoms of atelectasis which would generally not be noticeable to a patient. The grouping of this outcome with other outcomes related to the ear drum was discussed and all agreed that this would be more appropriate. IAB, AH and SD suggested combining as chronic otitis media. All agreed.

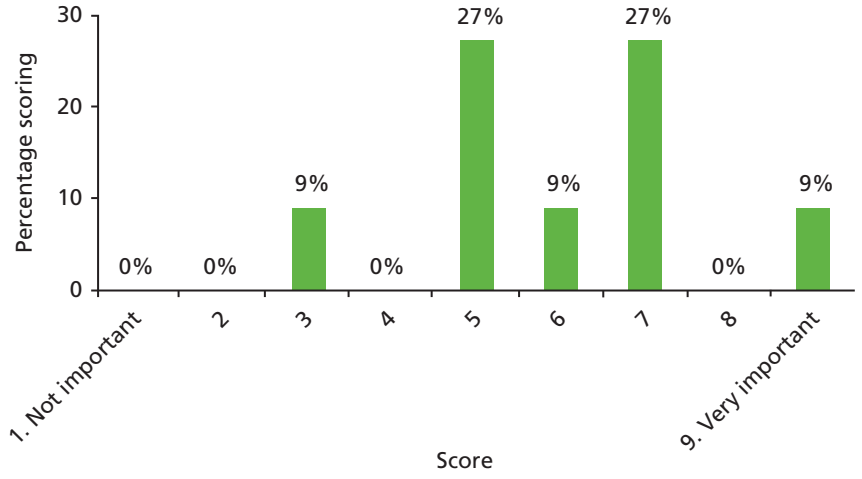


FIGURE 78 Results of scoring: atelectasis – your child not having problems inside their ear caused by having lots of ear infections over a long time (more than 3 months).

Outcome number	6
Outcome name	Cholesteatoma
Number of participants scoring 1–9	12
Number of participants unable to score	2

Notes
The consequences of cholesteatoma were discussed. SD mentioned that parents would not be routinely informed about this unless there was good evidence to suggest a cholesteatoma might occur as this would need surgery. JH and LH commented that parents do not have as much information to call on as clinicians. All agreed that this outcome should be revisited as there are possible wording and experience issues.

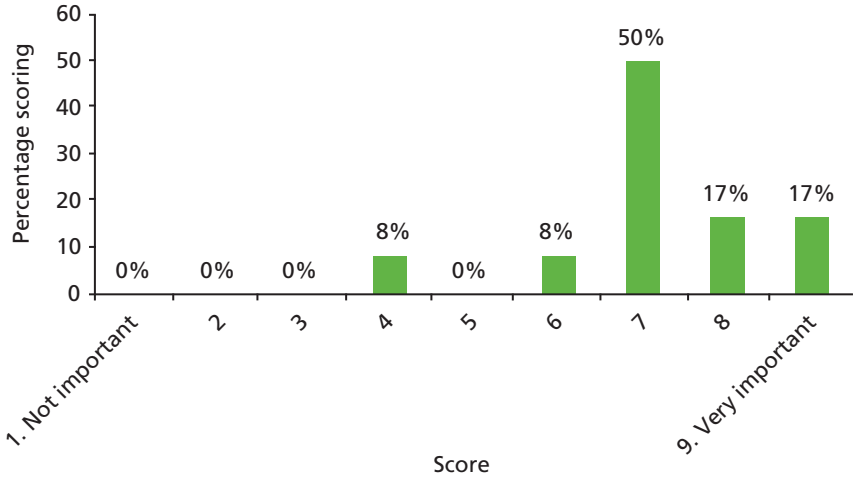


FIGURE 79 Results of scoring: cholesteatoma – your child not having problems inside their ear caused by bad skin growing behind their ear drum.

Outcome number	15
Outcome name	Psychosocial development
Number of participants scoring 1–9	14
Number of participants unable to score	0

Notes
AH commented that there are lots of factors which can influence psychosocial development. IAB commented that this would not be central in the way an ENT surgeon evaluates treatment. AB commented that the way that children interact is crucial and therefore it is incredibly important that this is included as a core outcome.

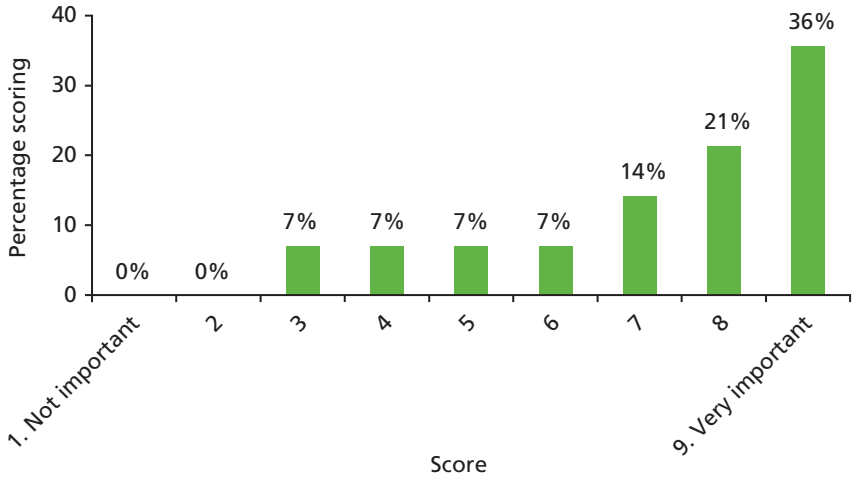


FIGURE 80 Results of scoring: psychosocial development – how well your child is learning to make friends and speak to new people.

Outcome number	25
Outcome name	AOM
Number of participants scoring 1–9	14
Number of participants unable to score	0

Notes
IAB commented that 'consensus in' might not have been voted for by ENT surgeons as you can have AOM as a consequence of glue ear and as a consequence of treatment for glue ear.
JH noted that this might not be the case if a new treatment were to be made available.

