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Using an epidemiological model to investigate unwarranted variation: the case of ventilation tubes for otitis media with effusion in England

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Abstract

Objectives: To investigate unwarranted variation in ventilation tube (VT) insertions for otitis media with effusion (OME) in children in England. This procedure is known to be ‘overused’ from audits of care provided, as only one in three VT insertions conform to the appropriateness criteria by the National Institute for Health and Clinical Excellence (NICE); but audits cannot identify the scale of ‘underuse’: i.e. patients who would benefit but are not treated.

Methods: To explore both ‘underuse’ and ‘overuse’ of VTs for OME we developed an epidemiological model based on: definitions of children with OME expected to benefit from VTs according to NICE guidance; epidemiological and clinical information from a systematic review; and expert judgement. A range of estimates was derived using Monte Carlo simulation and compared with the number of VTs actually provided in the NHS in 2010.

Results: About 32,200 children in England would be expected to benefit from VTs for OME per year (between 20,411 and 45,231 with 90% certainty). The observed number of VTs for OME-associated diagnoses however was 16,824.

Conclusions: The expected population capacity to benefit from VTs for OME based on NICE guidance appeared to exceed, by far, the number of VTs actually provided in the NHS. So, while there is known ‘overuse’, there also may be substantial ‘underuse’ of VTs for OME if NICE criteria were applied. Future investigations of unwarranted variation should therefore not only focus on patients who are treated, but consider potential to benefit at the population level.

Introduction

Systems of healthcare in countries that are under severe fiscal pressures¹ seek to do more for less: to increase the benefits from healthcare and reduce its costs. There is evidence of large and persistent variations in medical practice across small areas, which have been documented in various countries.² This evidence is generally seen as an indication of ‘overuse’: i.e. where reductions in rates of treatment could release resources with gains in health.³ In England, commissioners are allocated budgets for their populations and have to develop policies for services for which they are and are not prepared to pay. One such policy seeks to reduce unwarranted variation by restricting access to procedures listed as being of ‘low clinical value’.⁴ However, due to the lack of an objective reference point against which to evaluate ‘overuse’ (ineffective care that is more likely to harm than help the patient⁵) or ‘underuse’ (the failure to provide services from which the patient would likely benefit⁵), information on variations remains essentially ambiguous.⁶ The purpose of this article is to investigate unwarranted variations by modelling the scale of ‘underuse’ or ‘overuse’ of ventilation tubes (VTs; grommets) for children with otitis media with effusion (OME) in England.

VT insertions are a classic case of high geographic variation. Variations in England have been documented since the 1980s⁷ and have persisted: in 2010/11 there was about eight-fold variation across 151 commissioners (with a mean population of about 300,000).⁸ VTs have been listed by commissioners as a ‘low value’ procedure,⁴ which seeks to restrict referrals by general practitioners (GPs). Despite that, VT insertions remain one of the most frequent surgical interventions in children: with over 32,000 insertions in 2010/11, of which 23,500 were among children younger than 14 years.⁹ Clinical audits in the US¹⁰ and the UK,¹¹ using different criteria of appropriateness, found that only one in three VT

insertions were appropriate, suggesting substantial ‘overuse’. However, audits of care delivered cannot address the scale of ‘underuse’ of VTs for OME. We therefore developed an epidemiological model to estimate the number of children with capacity to benefit from VTs for OME, if NICE guidance¹² were being followed, and compared this with the number of VTs actually provided in England.

Recommended clinical pathway

OME is defined as an effusion in the middle ear cleft, in the absence of signs of acute inflammation. It may cause conductive hearing loss, which, if persistent, can affect speech and language development, educational performance and behaviour.¹³ By the age of four years, about 80% of children have had episodes of OME.¹⁴ As OME is transitory for most children, the NICE clinical pathway (Figure 1) recommends an initial period of active observation over three months and repeat audiological testing at the end of this period. At that stage, it is recommended that VTs are offered for children younger than 12 years who meet three ‘core’ criteria: (1) bilateral OME with (2) a hearing level in the better hearing ear of 25 to 30 dBHL or higher that (3) is documented over a period of three months. The crucial point is that NICE guidance does not define VTs as an intrinsically ‘low value’ procedure, but recognises their value in relation to a set of evidence-based criteria. In exceptional cases, VTs may also be offered if clinicians judge the impact of OME-related hearing impairment on the child’s development, well-being or social functioning to be substantial.¹²

Methods

Based on the NICE criteria, our epidemiological model to estimate population capacity to benefit from VTs for OME is formulated below. The modelling assumptions are

summarised in Table 1. The parameters, their definition and estimation are given in Table 2.

