International Journal of Audiology

ISSN: 1499-2027 (Print) 1708-8186 (Online) Journal homepage: http://www.tandfonline.com/loi/iija20

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**To cite this article:** Amanda J. Hall, Rachel Humphriss, David M. Baguley, Melanie Parker & Colin D. Steer (2015): Prevalence and risk factors for reduced sound tolerance (hyperacusis) in children, International Journal of Audiology, DOI: 10.3109/14992027.2015.1092055

**To link to this article:** http://dx.doi.org/10.3109/14992027.2015.1092055

Published online: 07 Dec 2015.

Taylor & Francis
Taylor & Francis Group

International Journal of Audiology 2015; Early Online: 1–7

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Original Article

# Prevalence and risk factors for reduced sound tolerance (hyperacusis) in children

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## Abstract

*Objective*: To estimate the prevalence of reduced sound tolerance (hyperacusis) in a UK population of 11-year-old children and examine the association of early life and auditory risk factors with report of hyperacusis. *Design*: A prospective UK population-based study. *Study sample*: A total of 7097 eleven-year-old children within the Avon longitudinal study of parents and children (ALSPAC) were asked about sound tolerance; hearing and middle-ear function was measured using audiometry, otoacoustic emissions, and tympanometry. Information on neonatal risk factors and socioeconomic factors were obtained through parental questionnaires. *Results*: 3.7% (95% CI 3.25, 4.14) children reported hyperacusis. Hyperacusis report was less likely in females (adj OR 0.64, 95% CI 0.49, 0.85), and was more likely with higher maternal education level (adj OR 1.72, 95% CI 1.08, 2.72) and with readmission to hospital in first four weeks (adj OR 1.98, 95% CI 1.20, 3.25). Report of hyperacusis was associated with larger amplitude otoacoustic emissions but with no other auditory factors. *Conclusions*: The prevalence of hyperacusis in the population of 11-year-old UK children is estimated to be 3.7%. It is more common in boys.

**Key Words:** ALSPAC; hyperacusis; hearing

Hyperacusis is defined as an abnormal lowered tolerance to sound (Baguley & Andersson 2007), and may arise within either the peripheral or central auditory system (Baguley et al, 2013a). An associated term is phonophobia (Jastreboff & Hazell, 2004), which is applied both to persons with a fear of certain sounds and also to the auditory sensitivity issues associated with migraine (Baguley et al, 2013a). Misophonia (Jastreboff & Hazell, 2004), is applied to persons with an aversive reaction to sounds, most commonly eating and respiration sounds. The term 'sensory over responsivity' has also been used but applies not only to auditory sensation but to tactile and visual stimulation also (Reynolds & Lane, 2008). Terminology in this area is unclear and it is challenging to distinguish between a subjective increase in the perceived intensity of sound, and the emotional reaction to that percept, particularly in children (Asha et al, 2010). In this paper we use the term hyperacusis to refer to a general reduction in sound tolerance. Many sounds have been reported to be associated with hyperacusis, including screams, whistles, thunder, and rattling of dishes, although adverse responses to less obtrusive noises such as the television, the telephone, and the car have been noted (Anari et al, 1999; Coelho et al, 2007a). Children complaining of hyperacusis can have lower loudness discomfort levels (LDLs) than those who do not complain, although this is not always the case (Anari et al, 1999; Coelho et al, 2007a), and the test-retest and inter-tester reliability of LDL measurements has been called into question (Baguley et al, 2013a). Care should be taken not to confuse hyperacusis with recruitment, which describes the narrowing of the auditory dynamic range due to poorer hearing thresholds, the result of outer hair cell damage (Moore, 2007). One differentiator is that whilst recruitment is not adversely modulated by emotional states such as fear and anxiety, hyperacusis commonly is (Baguley et al, 2013a).

