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Risk factors for failing the hearing screen due to otitis media in Dutch infants

Willeke Lok · Lucien J. C. Anteunis · Cor Meesters · Michelene N. Chenault · Mark P. Haggard

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Abstract Hearing loss from otitis media (OM) can affect young children's development. Some children with persistent OM-related hearing loss and associated problems can benefit from treatment, but researchers and clinicians are still unclear on how to identify them best. The present study aims to determine which factors are most related to the hearing loss in OM, as a first step towards an effective case-finding instrument for detecting infants with persistent OM-related hearing loss. The full PEPPER ('Persistent Ear Problems, Providing Evidence for Referral') item pool includes a wide range of risk factors for OM in a single questionnaire, and is easily completed by parents or guardians. The questionnaire was sent to all children invited for the universal hearing screen at age 9 months in Limburg, The Netherlands. Repeatedly failing of the hearing screen was used as outcome marker indicative of OM-related chronic hearing loss. Univariate analyses were conducted to determine statistically significant risk factors predicting 'fail' cases at this hearing screen. Five items were found as individually predictive of hearing screen failure and subsequent referral: 'having severe cold symptoms', 'attending day care with >4 children', 'having siblings', 'severe nasal congestion' and 'male gender'. Suitably worded parental questions document risk factors for OM-related hearing loss in infants, broadly consistent with past general literature on OM risk factors, but more focused. The findings justify further optimising and evaluation of an additive or multiplicative combination of these questions as a means for selecting and routing an infant with diagnosed or suspected OM to further care.

Keywords Otitis media · Hearing loss · Infants · Risk factors

Present Address:
W. Lok (✉)
Department of Otorhinolaryngology, Albert Schweitzer Hospital, P.O. box 444, 3300 AK Zwijndrecht, The Netherlands
e-mail: w.lok@asz.nl

W. Lok · L. J. C. Anteunis · M. N. Chenault
Department of Otorhinolaryngology and Head and Neck Surgery, Maastricht University Medical Centre, Maastricht, The Netherlands

W. Lok · L. J. C. Anteunis · M. N. Chenault
School of Mental Health and Neuroscience, Faculty of Health, Medicine and Life Sciences, Maastricht University, Maastricht, The Netherlands

C. Meesters
Department of Clinical Psychological Science, Faculty of Psychology and Neuroscience, Maastricht University, Maastricht, The Netherlands

M. N. Chenault
Department of Methodology and Statistics, Faculty of Health, Medicine and Life Sciences, Maastricht University, Maastricht, The Netherlands

M. P. Haggard
MRC Multicentre Otitis Media Study Group, Department of Experimental Psychology, University of Cambridge, Cambridge, UK

Introduction

Otitis media (OM), highly prevalent in the first 2 years of life [1–5], is characteristically fluctuating in duration [1, 3, 6] and often accompanied by varying degrees of hearing loss [7, 8]. OM-induced hearing loss often goes undiagnosed, as parents are unable to recognise it [9–11]. It can be detrimental to child development, including speech and language development, behaviour

and the general quality of life of the child [1, 12–14]. Until recently, screening for congenital or early acquired sensorineural or permanent conductive hearing loss, with an expected incidence of <0.2%, occurred at 9 months of age in the Netherlands. That screen had 5–7% fails, mostly due to OM-related hearing loss (data from 1995 to 2004, Dutch Society of the Deaf and Hearing Impaired Child; NSDSK) [15]. Children failing the hearing screen repeatedly were referred for treatment and many underwent tube insertion. Therefore, the screen also functioned as a tool for identification and treatment of persistent OM-related hearing loss. This hearing screen at 9 months of age has now been replaced by neonatal screening for earlier detection and rehabilitation of congenital hearing loss. Without the screen at age 9 months, detecting infants with hearing loss (e.g. due to OM) developing after the neonatal period would be more difficult, so reduced numbers of children receiving tubes might be expected. Surprisingly, however, more children, not fewer, are now treated with tubes at a very young age since [16]. The reasons for this increase in the number of children treated are unknown, but could be related to clinical uncertainty with subsequent over-treatment, implying a need for further bases of selection. The present study explores the risk factors for persistent OM-related hearing loss as a possible basis for selective referral from general practice. It uses a large sample of otherwise healthy young infants from the general Dutch population that was routinely invited for a population screen. Referral after a repeated hearing screen failure provides the relevant outcome. The approach is univariate, to help comparison with risk factors for OM.