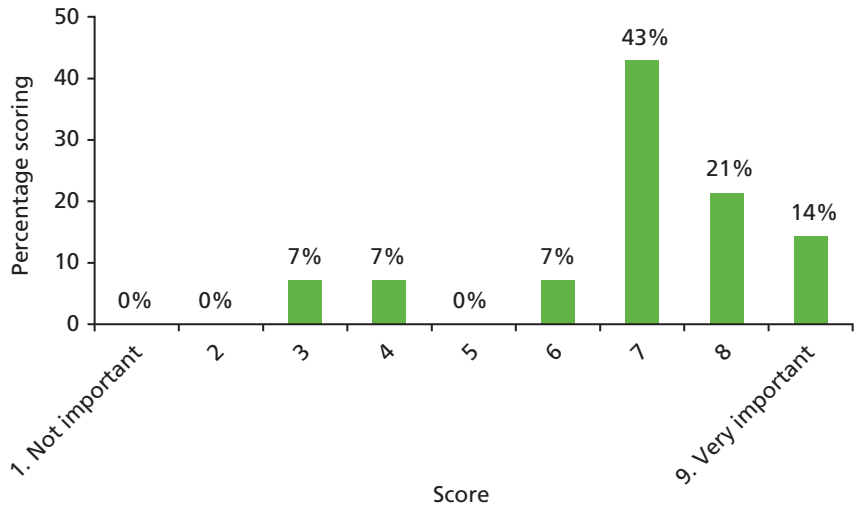


FIGURE 81 Results of scoring: AOM – your child not having ear infections.

Outcome number	28
Outcome name	Consonant production
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
'Speech intelligibility', 'consonant production' and 'expressive language skills' were scored by parents and children as one outcome: 'your child being able to say all of their words clearly so that adults and other children can understand what they said'.
Discussion as there were two outcomes related to consonant production: 'consonant production' and 'consonant production – cleft-related speech patterns'. Agreed that only one outcome 'consonant production' would be scored. Agreed that the outcomes scored as one 'your child being able to say all of their words clearly so that adults and other children can understand what they said' should be considered part of the 'how' of speech development and this will be discussed further with the SAG.

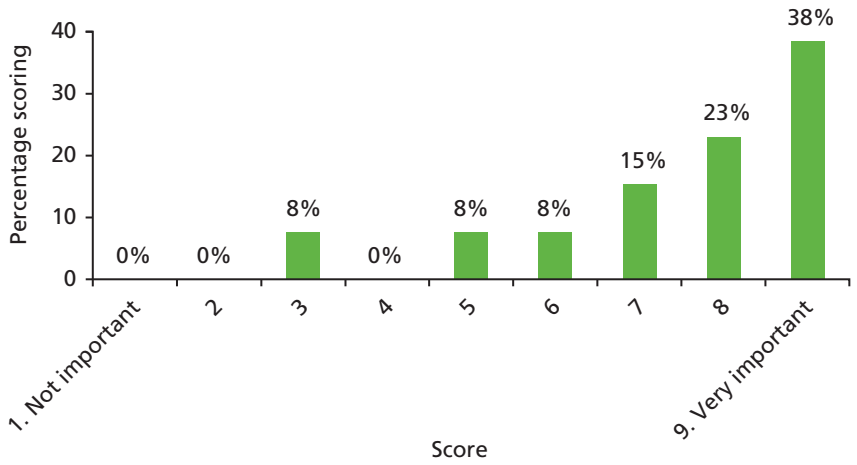


FIGURE 82 Results of scoring: consonant production – your child being able to say all of their words clearly so that adults and other children can understand what they say.

Outcome number	37
Outcome name	Necessity to remove VTs
Number of participants scoring 1–9	12*
Number of participants unable to score	1

Notes
Discussion that this is only relevant to studies involving VTs and not relevant to all types of intervention for the treatment of glue ear.

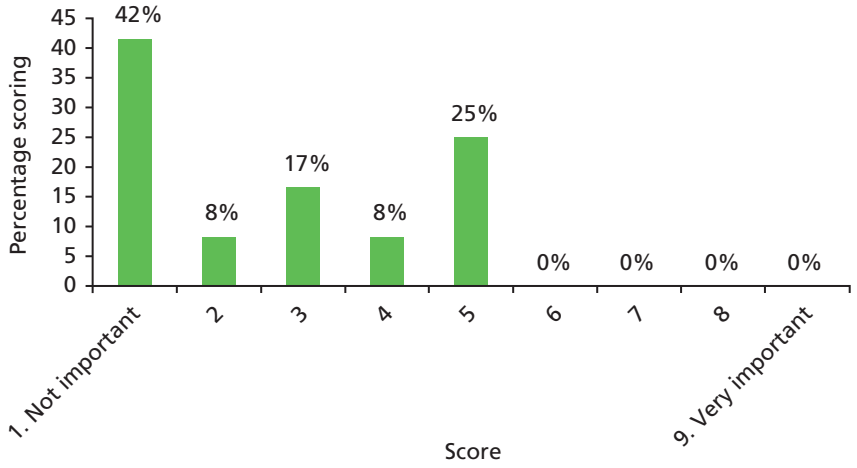


FIGURE 83 Results of scoring: necessity to remove VTs – your child not needing another operation to take grommets/VTs out.

Outcome number	38
Outcome name	Requirement for repeated VTs
Number of participants scoring 1–9	11*
Number of participants unable to score	2

Notes
Discussion that this is only relevant to studies involving VTs and not relevant to all types of intervention for the treatment of glue ear. All agreed not to include in COS.

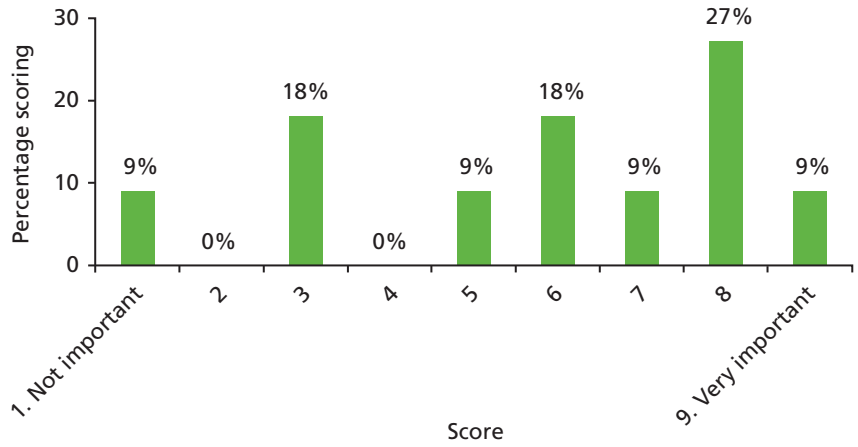


FIGURE 84 Results of scoring: requirement for repeated VTs – your child not needing another operation to take grommets/VTs because the old ones fell out.