Although the precise and presumably multiple mechanisms behind hyperacusis are not yet fully understood, increased gain of the central auditory system is thought to occur (Jastreboff & Hazell, 1993; Baguley, 2003; Eggermont, 2012); it has been

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*(Received 12 January 2015; revised 9 July 2015; accepted 2 September 2015)*

ISSN 1499-2027 print/ISSN 1708-8186 online 
DOI: 10.3109/14992027.2015.1092055

**Abbreviations**

| | |
|---|---|
| ALSPAC | Avon longitudinal study of parents and children |
| ASD | Autistic spectrum disorder |
| CI | Confidence interval |
| LDL | Loudness discomfort level |
| MOC | Medial olivary complex |
| OAE | Otoacoustic emissions |
| OR | Odds ratio |
| SCBU | Special care baby unit |
| SES | Socioeconomic status |

proposed that the neurotransmitter serotonin (5HT) might mediate this (Marriage & Barnes, 1995; Sattar, 2009), based upon the co-incidence of hyperacusis in serotonergic disorders such as depression and migraine. Increased central gain may be associated with reduced peripheral auditory sensitivity (Sun et al, 2011). Mild hearing loss therefore appears to be a risk factor for hyperacusis, a finding which is supported by animal models (Sun et al, 2011). This 'increased gain theory' is also thought to be the mechanism behind some cases of tinnitus and would therefore explain why tinnitus and hyperacusis are often comorbid (Jastreboff & Hazell, 1993; Anari et al, 1999; Coelho et al, 2007a; Baguley et al, 2013b). Another peripheral cause of hyperacusis not related to hearing sensitivity is facial nerve paralysis (e.g. Bell's palsy) where the stapedial reflex can be absent or of reduced amplitude (Katzenell & Segal, 2001).

In children, hyperacusis has been found to be associated with childhood developmental disorders. A high incidence of hyperacusis has been reported in persons with Williams syndrome (Levitin et al, 2005); high frequency hearing loss and recurrent OME are common in these children, and acoustic reflexes have been found to be deficient (Klein et al, 1990; Gothelf et al, 2006). Decreased sound tolerance is also a common feature of autistic spectrum disorder (ASD) (Rosenhall et al, 1999; Gomes et al, 2004; Khalfa et al, 2001) although empirical evidence is sparse. The suggested likely pathophysiology involves central pathways such as the limbic system, rather than the peripheral auditory system (Gomes et al, 2004). Oversensitivity to light and touch can also be common in these children (Gomes et al, 2004). Other non-auditory conditions which can be associated with hyperacusis include migraine, depression, Addison's disease, and minor closed head injury (Anari et al, 1999; Katzenell & Segal, 2001).

For some children, hyperacusis can be extremely distressing and can have a significant effect on behaviour. Children may complain that their ears hurt or may cover their ears with their hands when exposed to certain sounds (Gomes et al, 2004; Sattar, 2009). Avoidant behaviour may ensue and social interactions and daily activities can be affected (Klein et al, 1990; Aazh et al, 2011). Psychiatric comorbidities, particularly anxiety disorders, have been found to be more prevalent in hyperacusic adults (Juris et al, 2013). Hyperacusis has also been postulated as a precursor to tinnitus (Jastreboff & Hazell, 1993). It is therefore important that hyperacusis is given due attention, particularly in children where early intervention might prevent these sequelae.

The prevalence of hyperacusis in adults has been reported as between 8% and 23% (Rubinstein et al, 1996; Fabinjinska et al, 1999; Andersson et al, 2002). However, less is known about prevalence of hyperacusis in children. Coelho et al (2007a), using a sample of 506 children aged 5–12 years from one town in Brazil, reported the prevalence of hyperacusis (defined as 'lowered loudness discomfort levels with an abnormal annoyance to sounds') as 3.2%, and the prevalence of phonophobia as 9%. This is the only epidemiological study of hyperacusis in children that the authors are aware of; however, the limited geographical location (one town) and relatively small sample size calls into question the generalizability of these findings.

Other smaller-scale studies provide further limited information about the presentation of hyperacusis in children. In a sample of 100 normally-hearing children taken from an undefined clinical situation, Sattar (2009) found hyperacusis to be more common in males than females in a 2:1 ratio, and also found the most common age of presentation to be 3–4 years. Baguley et al (2013b) reviewed cases of young persons presenting with tinnitus to four expert European centres ($n = 88$): 39% were reported to have associated decreased sound tolerance.

The aim of the present study is to estimate prevalence of hyperacusis in 11-year-old children nested within a large UK population-based birth cohort study of child development. A secondary aim is to identify any early life and auditory risk factors for hyperacusis in this age group.