Methods

PEPPER item pool

The PEPPER item pool ('Persistent Ear Problems, Providing Evidence for Referral'), initially developed in the UK, embraces a wide range of OM-related factors in a single instrument for use in primary care and can be completed by the child's parents or guardians within 3 min. To facilitate use, the items were pooled into the PEPPER questionnaire. The English version of this questionnaire (Appendix) was translated into Dutch, and then back into English by an English native speaker.

Universal hearing screen

The hearing distraction test (CAPAS, Compact Amsterdam Paedo-Audiometrical Screening, a screen at 35 dB SPL based on visual re-enforcement audiometry; see Rovers et al. [15] for details) was a population screen conducted at age 9 months by special trained employees at the well-baby clinic. After failing the first test, children were screened a second time 1 month later, and again 1 month later after failing the second test. Children were referred to their general practitioner (GP) upon either failing the CAPAS three times or failing twice, combined with other problems warranting referral, for example developmental problems or suspected severe hearing loss. Referral (i.e. failing CAPAS repeatedly) is taken here as marking persistent hearing loss.

Information about infants failing the screen with a permanent conductive or a sensorineural hearing loss was provided by the regional audiology centre. Here, a specialised multidisciplinary team assessed the infants with a chronic hearing loss using voluntary response (visual re-enforcement audiometry) and fully objective tests (auditory brainstem response, auditory steady state response, tympanometry) and provided rehabilitation when needed. Children with such permanent impairments were excluded from our database, leaving the cases with OM-related hearing loss.

Study protocol

Parents of all children born between 1 June 2004 and 31 December 2004 in the province of Limburg, The Netherlands, received the routine CAPAS invitation, along with information regarding this study, a consent form and the PEPPER questionnaire, which the parents were asked to complete and bring to the well-baby clinic at the screening visit. The results of the questionnaire were not shared with the well-baby clinic doctors and therefore did not influence routine practice at the well-baby clinic.

Questionnaires were scanned into an SPSS file (Statistical Package for the Social Sciences; version 15.0), checked and merged with the CAPAS data. Excluded from the study database were children with sensorineural hearing loss or permanent conductive hearing loss and children with Down syndrome and cleft palate or other cranio-facial malformations.

Statistical analysis

Univariate logistic regression, using SPSS version 15.0, was applied to determine factors predicting referral. Response categories of some PEPPER items were combined when category counts were extremely small, as noted in "Appendix". The items 'number of children in day care' and 'breastfeeding' were dichotomised into 'attending day care with >4 children' and 'at least 3 months exclusive

breastfeeding' to facilitate comparison with earlier studies [17–22]. 'Early birth' means gestational age <37 weeks.

Comparison with other studies

A literature search using the terms 'otitis media', 'risk factors' and 'infants' resulted in a set of studies of risk factors for OM. Studies based on consultation rather than screening, that included children over 2 years, or studies with acute otitis media (AOM) or otorrhoea as outcome variable were excluded and only studies in developed countries were included. The search failed to find any study specifically on risk factors for the hearing loss related to OME at this age, but did yield eight studies on multiple risk factors for OM [17–25].

Results and study comparison

The response rate was 56.4% (6531 questionnaires sent, 3681 completed, 50.1% boys, 49.8% girls). Only 13 cases did not complete the questionnaire prior to the screen according to protocol, and leaving these cases out of the analysis did not change the results. Mean age at completion was 9 months 6 days (range 6 months 18 days to 16 months 9 days) and first CAPAS screening 9 months 24 days (range 7 months 21 days to 13 months 24 days) (see Fig. 1).

In Table 1, ORs and 95% CI are presented for each item. Question 1 and the category 'other' from questions 2, 3 and 4 were dropped because of inconsistent interpretation and irrelevant responses.

Four highly significant ($p < 0.001$) risk factors were found: 'having severe cold symptoms', 'attending day care with >4 children', 'having siblings' and 'male gender'. Risk factors significant for referral with a p value of <0.01 were 'severe nasal congestion', 'siblings with a history of ear/hearing problems' and 'father working part time'. However, when the factor 'siblings with a history of ear/hearing problems' ($p = 0.003$) was adjusted for 'having siblings', this item became non-significant (OR = 1.2; $p = 0.44$).

'Breastfeeding for at least 3 months' had a paradoxical 1.5 higher odds ($p = 0.01$), rather than being protective for referral. 'Season of CAPAS' being January–March or July–September had increased odds for referral with a $p = 0.05$. However, the CAPAS screen is no longer in use and therefore this factor will be irrelevant in the future.