Outcome number	41
Outcome name	Parental satisfaction with treatment
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
Voting only.

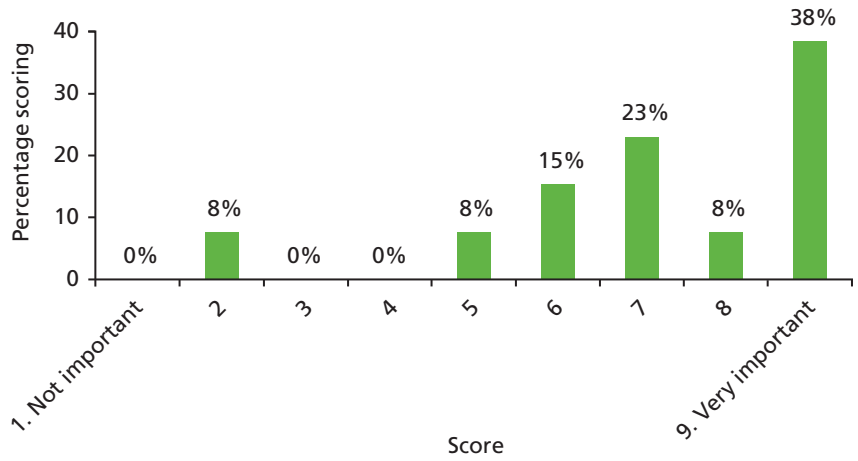


FIGURE 85 Results of scoring: parental satisfaction with treatment – how well you think that HAs or grommets have improved your child's hearing.

Outcome number	44
Outcome name	Child's satisfaction with treatment
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
Voting only.

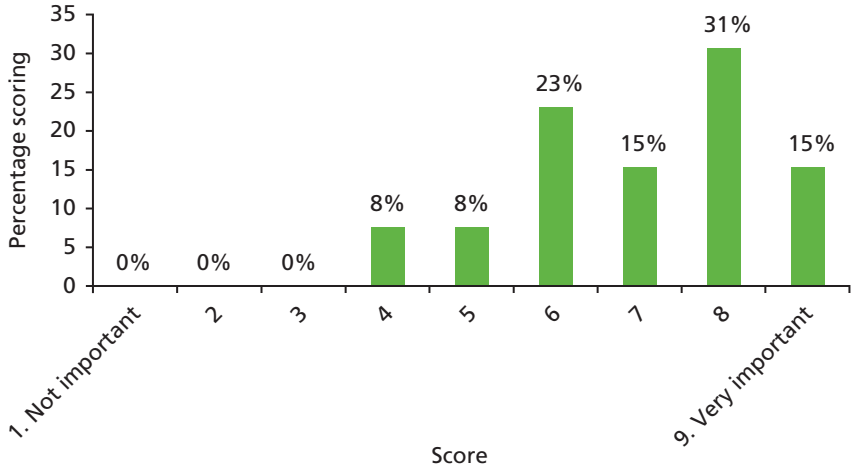


FIGURE 86 Results of scoring: child's satisfactions with treatment – how much your child thinks that treatment has made them better.

Outcome number	45
Outcome name	Child's perspective of speech
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
There was limited discussion for the outcome 'child's perspective of speech' and participants of the meeting moved quickly to voting on this outcome. For parent's perspective of speech there was discussion that this is linked to the outcome 'speech development' and represents 'how' this outcome is measured. For parental perspective of speech it was agreed that this should be included as a 'how'.

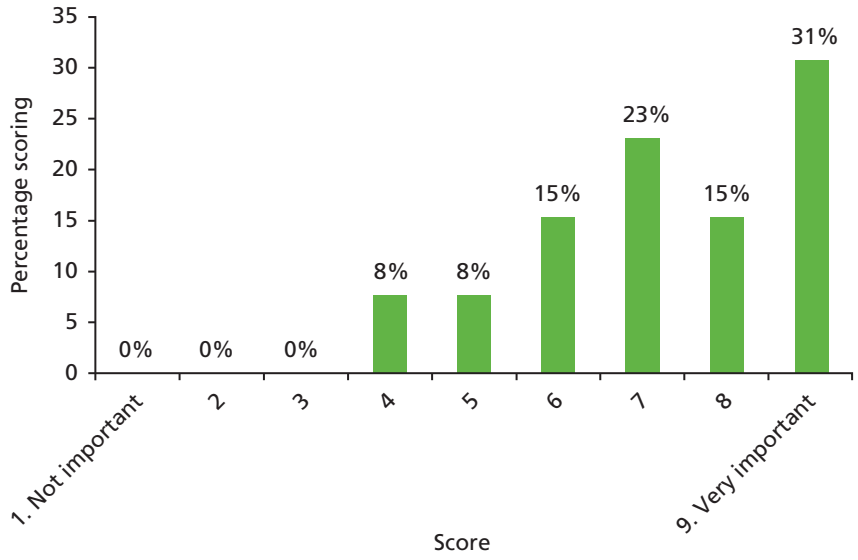


FIGURE 87 Results of scoring: child's perspective of speech – how normal your child thinks they sound when they are talking.

Outcome number	4
Outcome name	Persistent tympanic membrane retraction
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'chronic otitis media' and considered in 'how' the outcome is measured.

Outcome number	10
Outcome name	Cognitive development
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'how well your child is doing at school' and would be considered in 'how' this outcome is measured.

Outcome number	11
Outcome name	Developmental progress
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'how well your child is doing at school' and would be considered in 'how' this outcome is measured.

Outcome number	31
Outcome name	Consonant production – cleft-related speech patterns
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'consonant' and would be considered in 'how' this outcome is measured.