## Methods

*Avon longitudinal study of parents and children (ALSPAC)*

ALSPAC is a large prospective cohort study based in the UK. 14 541 pregnant women in the Avon area of the UK with expected delivery dates of 1 April 1991 to 31 December 1992 were recruited into the study. Further details about ALSPAC are given in Golding et al (2001) and Boyd et al (2013). The study website contains details of all the data that is available through a fully searchable data dictionary (www.bris.ac.uk/alspac/researchers/data-access/data-dictionary/).

At age 11, children attended a half day hands-on research clinic at the University of Bristol. Children attended a number of different sessions including physical measurements, psychological assessments, vision and hearing tests. A total of 7097 children attended the hearing test session, which formed our study sample. Ethical approval for the study was obtained from the ALSPAC Law and Ethics Committee and the Local Research Ethics Committees.

*Hyperacusis, tinnitus, and auditory measures*

Children attended a hearing session accompanied by their parents. A hyperacusis and tinnitus interview was carried out within this session, tailored to the level and understanding of the child. For the purposes of this study, hyperacusis was defined as over-sensitivity or distress to particular everyday sounds rather than to loud sounds. Specifically the child was asked whether they 'ever experience over-sensitivity or distress to particular sounds?' Children that answered yes to this question were further questioned on whether they stay away from places or activities because of sensitivity to sounds. Further contingent questions were asked about over-sensitivity to light/colours, touch, pain, smell or taste. The tinnitus methods are described by Humphriss et al (2015).

Air conduction hearing thresholds were measured at 0.5–8 kHz and bone conduction thresholds at 0.5–2 kHz using a GSI 61 audiometer in a sound treated booth. Middle-ear function was measured using a GSI 38 tympanometer. Tympanograms were classified using the Fiellau-Nikolajsen's modification of Jerger's

classification (Fiellau-Nikolajsen, 1983); Type A: peak at −100 to +100 daPa; Type C1: peak at −101 to –200 daPa; Type C2: peak at −201 to –300 daPa; Type B: flat trace, no middle-ear pressure recorded. The presence of grommets, perforations, or abnormal traces was also noted. On completion of tympanometry the presence of ipsilaterally evoked acoustic reflexes were measured at 85, 95, and 105 dB. The left ear was tested first and the lowest level sound at which the reflex was evoked was recorded. If no reflex was present it was recorded as absent.

Transient otoacoustic emissions (OAE) were measured using the Otodynamics ILO92 system. Click stimuli were presented at gain settings of −10.5 and −19.5 dB (reference 80 dB SPL) in the linear mode. OAE waveforms were extracted and analysed (see Hall et al, 2012 for detailed methods). OAE amplitude of the broadband response and at 1, 2, 3, and 4 kHz were analysed.

*Risk factors*

Neonatal risk factors, known to be associated with hearing outcomes in children were examined included gestational age (<37 weeks, ≥37 weeks gestation), birthweight (≤2500 g, >2500 g), child gender, admission to SCBU, special care baby unit (yes, no), and readmission to hospital in first four weeks (yes or never left, no). Information about neonatal factors was available from medical records. Diagnoses of autistic spectrum disorder (ASD) were available through health and education records (Williams et al, 2008).

Socioeconomic factors were available from parental questionnaires collected during pregnancy or at delivery, and those examined were highest maternal education level (low/minimal or vocational level, medium/qualifications obtained at16 years, high/qualifications obtained at ≥18 years), housing tenure (mortgaged/owned, private rented/other, council/housing association) and parental occupational social class based on the Registrar General's Classification (I professional, II intermediate, IIIN skilled non-manual, IIIM skilled manual/IV semi-skilled manual/V unskilled manual), using the highest of either parent.

*Statistical analysis*

Statistical analyses were carried out using STATA version 11.1. Associations between presence/absence of hyperacusis (outcome variable) and the range of categorical and continuous variables were examined using univariate and multivariate logistic regression, adjusting for the confounding variables specified above (and listed in Table 1).

## Results

A total of 7097 children attended the hearing session at age 11 years, described in Table 1. Of the 7097 children attending the hearing session, 7093 children answered the question about sound tolerance. A total of 261 (3.68%, 95% CI 3.25, 4.14) reported that they were over-sensitive to sound, including one set of twins; 157 (60.2%) male and 104 (39.8%) female. Of those, 112 (42.91%) reported behavioural avoidance of places or activities because of their sensitivity to sound, and 21 (8.04%) used ear protection for some sounds.