1) *Incidence*: The number of new cases of OME in any given year, $N(OME)$, is determined by the annual age-specific cumulative incidence (risk) I_j of OME multiplied by the susceptible population in a given age group S_j , summed over all eligible age groups j . The subgroup of cases with bilateral OME and a hearing level at NICE threshold level is expressed by

$$N(OME) = \sum_{j=0}^{12} (S_j * I_j * P(HL|Bilateral\ OME) * P(Bilateral\ OME|OME))$$

2) *Disease process*: We model the probability of OME persisting at time t from the onset of OME as an exponential process (adapted from¹⁵) of the form

$$P(OME | t) = \frac{1}{2^{\frac{t}{m}}}$$

3) *Capacity to benefit from VTs for OME*: As OME is transitory, the population with capacity to benefit will diminish as time passes since the onset of OME. Population capacity to benefit from VTs for OME is estimated as

$$PCB(t) = P(OME | t) * N(OME)$$

> Figure 1: NICE clinical pathway <

> Table 1: Modelling assumptions <

> Table 2: Model parameters <

Data sources and extraction

To estimate parameter values, we carried out a systematic literature review according to PRISMA guidelines¹⁶ (see Appendix A for details of the search strategy and data extraction, Appendix B for the rationale for the study inclusion criteria).

Setting and population

The setting is the National Health Service (NHS) in England. The population includes children younger than 12 years covered by NICE guidance. However, as we were unable to find incidence studies that met our inclusion criteria for the age groups 0, 1, 4 and 9 to 12 years, we focused the analysis on children aged 2 to 8 years (extrapolating the incidence for 4-year olds from 3-year olds) which is the age group with the majority of VT insertions (0 to 12 years: 19,805; 2 to 8 years: 16,824 procedures with OME-associated diagnoses in 2010/11⁹). To estimate the susceptible population, the total population of children has been corrected for an estimate of OME prevalence (Appendix C). We focused on children meeting the three NICE ‘core’ criteria for VT insertion. The number of exceptional cases, which are identified through clinical judgement, was not modelled. This means that estimates from our epidemiological model are probably conservative and underestimate the number of children with capacity to benefit from VTs.

Model validation

All modelling assumptions were iteratively refined in consultation with the Project Steering Group. During an expert workshop in September 2012, ten participants with complementary expertise in audiology, ENT, general practice and epidemiology were invited to conduct a structured ‘walkthrough’ to examine the model’s overall structure

and individual components. The group judged the model to be a fair representation of the NICE care pathway and of the disease process governing OME given the existing evidence base.

Sensitivity analysis

Data retrieved from the literature raised the issue of potential for bias in terms of internal validity (the extent to which the design of original studies ensured accurate measurement of the parameters of interest) and external validity (the extent to which studies conducted e.g. two decades ago in a different setting were applicable to the present UK context). While we recognise the relevance of the literature-based data, we felt the different sources of uncertainty in the evidence would merit supplementing this with expert judgement. We followed a structured approach to expert elicitation.¹⁷ We provided the panel of experts with the literature-based estimates, encouraged discussion and elicited fractiles of subjective probability distributions. We then used these estimates in a Monte Carlo simulation performed in @RISK 5.0 to gain an insight into the impact of the combined uncertainty in parameter estimates on the modelling results.¹⁸

Results

Figure 2 illustrates the combined uncertainty in the expected incidence of bilateral OME with a hearing level of +25 dB. Based on 10,000 iterations of the model and given the set of input distributions, the resulting distribution of the expected incidence ranges between 63,800 and 143,600 cases per year in England with 90% certainty (mean estimate: 102,083 cases). These results from the Monte Carlo simulation are used to model the expected number of children with capacity to benefit from VTs for OME as the total waiting time from the onset of OME is varied over a range.

Since OME is transitory, the expected population capacity to benefit from VTs for OME depends on the total waiting time from the onset of OME to the point where treatment is considered (Figure 3). NICE guidance recommends a three-month period of active observation following the first formal diagnosis. Thus, if we were to assume the first outpatient appointment took place instantaneously after the onset of OME, then the mean estimate of children for whom VTs would be clinically indicated would be approximately 51,000 (at $t=3$ months; between 32,400 and 71,800 with 90% certainty). There is currently no national guidance on the recommended waiting time from the onset of OME until the first outpatient appointment (waiting time intervals t_1 and t_2 in Figure 1). Since our model aims to provide a benchmark of expected care, rather than a reflection of actual practice, our assumptions about the length of these intervals (Table 2) represent clinically ‘ideal’ circumstances based on expert group consensus. Assuming a one-month buffer period before parents become concerned about the symptoms of OME and visit a GP and another month before children have their first outpatient appointment, we would expect approximately 32,200 children to benefit from VTs for OME (at $t=5$ months; 90% certainty interval 20,411 to 45,231). This contrasts with an ‘observed’ number of 16,824 VTs that were actually provided for OME-associated diagnosis codes in the age group of 2 to 8 years in 2010/11 in England. As can be seen in Table 3, even if we were to assume coding inaccuracies in VTs coded with OME-associated diagnoses, the conclusions would be unaffected.