Outcome number	42
Outcome name	Side effects of treatment
Number of participants scoring 1–9 (unable to score)	7*
Number of participants unable to score	1

Notes
PW noted that in a trial comparing two treatments it is usual to assess any potential side effects.
All agreed that this was important but that it might be covered by other outcomes.

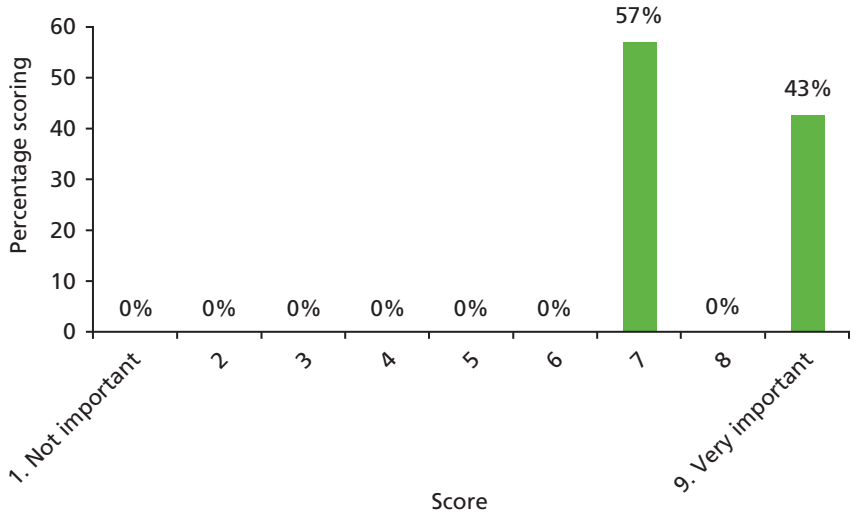


FIGURE 88 Results of scoring: side effects of treatment – your child not having problems, that sometimes happen, that are caused by a treatment they have for glue ear.

Outcome number	47
Outcome name	Listening skills
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
Minimal discussion, noted that this includes pre and post verbal skills.

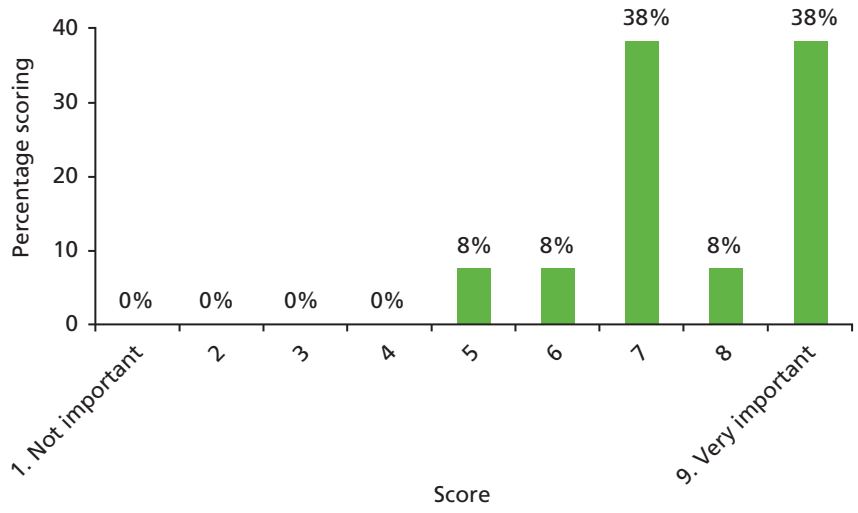


FIGURE 89 Results of scoring: listening skills (not scored by parents or children).

Outcome number	48
Outcome name	Psychosocial well-being
Number of participants scoring 1–9	13*
Number of participants unable to score	0

Notes
Not discussed voting only.

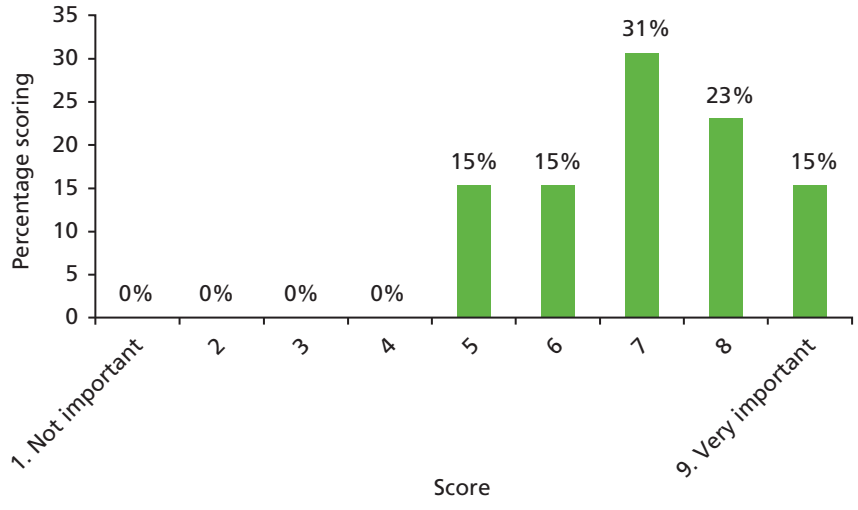


FIGURE 90 Results of scoring: psychosocial well-being (not scored by parents or children).

Outcome number	5
Outcome name	Tympanosclerosis
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'chronic otitis media' and considered in 'how' the outcome is measured.

Outcome number	8
Outcome name	Persistent tympanic membrane perforation
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'chronic otitis media' and considered in 'how' the outcome is measured.

Outcome number	17
Outcome name	Otalgia
Number of participants scoring 1–9	12*
Number of participants unable to score	0

Notes
Needs further discussion with parents. ENT may not have considered this important as children are often not referred because of pain. However, children with glue ear get recurrent earache. The outcome was scored but more input from parents needed.