Comparison with other studies

To aid comparison, the results of the reference studies investigating more than one risk factor are also summarised in Table 1 and mentioned here, leaving wider interpretation to the discussion section. There are two reports from one single study [19, 20]. One study [21] provided only ORs without statistical significance levels and another [23] is not included in the table as ORs were not given at all. Although populations can differ and some risk factors may be more specific for hearing loss rather than for OM, the large sample size makes the present study more powerful than others with fewer false-positive findings for expected trends.

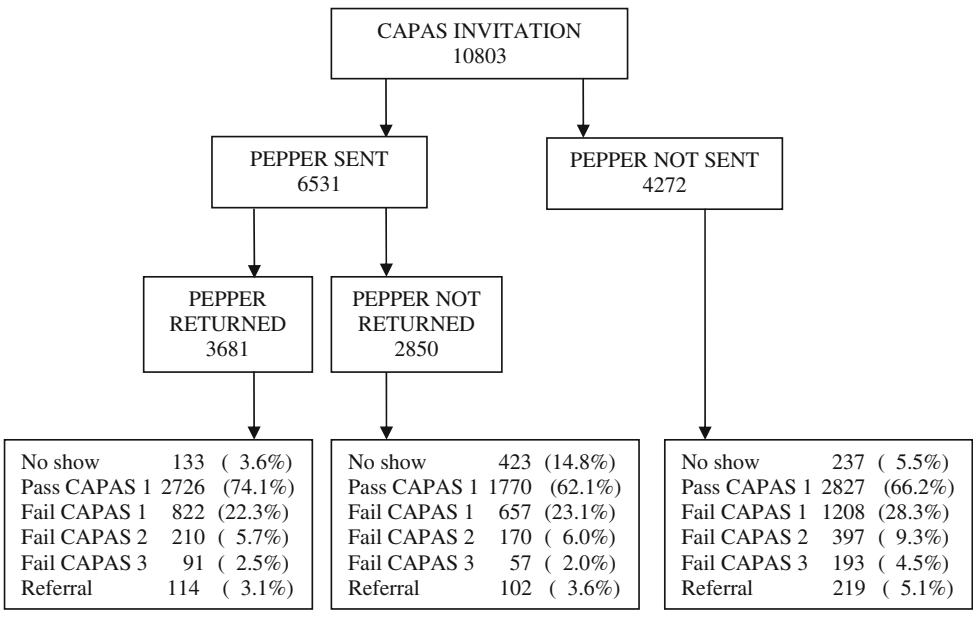

Fig. 1 Number of children invited for the hearing screen and response rates for the PEPPER questionnaire and hearing screen

Table 1 Questions in the PEPPER item pool with OR and 95% CI and results found in the literature for the corresponding risk factors for OM

	Lok OR (95%CI)	Alho et al. [24] OR (95%CI)	Dewey et al. [17] OR (95%CI)	Engel et al. [18] OR (95%CI)	Kraemer et al. [25] OR (95%CI)	Rovers et al. [19, 20] OR (95%CI)	Sassen et al. [21] OR	Zielhuis et al. [22] OR (p)
Q3: Delay in growth ~ low birth weight (yes/no)	1.1 (0.3–4.5)		1.3 (1.0–1.6)*	# **	1.3 (0.3–5.0)	0.4 (0.2–0.9)		1.61 (0.190)
Q3 a: Early birth (<37 weeks) (yes/no)	1.0 (0.5–1.9)		#	# *		0.5 (0.3–1.1)		#0.74 (0.354)
Q4: Meconium-stained amniotic fluid (yes/no)	0.9 (0.5–1.4)							
Q5: Age of first ear infection								
0–5 months	0.7 (0.3–1.6)				3.0 (1.2–7.4)			
6–9 months	1.0 (0.6–1.8)							
0–9 months ~ history of an ear infection	0.9 (0.6–1.4)			1.8 (1.3–2.6)***		1.3 (0.9–1.8)	2.12	
Q6: Age of first hearing problem								
0–5 months	0.9 (0.2–3.8)							
6–9 months	0.6 (0.1–4.0)							
0–9 months ~ history of a hearing problem	0.7 (0.2–2.4)			1.7 (1.0–2.7)*				
Q7: An ear infection in the last 3 months (yes/no)	1.2 (0.8–1.8)	#						
Q8: An ear infection in the last month (yes/no)	1.3 (0.8–2.3)	3.7 (1.2–11.2)						
Q9 a: Mouth breathing (yes/no)	1.4 (0.8–2.3)			1.3 (1.0–1.6)*				
Q9 b: Nasal congestion (yes/no)	1.8 (1.2–2.8)**				#			
Q9 c: Snoring (yes/no)	1.3 (0.8–2.3)			1.1 (0.9–1.4)				
Q9 d: Cold symptoms ~ URTI (yes/no)	1.9 (1.3–2.7)***			1.6 (1.2–2.0)***		2.6 (2.0–3.6)	1.46	
Q10: Smoking around child (yes/no)	1.0 (0.6–1.6)				#	1.0 (0.7–1.4)	0.95	1.11 (0.643)
Q11: Number of cigarettes smoked								
0–20	1.1 (0.7–1.7)				1.9 (0.7–5.3)			
10–19			1.6 (1.1–2.3)*		#			
>20	1.3 (0.4–4.2)		0.8 (0.5–1.3)			1.0 (0.7–1.4)		
Q12: Heating of the house (reference = central heating)								
Stove	0.6 (0.1–4.5)							
Central heating and also a stove	1.1 (0.7–1.8)							
Q13: Breastfeeding (yes/no)	1.5 (1.1–2.1)*		#	1.1 (0.8–1.6)	1.1 (0.5–2.7)	1.1 (0.8–1.5)	0.98	#0.71 (0.187)
Q14: Sleeping habit (reference = back)								
Side	1.1 (0.7–1.8)							