> Figure 2: Monte Carlo simulation <

> Figure 3: Capacity to benefit from VTs for OME <

> Table 3: Observed number of VT insertions in England, 2010/11 <

Discussion

This study shows that the expected capacity to benefit from VTs for OME among children in England, according to NICE guidance, exceeds the number of VTs that were actually provided in the NHS. Our model hence reveals the possibility of ‘underuse’ of VTs for OME at the aggregate national level. However, the findings also need to be interpreted in the light of the roughly eight-fold variation in treatment rates across PCTs in England,⁸ which suggests that ‘overuse’ might still occur in some regions.

Strengths and weaknesses of the study

The model draws on evidence-based clinical guidance to obtain an indicative estimate of the scale of potential ‘underuse’ or ‘overuse’ of VTs in a given population. This estimate does not represent the ‘right (treatment) rate’, which would also depend on informed patient choice. It attempts to approximate a level of treatment that the NHS would be expected to offer to patients, if NICE criteria were accepted as a valid basis for identifying patients with capacity to benefit from VTs. We recognise that NICE criteria can only be approximate predictors of ‘benefit’ from VTs for hearing outcomes, especially for cases located just above or below the +25dBHL threshold, with even more uncertainty over the impact of VTs on childhood development and the child’s Quality of Life. Thus, from a normative standpoint, our model can only give an approximate estimate of how many VTs ‘should’ be offered, which may change once better predictors of benefit become available.

The model uses best available evidence identified through a systematic review. The shortage of high-quality studies meeting our inclusion criteria did not allow for a meta-analysis, and we have demonstrated the consequent uncertainty in our parameter estimates and their combined impact on the modelling results by Monte Carlo simulation. The

observed number of VTs provided covers patients treated in the NHS; unfortunately we were unable to obtain estimates of the scale of private practice in England. However, total private sector expenditure on healthcare in the UK (2011) is 17.2%,¹⁹ which would not substantially affect the conclusions of our study.

Findings in relation to studies of utilisation

Our study using a population model complements utilisation-based studies of treatment appropriateness. A recent multi-centre study in England found that only 32.2% of VTs inserted complied with the three ‘core’ NICE criteria, while 54.8% of VTs were provided on the basis of ‘exceptional circumstances’.¹¹ Although NICE guidelines explicitly encourage the provision of VTs also beyond the three ‘core’ criteria if clinicians judge the impact of OME on the child’s development and social functioning to be substantial,¹² the apparent reframing of ‘exceptions’ under clinical guidance as the ‘rule’ in clinical practice does raise questions over treatment appropriateness. This study adds to these findings by illustrating that, while there may be deviation from NICE ‘core’ criteria, which could either reflect patient-oriented treatment or ‘overuse’ of VTs, ‘unmet clinical need’ according to these ‘core’ criteria may be present simultaneously.

Policy implications

An increasingly common policy among healthcare commissioners in England⁴ is to label VTs *per se* as ‘overused’ and ‘low value’ and hence restrict access to the procedure. Our findings highlight the possibility of substantial ‘underuse’ among children in England for whom VTs are deemed beneficial and thus call for a more nuanced policy response. Because there is no evidence of a systematic relationship between high rates of utilisation

and high rates of inappropriateness,²⁰ we need a policy that tackles overuse by clinical audit of treatment, and ensures access to effective care for children suffering from persistent bilateral OME with a degree of hearing loss that is disabling and may affect their health and development. This policy would use the ideas of epidemiologic surveillance of medical care²¹ to enlarge the framing of clinical appropriateness from audits of services delivered to population capacity to benefit. Understanding the number of people who might be expected to benefit, given local population characteristics and clinical guidance, has relevance also for other high-volume services such as cataract surgery, joint arthroplasty or spinal procedures: it could help widen clinical concerns from individual patients towards the entire population who could (not) benefit and should hence (not) be offered a procedure. This policy would require investments in: (1) recommended intervention criteria that are more directly related to patient benefit, based on evidence from everyday practice (high-quality clinical databases rather than RCTs) on the real-world impacts of surgery on health outcomes compared to a control group; and (2) good information on disease epidemiology.

Implications for research and quality improvement

To explain the discrepancy between ‘observed’ VT provisions and the ‘expected’ number of VTs offered, a multi-faceted qualitative and quantitative approach involving commissioners, professionals and families is needed to identify barriers along the whole pathway and then design interventions for improvement. As parents, teachers and nurseries may fail to recognise hearing loss associated with OME,²² it is possible that many patients do not present to primary care in the first place. GPs, school nurses and health visitors need the knowledge and capacity to identify patients with suspected OME and ensure timely referral and diagnosis according to NICE criteria; in a recent UK-based