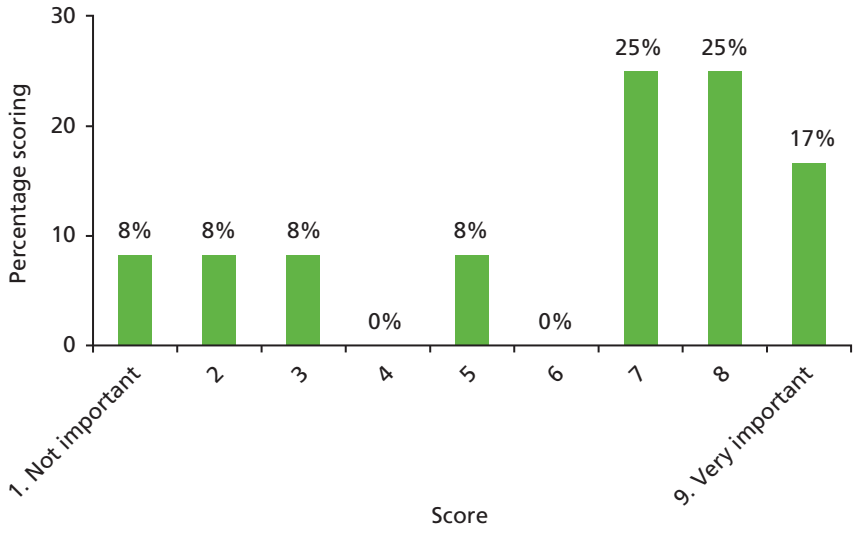


FIGURE 91 Results of scoring: otalgia – how painful your child's ear is.

Outcome number	18
Outcome name	Otorrhoea
Number of participants scoring 1–9	12*
Number of participants unable to score	0

Notes
No discussion, voting only.

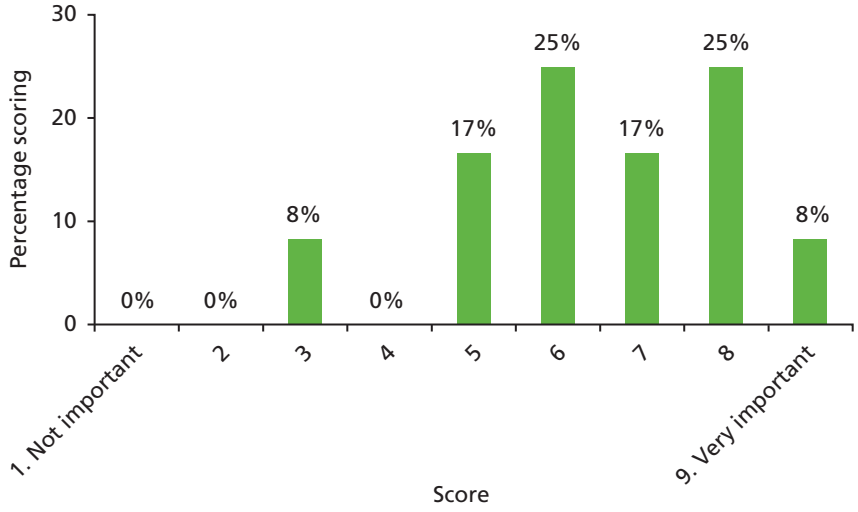


FIGURE 92 Results of scoring: otorrhoea – your child not having infected liquid (pus) leaking out of their ear.

Outcome number	21
Outcome name	Eustachian tube function
Number of participants scoring 1–9	11*
Number of participants unable to score	1

Notes
Wording might have influenced parental response. To discuss further with parents.

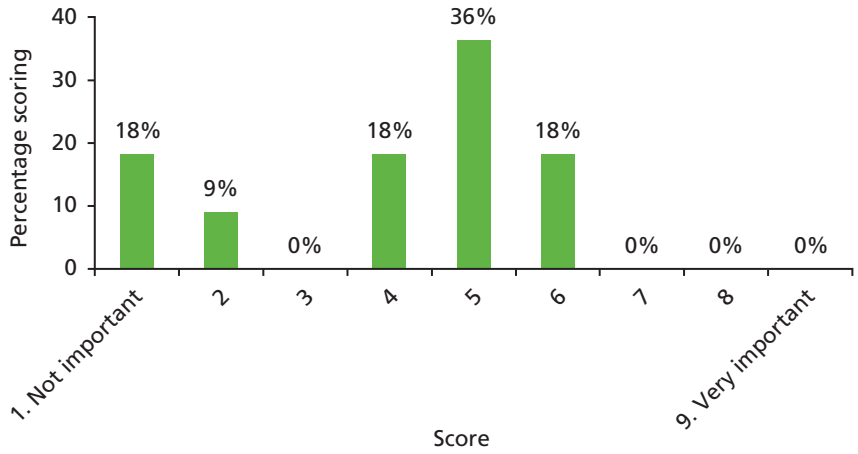


FIGURE 93 Results of scoring: eustachian tube function – how well a special tube in your child's ear works, if this doesn't work properly your child might hear popping and crackling noises.

Outcome number	29
Outcome name	Expressive language skills
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
Agreed that the outcomes scored as one 'your child being able to say all of their words clearly so that adults and other children can understand what they said' should be considered part of the 'how' of speech development and this will be discussed further with the SAG.
Agreed not to discuss further, no voting.

Outcome number	35
Outcome name	Speech signs of velopharyngeal insufficiency
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
Discussion around wording of outcome as this might need a different lay description. Agreed that this needs further discussion with parents, this outcome might also represent the 'how' of speech development.
No voting.

Outcome number	36
Outcome name	Early extrusion or blockage of VTs
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
Discussion that this is only relevant to studies involving VTs and not relevant to all types of intervention for the treatment of glue ear. All agreed not to include in COS.

Outcome number	39
Outcome name	Child stress
Number of participants scoring 1–9	8*
Number of participants unable to score	0

Notes
JH thought that this was important. RP suggested that this is not an issue for some and therefore some parents/children would not have scored highly as experience is likely to impact on perceived importance.