Table 1 continued

	Lok OR (95%CI)	Alho et al. [24] OR (95%CI)	Dewey et al. [17] OR (95%CI)	Engel et al. [18] OR (95%CI)	Kraemer et al. [25] OR (95%CI)	Rovers et al. [19, 20] OR (95%CI)	Sassen et al. [21] OR	Zielhuis et al. [22] OR (p)
Stomach	1.2 (0.8–1.8)							
Q15 a: Sucking is weak (yes/no)	1.2 (0.5–2.7)							
Q15 b: Slow to feed (yes/no)	1.6 (0.9–2.9)							
Q16: Day care (yes/no)	1.4 (1.0–1.9)	2.56 (1.2–5.6)*		1.6 (1.1–2.2)**		1.9 (1.3–2.6)	1.67	1.88 (0.007)
No. of children attending: >4	1.9 (1.3–2.7)***		1.4 (1.0–1.8)*					
Q17: Having siblings (yes/no)	2.8 (1.9–4.1)***	1.77 (0.96–3.3)	1.5 (1.2–1.9)*		0.6 (0.3–1.3)	3.2 (2.3–4.4)	1.97–5.60	# (0.010)
More than one compared to one				1.5 (1.2–2.0)*				
Q18: Family history of ear/hearing problems								
(yes/no)	1.0 (0.7–1.4)			0.8 (0.6–1.0)	1.9 (0.9–3.6)	1.9 (1.4–2.5)	1.73–2.68	
Siblings with a history of OME (yes/no)	1.7 (1.2–2.5)**							1.85 (0.005)
Q19: Father working (reference = full time)								
Part time	2.0 (1.2–3.3)**							
Not working	0.8 (0.4–1.5)							
Q20: Mother working (reference = not working)								
Part time	1.2 (0.9–1.8)							
Not working	1.3 (0.6–3.1)							
Gender = male	1.9 (1.3–2.7)***	2.17 (1.4–3.4)*		1.0 (0.8–1.3)		1.4 (1.0–1.8)	1.24	1.50 (0.055)
Season (reference = October–December)								
January–March	#	#	1.7 (1.4–2.1)*					#
April–June	#	#	1.0 (0.8–1.2)					#
July–September	#	#	0.5 (0.4–0.6)*					#

*** p-value < 0.001; ** p-value < 0.01; * p-value < 0.05

The risk factor is mentioned in the article without specifying the RR or OR, or the risk factor is not exactly the same and cannot be added to the table

Classical risk factors

Siblings

'Having siblings' is a risk factor, consistent with the reference studies [17–22], although our estimate is greater in magnitude. Risk probably increases with more siblings, as reported elsewhere [21], but we were unable to test this with our data.

Day care

'Attending day care' was a risk factor in five of the comparison studies [18, 20–22, 24], but not in ours. We did find increased odds with 'attending day care with >4 children' (OR = 1.9; $p < 0.001$) indicating that it is indeed the number of children an infant is exposed to which is relevant [4].