study, participating GPs correctly identified OME only in 53% of cases, which is not much higher than chance.²³ Since VTs feature widely as a ‘low value’ procedure,⁴ GPs might also tend to withhold referrals even for patients for whom VTs could be a clinically and cost-effective option. Delays in care and a long history of ‘watchful waiting’ in community services may thus, in practice, exceed the two-month interval from the onset of OME to formal diagnosis which we assumed as a clinically ‘ideal’ benchmark in our model. To overcome fragmentation, GPs, audiologists and ENT specialists need to work together to ensure early recognition and referral of children with capacity to benefit from treatment. Patients and carers deliberately choosing non-surgical treatment alternatives, such as hearing aids or medical management, may also in part explain the apparent discrepancy between ‘expected’ and ‘observed’. However, many patients and carers may not be given the opportunity to discuss and understand their options for treatment, resulting in uninformed use of other care. Future research might therefore also examine regional variations in patient preferences and approaches to shared decision-making²⁴ and how these add to, or interact with, differences in local commissioning criteria and socio-economic inequalities.

Conclusions

This study has highlighted the case of VTs for OME which, although known to be ‘overused’ based on audits of care provided, simultaneously seem to exhibit substantial ‘underuse’ at a population level in England based on NICE guidance. Because ‘overuse’ and ‘underuse’ may co-exist as sources of unwarranted variation, clinicians and managers should examine if all children who would be expected to benefit from VTs for OME also have access to the procedure. The study is of one condition in England but raises an important general issue over using studies of medical practice variations to inform policies

to reduce 'overuse' and thus release resources to meet rising demand in times of austerity. To maximise benefits for patients within resource constraints, policies where medical practice varies ought to tackle 'overuse' by auditing care that is provided, and 'underuse' by assessing capacity to benefit in populations.

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Table 1. Modelling assumptions

	Assumption	Comment
1	Exponential disease process	For the population level an exponential and rate-constant recovery process is applied based on Zielhuis et al. ¹ The authors found a good fit ($r^2 = 0.98$) between the exponential model estimated with Kaplan-Meier technique and the empirical data from a prospective cohort study (n=816 children with valid measurements). For a discussion of the epidemiological models for representing the natural course of OME see ² However, this may mask the few children suffering from highly persistent OME. At the individual level, OME may also be more episodic.
2	Stationary population	Assumes a stable age distribution within each age group and year (based on mid-year population estimates).
3	t	Total waiting time t represents a parameter that reflects demand- and supply-side aspects of patient utilisation behaviour, access and referral policies and the organisation of care delivery. Is varied over a range to account for uncertainty in three distinct sub-intervals: <ul style="list-style-type: none"> • <i>t₁, time to presentation in primary care:</i> Seeks to account for the time lag for detecting hearing loss associated with OME. As OME is an often asymptomatic or ‘silent’ condition, conductive hearing loss is likely to be noted by parents, teachers or carers only after some time (if at all). • <i>t₂, time from presentation in primary care to diagnosis in specialist care:</i> According to the NHS Constitution, patients have a right to be seen by a consultant within maximum 18 weeks after referral.³ This is a political rather than clinical standard. It also refers to maximum not to optimum waiting times. National HES data confirms a median waiting time of 7.3 weeks (51 days) for grommets⁴ from the decision to admit to actual admission (excluding days of deferment and suspension). • <i>t₃, time from diagnosis to confirmation:</i> supposed to be 3 months according to NICE guidance.
4	Incidence is represented as a function of age	Age-based incidence rates are used as the association of OME with age is well-established and most reliably documented. ⁵
5	Incidence rates are at a population level and include both first and recurrent cases	About 50% of children recovering from OME experience a further episode of OME ^{6; 1} . However, due to the often asymptomatic character of OME, even robust incidence studies cannot rule out the possibility that a child has previously suffered from OME. Modelling history of OME could thus lead to an overestimation of cases. Therefore incidence rates used in the model do not differentiate between first-time and recurrent cases and are assumed to include both.
6	Incident cases	Potential underestimation of transient cases occurring and recovering between the screening intervals of 3 months ⁷ or 4 months. ⁸ However, OME is considered a disease occurring only after several weeks of middle ear pathology. ²
7	Seasonal variation in incidence is averaged out over one year.	The incidence of OME is known to be higher in winter; however, the incidence data used in the model and the model output represent an annual average.
8	Fixed proportion of bilateral OME.	Reflects the nature of the data that has been collected at (discrete) screening time points; although at individual level, children may switch between unilateral and bilateral states .

^a References are given in Appendix D.