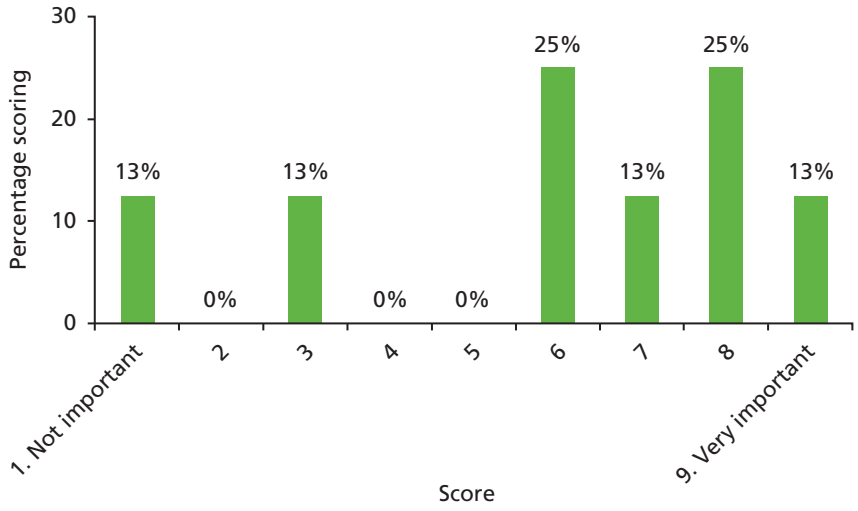


FIGURE 94 Results of scoring: child stress – how often your child feels tense or upset.

Outcome number	48
Outcome name	Psychological well-being
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
Not discussed, need to revisit wording and provide explanation so that the difference between psychosocial and psychological well-being is clear.
No voting.

Outcome number	1
Outcome name	Internalising behaviour
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
Not discussed, all agreed not in COS.
No voting.

Outcome number	2
Outcome name	Externalising behaviour
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
Not discussed, all agreed not in COS.
No voting.

Outcome numbers	9–14
Outcome name	Academic achievement
Number of participants scoring 1–9	12*
Number of participants unable to score	0

Notes
Parents scored 'How well your child is doing at school or college', this combined the outcomes 'academic achievement', 'cognitive development', 'developmental progress', 'intelligence', 'literacy' and 'phonological memory'.
All agreed that one outcome should be considered and the details determined as part of the 'how'. Therefore all considered the outcome 'how well your child is doing at school or college' when scoring.

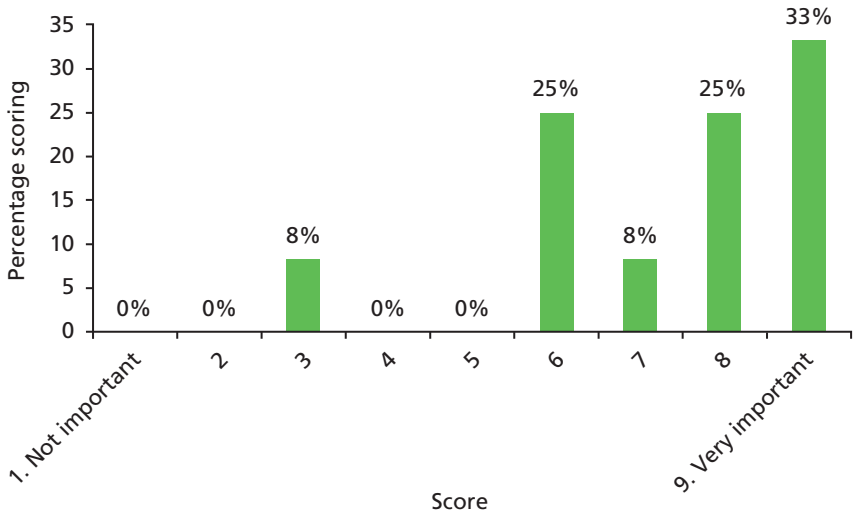


FIGURE 95 Results of scoring: academic achievement – how well your child is doing at school or college.

Outcome number	13
Outcome name	Literacy
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'how well your child is doing at school' and would be considered in 'how' this outcome is measured.

Outcome number	14
Outcome name	Phonological memory
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting, was agreed that this should be included with 'how well your child is doing at school' and would be considered in 'how' this outcome is measured.

Outcome number	19
Outcome name	Tinnitus
Number of participants scoring 1–9	12*
Number of participants unable to score	0

Notes
IAB gave an explanation that tinnitus affected the inner ear rather than the middle ear (like glue ear). AHB raised that parents discuss tinnitus in speech clinics but do not realise they can go to ENT about it. JH confirmed that would not go to ENT with tinnitus.

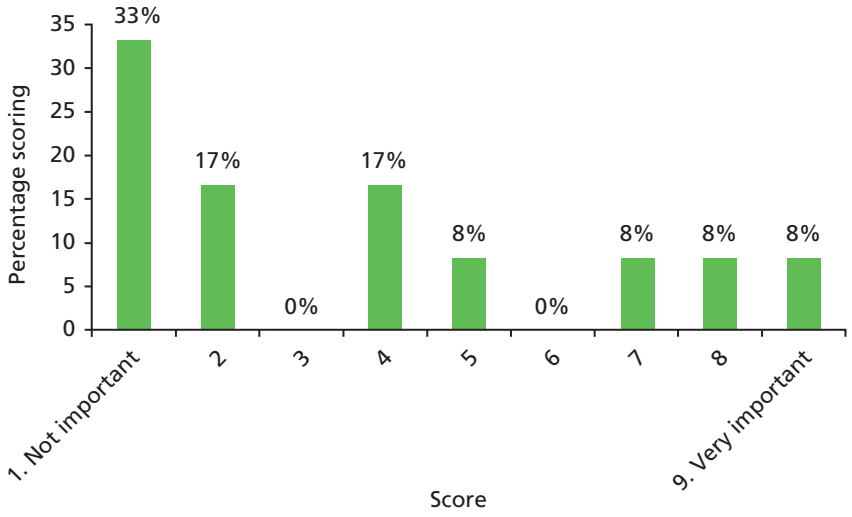


FIGURE 96 Results of scoring: tinnitus – how much your child hears ringing or buzzing noises.

Outcome number	20
Outcome name	Vertigo
Number of participants scoring 1–9	12*
Number of participants unable to score	0

Notes
Minimum discussion – not important to parents attending.