Gender

Boys have almost twice the odds for developing OM-related hearing loss (OR = 1.9; $p < 0.001$) and this agrees with one other included study [24], as well as with the general background of literature on OM. One study [22] showed a marginal trend (OR = 1.5; $p = 0.055$) and another [21] reported an increased risk without specifying significance. Overall, boys are at raised risk for OM [4, 24, 26].

Genetic disposition

'Siblings with a history of ear/hearing problems' emerged as a risk factor (OR = 1.7; $p = 0.003$), consistent with one previous study [22]. However, upon adjustment for 'having siblings' this risk decreased and was no longer significant. It seems that 'having siblings', an environmental risk factor, is a stronger risk factor than genetic disposition. Having 'parents with a history of ear/hearing problems' was not a risk either.

Parents' working status

'Father working part time' had significantly higher odds (OR = 2.0; $p = 0.007$), while mother's working status appeared to be unrelated to referral. There were no studies for comparison.

Breastfeeding

'Breastfeeding' emerged as an apparent risk rather than a protective factor. One reference study [23] reported breastfeeding as protective, while six other studies [17–22, 25] were unable to do so. One [22] did report that the longer a child was breastfed, the less the risk for developing OM. Varying definitions of breastfeeding or the absence thereof make comparison difficult (>7 months of breastfeeding [22]; exclusive breastfeeding for 6 months [25]; exclusive breastfeeding for >4 months [17, 22]; median of 2 months breastfeeding [18]; at least 6 months breastfeeding [19, 20]; no definition [23] [21, 24]).

Upper respiratory tract infection symptoms (URTI)

Mouth breathing, snoring and nasal congestion can all be symptoms of URTI and can be related to adenoid hypertrophy. These factors could therefore impose a risk for developing OM and OM-related hearing loss. Neither 'severe mouth breathing' nor 'severe snoring' appeared to be risks here or in another study [18].

'Severe nasal congestion' appeared as a risk here (OR = 1.8; $p = 0.006$), and in one reference study [25] which showed that the risk increased with growing number of days with nasal congestion. 'Severe cold symptoms' (OR = 1.9; $p < 0.001$), which is less specific, embracing coughing, common cold symptoms and sore throat, was also significant in all four studies reporting on it [18–21, 23].

The understanding of 'severe cold symptoms' as well as 'severe nasal congestion' probably varies much amongst parents and might be rather imprecise. Although we are obviously not dealing with a homogenous group, the results show that these items do predict referral and therefore they can be of interest.

Ear and hearing problems

Having had a history of hearing problems [18], ear infections [18, 21, 24, 25] or early OME [23] or early otitis [25] have all been reported as a risk for developing chronic OM. However, they were not related to hearing in our study, or in one other study [19, 20]. Our study population was very young, making it difficult for their parents to detect hearing loss or ear infections, and this could have influenced the results.

Smoking

Smoking around the child by household members or the number of cigarettes smoked inside the house did not appear to be a riks factor. This is in line with the results of several recently completed studies [18–23]. However, one of the reference studies [17] found that smoking 10–19 cigarettes per day was a risk, although smoking >20 was not. Another study [25] found that the number of smokers around the child did increase the risk of persistent OM,

although in that study the number of cigarettes smoked was not a significant risk. We did not ask about smoking habits around the child outside the house, and therefore the results may be an underestimation.

Season

OM is least prevalent in summer [3, 5, 22] and OM first detected in fall and winter has a greater tendency to persist [6]. Two reference studies [17] [18] confirmed this classical effect. We did not find season to be a risk factor when considering the four traditional seasons. However, when October through March were compared to April through September, we did find a 1.5 higher odds for referral ($p = 0.065$), which was in concordance with two other studies [22, 24].

Less conventional risk factors

Prenatal and birth characteristics

Low birth weight was not defined uniformly, hindering comparison (<2500 g [19, 22, 25]; <3100 g [17]; <3400 g [18]). We used the less specific term 'delay in growth' as a marker for low birth weight. It did not appear to be a risk factor. Two reference studies [17, 18] found low birth weight to be a risk factor, two did not [22, 25], and one [19] even found low birth weight to be protective.

Prematurity is usually defined as a gestational age <37 weeks [17, 19, 20], although <38 weeks has also been used [22]. One study [18] analysed prematurity appropriately as a continuous measure, finding all children born before a gestational age of 40 weeks having an increased risk for OM, this risk being greater the more premature the birth was. None of the other studies, including ours, found prematurity to be a risk factor.