A small proportion of the children reporting hyperacusis also reported other sensory sensitivities, the most common of these being sensitivity to light and/or colours (Table 2).

**Table 1.** Characteristics of the study sample.

| | | *Study sample* n *(%)* *(n = 7097)* |
|---|---|---|
| Child factors | Sex | |
| | Male | 3485 (49.11%) |
| | Female | 3612 (50.89%) |
| | Missing | 0 |
| | Birthweight | |
| | ≤2500 g | 329 (4.64) |
| | >2500 g | 6323 (89.09) |
| | Missing | 445 (6.27) |
| | Gestational age | |
| | <37 weeks | 360 (5.07) |
| | ≥37 weeks | 6375 (89.83) |
| | Missing | 362 (5.10) |
| | Admission to SCBU | |
| | Yes | 425 (6.60%) |
| | No | 6015 (93.40%) |
| | Missing | |
| | Readmitted to hospital/ still in at four weeks | 6106 (86.04) |
| | No | 320 (4.51) |
| | Yes/never left | 671 (9.45) |
| | Missing | |
| Socioeconomic factors | Highest maternal education level | |
| | Low | 1421 (20.02%) |
| | Medium | 2297 (32.37%) |
| | High | 2768 (42.68%) |
| | Missing | 611 (8.61%) |
| | Housing tenure | |
| | Council/housing association | 510 (7.19%) |
| | Private rented/other | 604 (8.51%) |
| | Mortgaged/owned | 5403 (76.13%) |
| | Missing | 580 (8.17%) |
| | Parental social class | |
| | I | 962 (13.56%) |
| | II | 2776 (39.12%) |
| | III non-manual | 1562 (22.01%) |
| | III manual/IV/V | 892 (12.57%) |
| | Missing | 905 (12.75%) |

**Table 2.** Proportion of children with hyperacusis ($n = 261$) who reported other sensory sensitivities.

| *Sensory sensitivity* | *N (% of those with hyperacusis)* |
|---|---|
| Light/colours | 22 (8.42) |
| Touch | 17 (6.51) |
| Smell | 14 (5.36) |
| Taste | 10 (3.83) |
| Pain | 9 (3.45) |

Data on presence/absence of tinnitus were available for 260 of the 261 children with hyperacusis. A total of 109 (41.92%) of the children reporting hyperacusis also reported *any* tinnitus, a strong association (unadjusted odds ratio (OR) 1.89 [95% CI 1.47, 2.43], $p < 0.001$; adjusted OR 1.88 [95% CI 1.43, 2.48], $p < 0.001$).

Reporting of hyperacusis was more likely in children from families with a higher level of maternal education, in boys, and in those children still in hospital/readmitted to hospital during the first four weeks of life (Table 3). No strong associations were found with any of the other risk factors examined.