Table 2. Model parameters

Parameter	Definition	Base value used in model	References ^a	Distribution for sensitivity analysis	Lower quartile; upper quartile ^{10c}
S_j	Number of susceptible children in age group j at risk of developing OME in a given year (reference year 2010).	See Appendix B	¹	-	-
I_j	Age-specific cumulative incidence (risk) of transiting to the OME state over a period of one year by year of age. Diagnosis based on type B tympanogram by the Jerger classification and otoscopy.				
2		0.350	²	β (1.93;1.93;0.15;0.54)	0.280;0.420
3		0.160	²	β (1.93;1.93;0.06;0.25)	0.128;0.192
4		0.160	^{10a}	β (1.93;1.93;0.06;0.25)	0.128;0.192
5		0.278	³	β (1.93;1.93;0.12;0.43)	0.222;0.334
6		0.151	³	β (1.93;1.93;0.06;0.23)	0.121;0.181
7		0.111	³	β (1.99;1.99;0.04;0.17)	0.088;0.133
8		0.065	³	β (1.93;1.93;0.03;0.11)	0.056;0.084
P (Bilateral OME OME)	Conditional probability of bilateral OME given a diagnosis of OME.	0.4	³	β (303;455)	0.38;0.41
P (HL Bilateral OME)	Conditional probability of a hearing level of +25dB given a diagnosis of bilateral OME.	0.35	⁴	β (11;11)	0.3;0.4
m	Median time to recovery ('half life' of OME)	3 months (three-month recovery rate of 0.5)	⁵⁻⁸	Used as deterministic value in the model as found to be consistent across different settings and time periods by various studies.	
t	Total waiting time t from OME onset	$t_1 + t_2 + t_3$	See Table 1	Varied over a range from 0 to 25 weeks	
t_1	Time from OME onset to presentation in primary care	1 month	^{10b}		
t_2	Time from presentation in primary care to formal diagnosis	1 month	^{10b}		
t_3	Time from formal diagnosis to offer of treatment ('active observation' or 'watchful waiting')	3 months	⁹		

^a References are given in Appendix D.

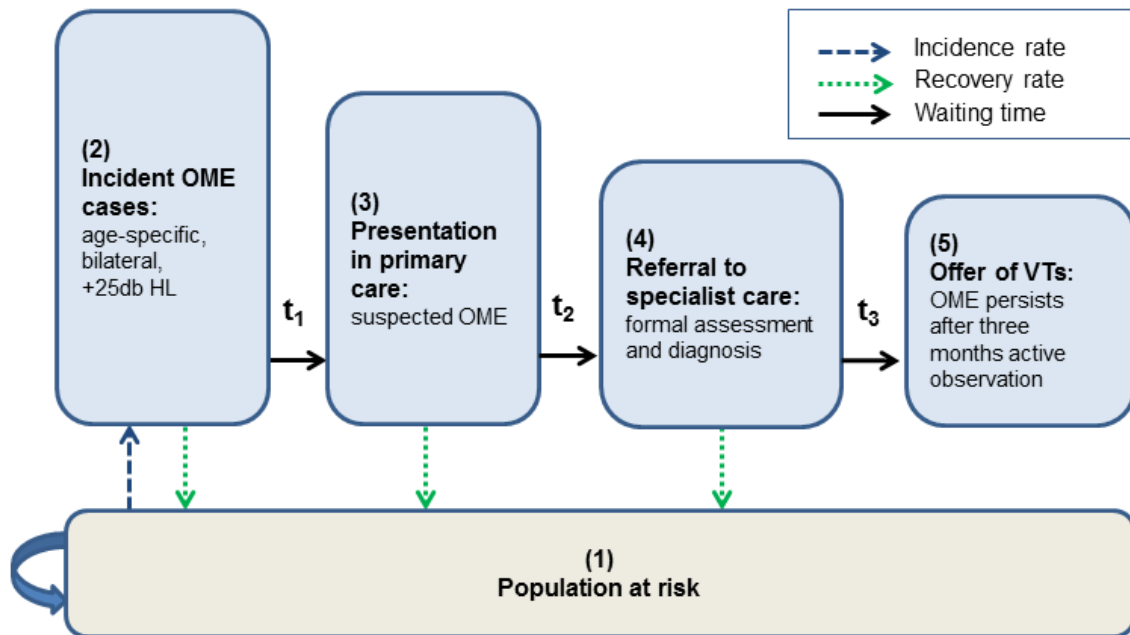
Table 3. Observed VT insertions in England, 2010/11

Observed VT insertions	Count
Total admissions	32,716
Day case	29,566
Age 0-14	23,459
Age 0-12, OME-associated diagnosis codes (2010/11)*	19,805
Age 2-8, OME-associated diagnosis codes (2010/11)*	16,824

Source: NHS Information Centre. Main procedures and interventions: 4 character. Hospital Episode Statistics for England. Inpatient statistics, 2010-11.