'Meconium-stained amniotic fluid' has been studied as a risk in developing OM, using varying definitions and subsequently finding conflicting results [27–29]. Overall, these studies, including ours, did not find the mere presence of meconium-stained amniotic fluid to be a risk [27, 29]. A recent study [28], using the stringent definition of meconium-stained amniotic fluid being present at birth with pulmonary aspiration requiring tracheal suction and treatment at a neonatal intensive care unit, did find an increased risk for AOM. That study mentioned that the mechanism was obscure but presumably related to immune immaturity, although it might also be related to treatment.

Sucking and eating

The items 'sucking is weak' and 'slow-to-feed' could be seen as symptoms of nasal congestion and/or adenoid hypertrophy, being proxies for mouth breathing, but more appropriate questions to ask in the child under 6 months. These items were not predictive overall for referral and there were no studies for comparison.

Sleeping position

Sleeping in the prone position has been reported as an increased risk for coughing, earache and hearing problems in the young infant [30] or for 'having ear infections' and developing a 'stuffy nose at 6 months of age' [31]. In our study however, it may not have emerged as a risk, as the number of children sleeping prone was too few to detect a difference at statistical significance.

Heating of the house

We found no studies on 'heating of the house', which could reflect socioeconomic status, general environmental stress or indoor air quality. The effect of using secondary home heating sources (a fireplace, wood-burning stove, kerosene heater or a gas stove) has been studied before in developing AOM [32]. Neither in that study nor in ours was a significant association found. Although indoor air quality does seem to affect URTI [33] and hence possibly OM, the absence of central heating in the Netherlands is too rare to detect any such effect.

General discussion

Researchers as well as clinicians know that there are children with persistent OM-related hearing loss who might benefit from treatment [34, 35]. For example, one study [36] showed that with every dB improvement in hearing, the comprehensive language development improved with 0.05 month. Identifying these children is however difficult and screening for hearing loss in a healthy population and subsequent treatment with tubes is not effective [34, 35]. The increase in number of very young children treated with tubes in the Netherlands [16] may reflect clinical uncertainty and consequent over-treatment. It shows the need for more selective case finding. Risk factor studies in OM have not used OM-related hearing loss as the outcome, although hearing loss resulting from OM should be a major concern, as it is the presumed route to developmental delays, so justifying parental concern and medical attention. This serious omission is met by our study.

In the present study, we cannot distinguish between general factors leading to non-persistent OM and specific factors leading on to persistent OM and/or on to subsequent OM-related hearing loss. However, it is reasonable to assume that the overlap is substantial, allowing us to

compare our findings with those for OM [17–25]. The results of our study do, to some extent, differ from the results of the studies used for comparison, but no more than those studies' results differ from each other, so the former contrast should not be overplayed. Differences in the reference group used and how the potential risk factors have been operationalised will clearly affect the magnitude of ORs, while sample size will affect the statistical significance. Furthermore, differing results may reflect varying exposure to risk factors between populations [37] and international or policy differences in care seeking for ear problems [38].

The historical opportunity form the change in system places our results in 2004; extrapolating these results to the present day should be done with some caution, as some of the studied items are not proximate risks for developing OM-related hearing loss, instead for example social conditions (work, socioeconomic status) are mere proxies for underlying risks. These social conditions can change over time; however to our knowledge, no such major change has been recorded.

Conflicting or surprising findings

Our study did not find parent-reported ear infections to predict referral. As some OM history is fairly common, the inconsistency here may be due to whether the controls are disease free or at least unselected, and the difficulty in capturing strength of history in a still young infant.

The risk of having 'siblings with a history of ear/hearing problems' decreased after adjustment for 'having siblings' and was no longer significant. We assume that the risk of having 'siblings with a history of ear/hearing problems' is not a marker of genetic disposition, but rather a marker for shared environmental risk factors. This explains why the risk diminished when adjusting for 'having siblings'.