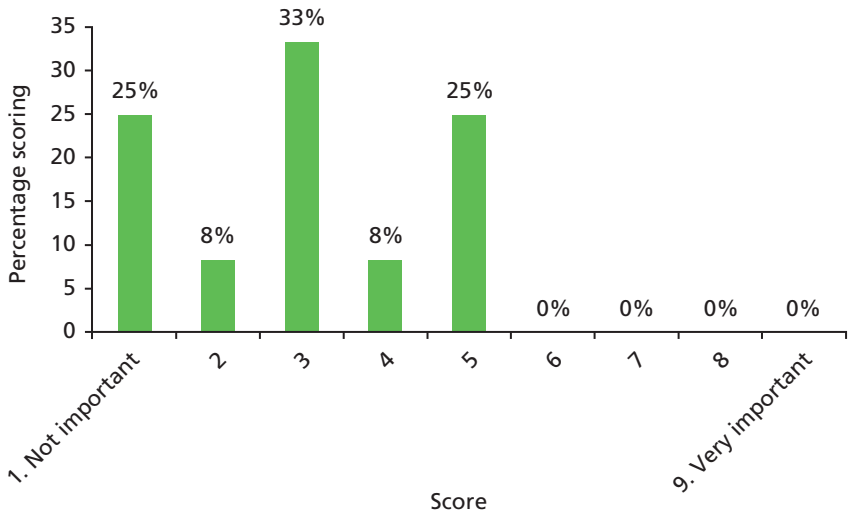


FIGURE 97 Results of scoring: vertigo – how dizzy your child feels.

Outcome number	22
Outcome name	Stapedial reflex
Number of participants scoring 1–9	10*
Number of participants unable to score	1

Notes
None.

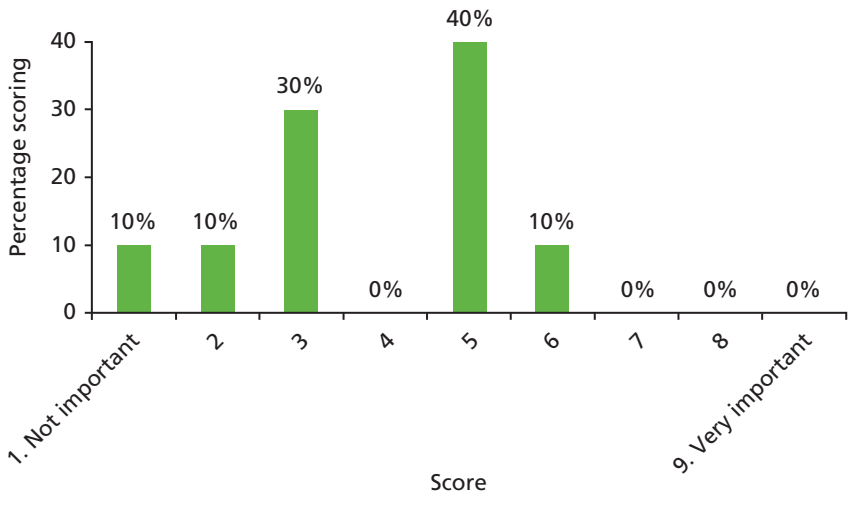


FIGURE 98 Results of scoring: stepedial reflex – how well your child's ear works when it hears a loud noise.

Outcome number	32
Outcome name	Parent's perspective of speech
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
No voting. All agreed that this part of the 'how' for the outcome 'speech development'.

Outcome number	40
Outcome name	Parental stress
Number of participants scoring 1–9	7*
Number of participants unable to score	0

Notes
LH noted that only specialist cleft nurses and clinical psychologists were likely to be made aware of child stress. Both JH and LH agreed that child stress was more important to them than parental stress.

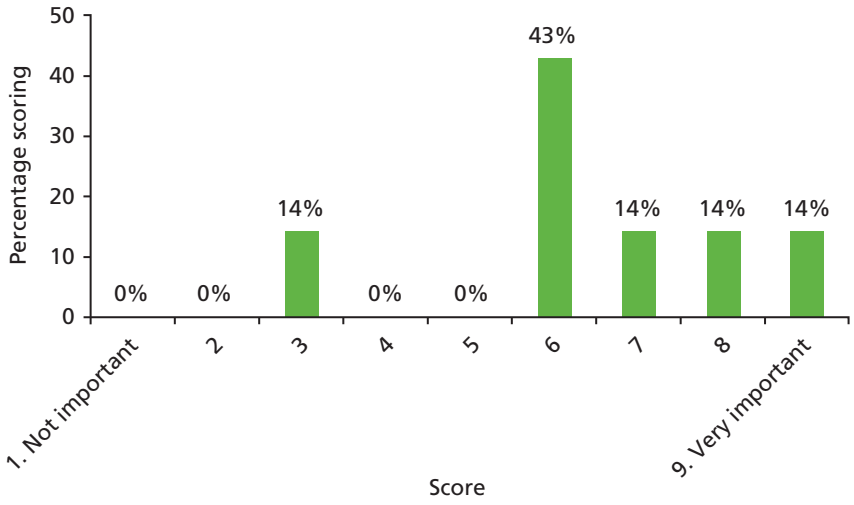


FIGURE 99 Results of scoring: parental stress – how often you feel tense or upset.

Outcome number	27
Outcome name	Temporary tympanic membrane perforation
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
All agreed that this should not be included in the COS – no further discussion or voting.

Outcome number	43
Outcome name	Upper respiratory tract infection
Number of participants scoring 1–9	7*
Number of participants unable to score	0

Notes
Noted that there are potentially some issues with the wording provided to parents. Health professionals would consider this as colds and viruses, which could happen regardless of treatment. For the parents wording the word 'ear' should be removed. Needs further discussion with parents.