* Procedure code D15.1 Myringotomy with insertion of ventilation tube through tympanic membrane for DIAG1=H652: Chronic serous otitis media or H653: Chronic mucoid otitis media or H654: Other chronic nonsuppurative otitis media or H659: Nonsuppurative otitis media, unspecified. Both as primary and secondary procedure (e.g. besides adenoidectomy); including both elective and emergency admissions, in- and outpatient cases

Figure 1. Conceptual model: NICE pathway of care



Explanation:

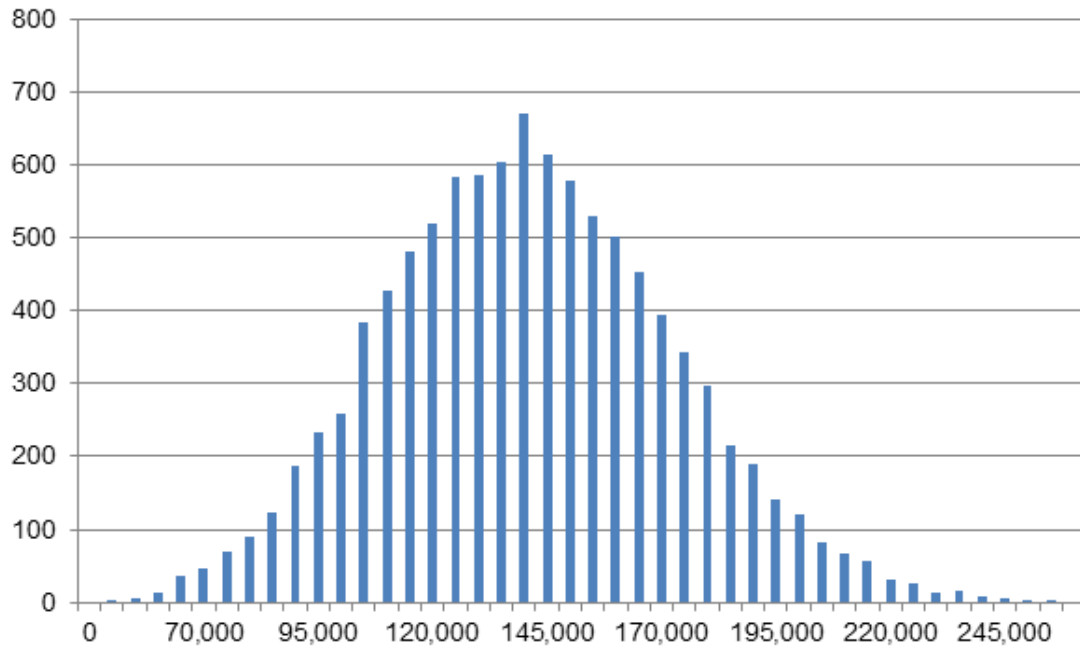
- (1) The model starts with a population of children at risk of developing OME.
- (2) Of these children, some will develop bilateral OME with a hearing level of +25 dBHL.
- (3) The recovery rate determines the proportion of children recovering and ‘returning’ to the susceptible population. The remaining (persistent) cases present in primary care.
- (4) Children who are referred to specialist care undergo formal assessment and diagnosis.
- (5) Patients for whom a diagnosis of OME is confirmed after three months ‘watchful waiting’ have a capacity to benefit from VTs for OME and should be considered for surgical intervention according to NICE guidance.

Legend:

Boxes represent mutually exclusive, collectively exhaustive states in which parts of the population of children find themselves.

Arrows represent the transition probabilities (incidence and recovery rates) and the waiting time that link the states.

Figure 2. Monte Carlo simulation of expected annual incidence of bilateral OME with a hearing level of +25 dB in England (reference year 2010, age groups 2 to 8 years)