The fact that breastfeeding appeared to be a risk rather than a protection might seem surprising. Perhaps the protective effect of breastfeeding is small, short lasting or hard to capture, as children could be (partly) switched to bottle feeding at some time. Another explanation may be that breastfeeding and referral are both associated with the socioeconomic status of the parents. More highly educated parents usually extend the period of breastfeeding [39, 40] and higher SES is associated with greater uptake of health care [41]. A child could be breastfed for a longer period of time, but also be referred despite having fewer or milder problems because of parental concern expressed at the well-baby clinic, as there is room for such individual concern in the referral process. Selection bias may also play a role here, as parents who breastfeed may also be more likely to have concerns about their child's ear status and to return questionnaires. Furthermore, we are studying OM without signs of an acute infection. A recent study investigating the risks of formula feeding did find a protective effect of breastfeeding against developing acute OM [42]. Together, these considerations could explain the marginal inverse effect found here and some of the inconsistencies in findings on breastfeeding generally.

The items on parents' working status were included to capture socioeconomic status. The increased risk with a father working part time is puzzling, possibly a random finding or an obscure socioeconomic marker in this population. If this item is included in future studies for replication, it would be useful to ascertain the type of work the father does, the reasons for working part time and the number of hours worked per week.

Home air quality deserves consideration alongside smoking, although both have become difficult to study with changes in standard of living and lifestyle, perhaps contributing to null results. Our study only showed a trend in the number of cigarettes smoked in the house, whilst reported smoking around the child was not statistically significant at all. Results on consumption of addictive substances are known to be distorted by a social desirability bias in reporting known or believed risk factors under some degree of voluntary control. A study with an objective marker of exposure to smoking did find an accompanying risk for OM and recurrent OM [43].

Eight items were found to be associated with a higher risk of referral after failing the hearing screen. The results of both the items 'breastfeeding' and 'father working part time' were puzzling. Furthermore, 'siblings with a history of ear/hearing problems' became non-significant when it was adjusted for 'having siblings'. Therefore five reliable risk factors, consistent with past literature, are simply predictive for referral: 'having severe cold symptoms', 'attending day care with >4 children', 'having siblings', 'severe nasal congestion' and 'male gender'.

Study limitations

Response rate

Our response rate was 56.4%, which although reasonable for a service-based population study [44] invites replication in other studies using perhaps fewer items. Response rates do not necessarily influence which items are found to be predictive, although they obviously influence statistical reliability and can influence the OR of a specific item [45]. To determine any participation bias, we compared the responders to the non-responders. Fewer children of responders failed to show up at the first hearing screen, compared to non-responders (3.6 vs. 14.8%, see Fig. 1). As the PEPPER questionnaire should have been completed

and brought to the well-baby clinic during the first testing, we received fewer questionnaires from the group not attending the hearing screen. Furthermore, the non-responders lived in areas with relatively higher percentage migrant population, more unemployment and lower incomes, all of which co-indicate a lower SES, and this could explain the non-responding [46]. At the same time, there appears to be no real bias in referral. Slightly fewer children from responders were referred compared to non-responders (3.1 vs. 3.6%, not statistically significant different). Thus, we appear to be reporting on a sub-population with a high participation rate both in research and routine service, but without difference in outcome measurement.

Individual outcomes from the screen

Precise reasons for screen failure are unknown in individual cases. However, most infants who repeatedly fail a distraction hearing screen have conductive hearing loss due to OM [15, 47, 48]. A previous study [15] reported that 58% of the children failing the CAPAS and referred to an ENT department were diagnosed with bilateral OME and 70% with bilateral or unilateral OME, as confirmed by otoscopy and tympanometry at the ENT department. There was a delay between the hearing screen and the diagnosis at the ENT department; although this was not long (0.8 months) it could however have led to an underestimation of the percentage of children with bilateral OME at the time of referral. The time for spontaneous resolution varies, but can be 11–40% in 4–6 weeks, depending on criteria used [1, 3, 6]. The percentage of children with bilateral OME at the time of referral was probably higher than the reported 58%. We therefore assume that most hearing screen fails are indicative of persistent OM-related hearing loss.

The present study is cross-sectional and thus precludes capturing all children who may be prone to long-persisting OM with accompanying hearing loss. Obviously, children may develop OM-related hearing loss after passing the hearing screen, due to OM being a seasonally fluctuating condition. Such children will have entered the control group. This will not necessarily distort the profile of risk factors, but it will reduce sensitivity.

Implications for future research

The present results are encouraging for low-cost questionnaire-based screening. Future research should focus on creating a practical short case-finding instrument for persistent OM-related hearing loss. The PEPPER questionnaire can be completed by the child's parents or guardians within 3 min. However, processing the data from even a short questionnaire requires staff time and facilities. We do not yet feel justified in recommending the five selected items as a short form for immediate use with simple equal-weight scoring.