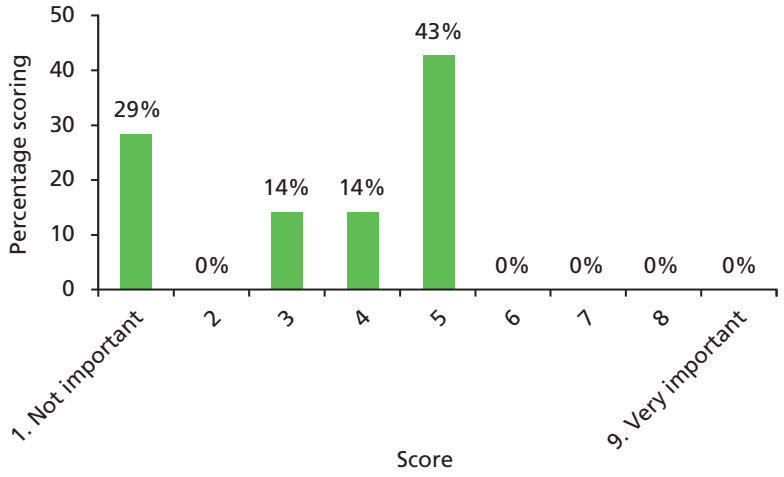


FIGURE 100 Results of scoring: upper respiratory tract infection – your child not having infections in their ear, nose or throat.

Outcome number	24
Outcome name	Rhinitis
Number of participants scoring 1–9	–
Number of participants unable to score	–
Results of discussion	

Notes
All agreed that this should not be included in the COS – no further discussion or voting.

Outcome number	49
Outcome name	Hyperacusis
Number of participants scoring 1–9	–
Number of participants unable to score	–

Notes
All agreed that this needs further discussion with parents.

Appendix 10 Electronic search strategy to identify published model-based economic evaluations

MEDLINE, EMBASE and the American Economic Association's electronic bibliography

URLs: MEDLINE, www.nlm.nih.gov/bsd/pmresources.html; EMBASE, www.elsevier.com/online-tools/embase; EconLit, www.aeaweb.org/econlit/; accessed via Ovid – https://ovidsp.ovid.com/

Date range searched: MEDLINE, from inception to 16 January 2014; EMBASE, from inception to 16 January 2014; EconLit, from inception to 16 January 2014.

Date search performed: January 2014.

Search strategy

1. otitis media.mp.
2. (glue ear or glue-ear).mp.
3. (middle ear effusion or effusion of the middle ear or middle ear infection or infection of the middle ear or middle ear inflammation or inflammation of the middle ear).mp.
4. or/1-3
5. (cleft$ or cheiloschisis or palatoschisis).mp.
6. 4 and 5
7. (economic$ analys$ or economic$ evaluation$).mp.
8. (cost-effective$ or cost effective$).mp.
9. (cost-utility or cost utility).mp.
10. (cost-benefit or cost benefit).mp.
11. (cost-minimi$ or cost minimi$).mp.
12. (cost-consequence$ or cost consequence$).mp.
13. (value-of-information analys$ or value of information analys$).mp.
14. (decision-tree model$ or decision tree model$).mp.
15. (markov model$ or state-transition model$ or state transition model$).mp.
16. simulation model$.mp.
17. (individual-patient simulation or individual patient simulation).mp.
18. (individual patient-level model$ or individual patient level model$).mp.
19. (health-economic$ model$ or health economic$ model$).mp.
20. (decision-analytic$ model$ or decision analytic$ model$).mp.
21. (quality-adjusted life-year$ or quality-adjusted life year$ or QALY$).mp.
22. (disability-adjusted life-year$ or disability-adjusted life year$ or DALY$).mp.
23. or/7-22
24. 6 and 23

Appendix 11 Electronic search strategy to identify estimates of impact on health-related quality of life and utility values

MEDLINE, EMBASE and the American Economic Association's electronic bibliography

URLs: MEDLINE, www.nlm.nih.gov/bsd/pmresources.html; EMBASE, www.elsevier.com/online-tools/embase; EconLit, www.aeaweb.org/econlit/; accessed via Ovid – https://ovidsp.ovid.com/

Date range searched: MEDLINE, from inception to 16 January 2014; EMBASE, from inception to 16 January 2014; EconLit, from inception to 16 January 2014.

Date search performed: January 2014.

Search strategy

1. hearing.mp.
2. otitis media.mp.
3. (glue ear or glue-ear).mp.
4. (middle ear effusion or effusion of the middle ear or middle ear infection or infection of the middle ear or middle ear inflammation or inflammation of the middle ear).mp.
5. or/1-4
6. (utility index or utility-index).mp.
7. (utility score$ or utility-score$).mp.
8. (utility preference$ or utility-preference$).mp.
9. (utility value$ or utility-value$).mp.
10. (utility weight$ or utility-weight$).mp.
11. (health state utility or health-state utility or health-state-utility).mp.
12. (health state utilities or health-state utilities or health-state-utilities).mp.
13. (health utility index or health-utility index or health-utility-index).mp.
14. (health utility or health-utility).mp.
15. (utility measure$ or utility-measure$).mp.
16. (health related utility or health-related utility or health-related-utility).mp.
17. (health related utilities or health-related utilities or health-related-utilities).mp.
18. (time-trade-off or time trade-off or TTO).mp.
19. (standard gamble or standard-gamble or SG).mp.
20. (EQ-5D or EQ5D or EQ 5D or EuroQoL-5D or EuroQoL 5D or EuroQoL-five dimensions or EuroQoL five dimensions).mp.
21. (SF-6D or SF6D or SF 6D or short form-6D or short form 6D or short-form six-dimensions or short form six dimensions or short form-six dimensions).mp.
22. (quality of well being scale or quality of well-being scale or QWB).mp.
23. (quality of well being scale self administered or quality of well-being scale self-administered or QWB-SA).mp.
24. (health utilities index or HUI2 or HUI-2 or HUI-II or HUI3 or HUI-3 or HUI-III).mp.
25. (cost-utility or cost utility).mp.
26. (quality-adjusted life-year$ or quality adjusted life year$ or QALY$).mp.
27. or/6-26
28. 5 and 27

29. grommet$.mp.
30. (ventilation tube$ or ventilation-tube$ or tympanostomy tube$ or tympanostomy-tube$).mp.
31. (hearing aid$ or hearing-aid$).mp.
32. (watchful waiting or watchful-waiting or do nothing or do-nothing).mp.
33. (adenoid$ or adenoidectomy).mp.
34. or/29-33
35. 28 and 34
36. 35 and (child or children).mp.
37. remove duplicates from 36

EME
HS&DR
HTA
PGfAR
PHR

Part of the NIHR Journals Library
www.journalslibrary.nihr.ac.uk

Published by the NIHR Journals Library