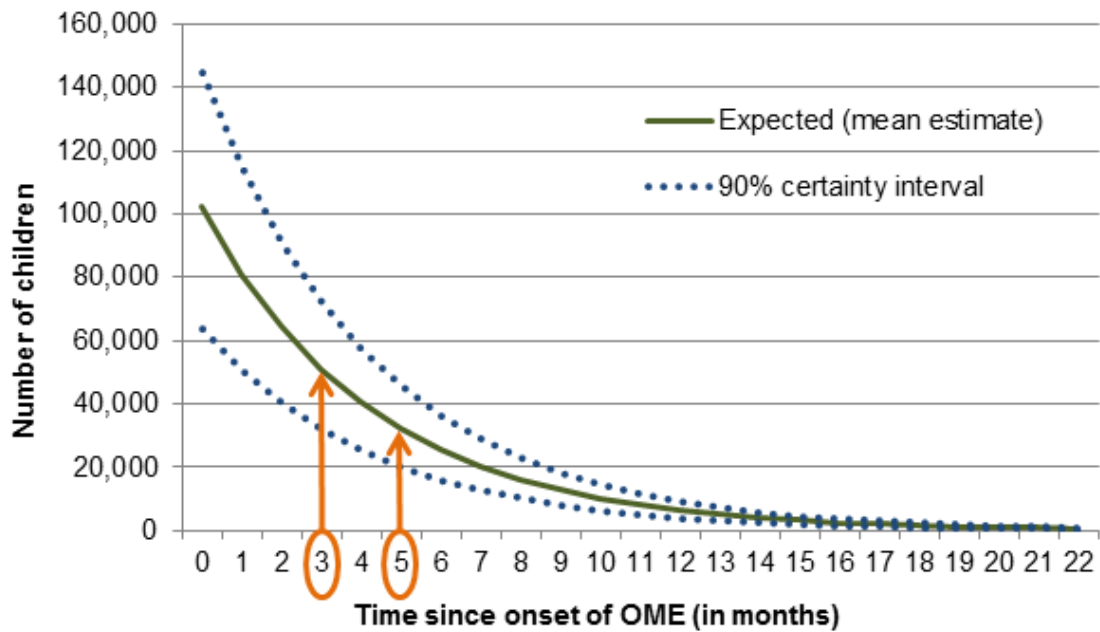


Legend:

x-axis: expected annual incidence of bilateral OME with +25 dBHL in England (2010).

y-axis: frequency of observing a particular output value based on 10,000 iterations of the simulation model.

Figure 3. Expected number of children with capacity to benefit from VTs for OME depending on total waiting time in England (reference year 2010, age groups 2 to 8 years)*



*Given different starting estimates of the total annual incidence of bilateral OME with hearing level of +25dB for the age groups 2 to 8 from the Monte Carlo simulation (Figure 2) of approximately 102,083 cases (mean estimate); 63,800 cases (lower 5% bound); and 143,600 cases (upper 95% bound).

Appendix A. Systematic literature review: Search strategy and data extraction

A systematic literature review was carried out using the databases PubMed, DARE, Scopus, Web of Science and the Cochrane Library (timespan: all available years; restriction to studies in English language). After removing duplicates, 1302 studies were screened independently by the first and second authors based on pre-defined criteria. To be eligible, studies needed to (i) be population-based screening studies; (ii) have a prospective design; (iii) follow defined case finding and diagnostic methods; (iv) provide incidence rates by year of age; and (v) be conducted in Europe or North America. The detailed rationale for each criterion is stated in Appendix B. Study selection was discussed among members of the research team, with the Project Steering Group and during a workshop with UK-based clinical and epidemiological experts. Those studies judged to be in line with the selection criteria were retained.

Database	Search criteria	Number of results
DARE	((("otitis media with effusion" OR "glue ear" OR "non suppurative otitis media" OR "serous otitis media" OR "secretory otitis media" OR "middle ear effusion" OR "purulent otitis media with effusion")) AND (("prevalence" OR "incidence" OR "epidemiology" OR "occurrence")) AND (("child*" OR "kid*" OR "infan*"))	15
Cochrane library	("otitis" "media" "with" "effusion" OR "glue" "ear" OR "non" "suppurative" "otitis" "media" OR "serous" "otitis" "media" OR "secretory" "otitis" "media" OR "middle" "ear" "effusion" OR "purulent" "otitis" "media" "with" "effusion") and ("prevalence" OR "incidence" OR "epidemiology" OR "occurrence") and ("child*" OR "kid*" OR "infan*") not ("acute" "otitis" "media" OR "recurrent" "acute" "otitis" "media") not ("adult*") NOT ("animal*") NOT ("cleft" "palate" OR "down's" "syndrome" OR "down" "syndrome") in Cochrane Database of Systematic Reviews"	57
Web of science	Topic=((("otitis media with effusion" OR "glue ear" OR "non suppurative otitis media" OR "serous otitis media" OR "secretory otitis media" OR "middle ear effusion" OR "purulent otitis media with effusion")) AND Topic=("prevalence" OR "incidence" OR "epidemiology" OR "occurrence") AND Topic=("child*" OR "kid*" OR "infan*")) NOT Topic=((("acute otitis media" OR "recurrent acute otitis media"))NOT Topic=("adult*")NOT Topic=("animal*")NOT Topic=("cleft palate" OR "down's syndrome" OR "down syndrome")) Refined by: Languages=(ENGLISH) AND [excluding] Subject Areas=(PHYSICS OR URBAN STUDIES OR PLANT SCIENCES OR HISTORY) Timespan=All Years. Lemmatization=On	635
PubMed	(((((("otitis media with effusion"[All Fields] OR "glue ear"[All Fields] OR "non suppurative otitis media"[All Fields] OR "serous otitis media"[All Fields] OR "secretory otitis media"[All Fields] OR "middle ear effusion"[All Fields] OR ("otitis media, suppurative"[MeSH Terms] OR ("otitis"[All Fields] AND "media"[All Fields] AND "suppurative"[All Fields]) OR "suppurative otitis media"[All Fields] OR	538