Multi-variable modelling can reveal optimal combinations of risk factor items and optimal scaling of item response levels. An optimum scoring algorithm based on such an item selection then needs to be evaluated and the predictive value should be tested again, thereby verifying the optimum scoring. At the same time issues of practical implementation should be addressed, where after routine implementation can be organised.

Conclusions

Five reliable items, consistent with the reviewed literature, are associated with a higher risk of referral after failing the hearing screen: 'having severe cold symptoms', 'attending day care with >4 children', 'having siblings', 'severe nasal congestion' and 'male gender'. Combinations of these factors via multi-variable models might be worth optimising to see whether a case-finding instrument for routine referral of young children with chronic OM-related hearing loss can be developed.

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Conflict of interest There were no conflicts of interest.

Appendix

Pepper ear problem checklist (DOC 29 kb)

PEPPER EAR PROBLEM CHECKLIST

Q1 Is your child healthy or has (s)he a special condition? Yes/no.
Q2 Does your child have special condition? Yes/no.
Special condition: Down syndrome, Cleft syndrome, other.
Q3 Were there special events during the pregnancy? Yes/no.
Special events: infection during pregnancy, growth retardation, early birth, other.
Q3*: 'early birth is a gestational age <37 weeks'. Yes/no.
Q4 Were there special events during delivery? Yes/no.
Special events: meconium stained birth water, slow start, other.
Q5: Please give your best estimate of how old your child was when (s)he first had an ear infection?
Answers: Younger than 3 months, 3-5 months, 6-7 months, 8-9 months, not applicable.
Q6: Please give your best estimate of how old your child was when (s)he first had a hearing problem?
Answers: Younger than 3 months, 3-5 months, 6-7 months, 8-9 months, not applicable.
Q7: In the last three months including today, how many ear infections has your child had?
Answers: None, 1, 2, 3, more than 3.
Q7*: 'having had at least one ear infection in the last three months'. Yes/no.
Q8: How many of these ear infections occurred just in the last month?
Answers: None, 1, more than 1.
Q8*: 'having had at least one ear infection in the last month'. Yes/no.
Q9a: In the last three months, has your child breathed mainly through the mouth?
Q9b: In the last three months, has your child sounded as if she/he had a stuffed nose?
Q9c: In the last three months, has your child snored?
Q9d: In the last three months, has your child suffered from any of coughs, colds or sore throats?
Answers 9a-9d: never, rarely, only during a cold, often, always, not sure.
Q9a-9d*: Answer 'yes': 'often' and 'always'.
Q9a-9d*: Answer 'no': 'never, 'seldom', 'only with a cold' and 'not applicable'.
Q10: Which members of the household currently smoke in the same room as the child?
Answers: none, mother, father, childminder, other.
Q10*: 'at least one smoker around the child'. Yes/no.
Q11: How many cigarettes altogether are smoked in the home each day?
Answers: none, less than 20, 20 or more.
Q12: How is the house being heated?
Answers: with central heating, with a wood burning stove, with central heating but there is also a fireplace/wood burning stove.
Q13: What sort of milk feeds did your child have during the first 6 months?
Answers: breast milk from birth until ... months of age and infant formula from ....months, only infant formula.
Q13*: 'at least 3 months of breastfeeding'. Yes/no.
Q14: As a baby, does your child usually sleep on his/her.....?
Answers: front, side, back.
Q15a: Would you describe your baby (up to the age of 6 months) as having a weak suck? Yes/no.
Q15b: Would you describe your baby (up to the age of 6 months) as being slow to feed? Yes/no.
Q16: Does your child attend nursery, playgroup, and childminder? Yes/no.
Q16 extra: How many children are there next to your child? .....
Q16*: 'attending day care with > 4 children'. Yes/no.
Q17: How many children living at home (not counting this child) attend school, nursery or playgroup?
Answers: one or more older children, other child at home, but does not attend, no other child at home.
Q17*: 'having at least one sibling'. Yes/no.
Q18: This question refers to family members other than this child and only to blood relatives. Has either parent, or any brother/sister of this child, has similar ear or hearing problems?
Answers: yes, needed operation, yes, but no operation, not sure, not had a problem.
Q18*: 'having at least one family member with ear of hearing problems' (father, mother or sibling). Yes/no.
Q19: Does father work: part time/full time/not applicable.
Q20: Does mother work: part time/ full time/not applicable.

Response categories of some PEPPER items were combined when category counts were extremely small, noted above with *

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