	<p>("purulent"[All Fields] AND "otitis"[All Fields] AND "media"[All Fields]) OR "purulent otitis media"[All Fields]) AND effusion[All Fields]) AND ("prevalence"[All Fields] OR "incidence"[All Fields] OR "epidemiology"[All Fields] OR "occurrence"[All Fields])) AND ("child*"[All Fields] OR "kid*"[All Fields] OR "infan*"[All Fields])) NOT ("acute otitis media"[All Fields] OR "recurrent acute otitis media"[All Fields])) NOT "adult*"[All Fields] NOT "animal*"[All Fields] NOT ("cleft palate"[All Fields] OR "down's syndrome"[All Fields] OR "down syndrome"[All Fields]) AND ("humans"[MeSH Terms] AND English[lang])</p>	
Scopus	<p>(ALL(("otitis media with effusion" OR "glue ear" OR "non suppurative otitis media" OR "serous otitis media" OR "secretory otitis media" OR "middle ear effusion" OR "purulent otitis media with effusion")) AND ALL(("prevalence" OR "incidence" OR "epidemiology" OR "occurrence")) AND ALL(("child*" OR "kid*" OR "infan*")) AND NOT ALL(("acute otitis media" OR "recurrent acute otitis media")) AND NOT ALL(("adult*")) AND NOT ALL(("animal*")) AND NOT ALL(("cleft palate" OR "down's syndrome" OR "down syndrome")) AND (LIMIT-TO(LANGUAGE, "English")) AND (EXCLUDE(SUBJAREA, "AGRI") OR EXCLUDE(SUBJAREA, "PHYS"))</p>	947

Appendix B. Study inclusion criteria

Inclusion criteria	Rationale	Exclusion criteria
Population-based screening study	<p>For valid estimates of incidence, the denominator should include all, or a representative sample of, individuals at risk.</p> <p>As regards hearing loss: most literature focuses on clinical populations. Thus, current hearing loss data is from a community-based study focused on bilateral middle ear effusion¹ (p.44).</p>	<p>1) <i>Utilisation-based studies</i> (i.e. with clinical populations actually visiting the doctor as denominator). Single hospital or practice cannot usually be assumed to provide care for a well-defined population that is representative of a larger group.²</p> <p>2) <i>Trial-based studies</i>. Results may be difficult to generalise to a general population setting, if particular groups are over- or underrepresented.</p> <p>3) <i>Studies with high-risk populations</i> (e.g. pre-term babies on intensive care units, exclusive focus on children in daycare).</p> <p>4) <i>Clinical specialist populations</i> (for estimating the proportion of hearing loss among all OME cases). If the denominator are children who have already been referred to ENT,³⁻⁵ this may either lead to overestimation (due to selectivity of more severe cases) or underestimation (due to bias in detection and presentation among parents and/ or gaps in referral from primary care).</p> <p>5) <i>Self-report studies</i>. As regards incidence and hearing loss, parents have been shown to be inaccurate in their judgments regarding the presence of hearing loss that may accompany an episode of OME.⁶</p>
Prospective design	OME often presents asymptotically, which complicates retrospective diagnosis of OME.	<i>Retrospective designs</i> (e.g. parent interviews or analysis of doctor consultations). These will substantially underestimate the true incidence of OME ⁷ and are thus not a reliable case finding design for OME.
Case finding methods and diagnosis	<p>The recommended diagnostic algorithm for OME combines impedance audiometry (tympanometry) with pneumatic otoscopy.⁸</p> <p>OME is diagnosed when tympanometry reveals a flat curve (relative gradient less than 0.1, type B) or middle ear pressure between -399 to -200 daPa (C2 curve), when the tympanic membrane has no or reduced mobility, or fluid or air bubbles are evident behind the ear drum.⁹</p>	Studies that do not provide correspondingly defined case finding and diagnostic methods.
Stratified by year of age	Incidence of OME is known to vary considerably by age. ¹⁰	<i>Aggregate (e.g. five-year) rates</i> . This is likely to obscure key differences in incidence across age groups.
Studies conducted in Europe or North America	Incidence of OME may be influenced by climatic settings. ¹¹	Studies conducted in different climatic settings than England.

Appendix C. Estimation of susceptible population

For valid estimations of incident cases, children with prevalent OME at the beginning of the study period need to be subtracted from the total population to obtain an estimate of the susceptible population (i.e. the population at risk). This is because the denominator of the cumulative incidence is defined as the number of children at risk at beginning of the study period rather than the total population.¹² Point prevalences are taken from population-based studies. The estimates are lower than those reported in a review by Zielhuis et al,¹⁰ which may be due to the amalgamation of point and period prevalences (time frames over which prevalence has been measured are not reported).

j Age group	P _j Point prevalence (%)	Reference	N _j Total population	S _j = N _j - (N _j *P _j) Susceptible population
2	10.61	¹³	667,185	596,423
3	9.8	²	640,232	577,489
4	8.8	²	620,326	565,737
5	10	²	606,770	546,093
6	6.1	²	598,725	562,203
7	3.04	¹³	577,767	560,183
8	1.11	¹³	560,460	554,233

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Appendix D: Reference list for Tables 1 and 2

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