**Table 3.** Associations of child and socioeconomic factors with hyperacusis (OR for hyperacusis).

| | *N with hyperacusis (%)* | *N without hyperacusis (%)* | *Unadjusted OR [95% CI]* | *P* | *Adjusted OR* [95% CI] N = 5764* | *P* |
|---|---|---|---|---|---|---|
| Child factors | | | | | | |
| Sex | | | | | | |
| Male | 157 (60.15) | 3325 (48.67) | Ref | | Ref | |
| Female | 104 (39.85) | 3507 (51.33) | 0.62 [0.48, 0.80] | <0.001 | 0.64 [0.49, 0.85] | 0.002 |
| Birthweight | | | | | | |
| >25000g | 229 (94.24) | 6091 (95.10) | Ref | | Ref | |
| ≤2500g | 14 (5.76) | 314 (4.90) | 1.18 [0.68, 2.05] | 0.544 | 1.25 [0.59, 2.66] | 0.546 |
| Gestational age | | | | | | |
| ≥37 weeks | 230 (94.26) | 6141 (94.67) | Ref | | Ref | |
| <37 weeks | 14 (5.74) | 346 (5.33) | 1.08 [0.62, 1.87] | 0.783 | 0.99 [0.48, 2.04] | 0.988 |
| Admission to SCBU | | | | | | |
| No | 214 (92.24) | 5799 (93.47) | Ref | | Ref | |
| Yes | 18 (7.76) | 405 (6.53) | 0.83 [0.50, 1.35] | 0.458 | 0.99 [0.53, 1.84] | 0.977 |
| Readmitted to hospital/still in hospital at four weeks | | | | | | |
| No | 213 (91.03) | 5889 (95.17) | Ref | | Ref | |
| Yes | 21 (8.97) | 299 (4.83) | 1.94 [1.22, 3.08] | 0.005 | 1.98 [1.20, 3.25] | 0.007 |
| Socioeconomic factors | | | | | | |
| Highest maternal education level | | | | | | |
| Low | 39 (16.25) | 1381 (22.12) | Ref | | Ref | |
| Medium | 79 (32.92) | 2217 (35.52) | 1.26 [0.85, 1.86] | 0.242 | 1.52 [0.97, 2.40] | 0.066 |
| High | 122 (50.83) | 2644 (42.36) | 1.63 [1.13, 2.35] | 0.009 | 1.72 [1.08, 2.72] | 0.020 |
| Housing tenure | | | | | | |
| Mortgaged/owned | 205 (84.71) | 5195 (82.84) | Ref | | Ref | |
| Private rented/other | 20 (8.26) | 583 (9.30) | 0.86 [0.54, 1.38] | 0.557 | 0.90 [0.47, 1.73] | 0.770 |
| Council/housing association | 17 (7.02) | 493 (7.86) | 0.87 [0.52, 1.44] | 0.599 | 0.93 [0.54, 1.60] | 0.817 |
| Parental social class | | | | | | |
| I (professional) | 38 (16.67) | 924 (15.50) | Ref | | Ref | |
| II (intermediate) | 122 (53.51) | 2652 (44.50) | 1.11 [0.77, 1.62] | 0.555 | 1.19 [0.81, 1.74] | 0.360 |
| III (skilled non-manual) | 46 (20.18) | 1515 (25.42) | 0.73 [0.47, 1.14] | 0.174 | 0.88 [0.54, 1.42] | 0.612 |
| IV (type III manual, IV semiskilled manual, V unskilled manual) | 22 (9.65) | 869 (14.58) | 0.61 [0.36, 1.04] | 0.075 | 0.72 [0.39, 1.33] | 0.299 |

*Adjusted for maternal education, housing tenure, parental social class, sex of child, birthweight, gestational age, admission to SCBU and readmission to hospital.



In addition, 29 children with ASD attended the age 11 clinic. Twelve of these 29 children reported hyperacusis (41.37%); after adjustment for gender, the OR of autism predicting hyperacusis was 17.32 [95% CI 8.14, 36.88; $p < 0.001$]. The small sample size means that these results should be interpreted with caution.

Children with hyperacusis were not more likely to have middle-ear effusion or raised acoustic reflex thresholds at the sample point (Supplementary Tables S1 and S2, available in the online version of the journal). No strong associations between hyperacusis and hearing thresholds at 11 years were found (Supplementary Table S3, available in the online version of the journal).

There were associations between both left and right ear OAE amplitude (to low stimulus levels) and hyperacusis report (Table 4). Children with hyperacusis had increased odds of having larger OAEs; the associations were strongest and effect sizes were largest for OAEs generated at lower intensity stimulus levels. These associations were only apparent after statistical adjustment; the higher prevalence of hyperacusis in boys and the higher OAE amplitude in girls is likely to have been the reason for this difference not being observed in the unadjusted analyses. The largest differences in OAE amplitude were observed at 2 kHz: adjusted mean differences at this frequency for low level stimuli were 2.69 dB (95% CI 1.39, 3.98) on the right ear and 2.32 dB (95% CI 1.03, 3.60) on the left.

## Discussion

### *Prevalence*

We have estimated the prevalence of hyperacusis in 11-year-old UK children to be 3.68%. This value is similar to that of Coelho et al (2007a) who with a much smaller Brazilian sample of children aged 5–12 years, and with a definition of hyperacusis which included loudness discomfort levels, estimated the prevalence of hyperacusis to be 3.2%. ALSPAC is unique in being a much larger population-based study: we are not aware of any other comparable epidemiological studies that have looked at hyperacusis in children.

The present study, which was part of a larger study of child development, utilized a single question about sound tolerance embedded within a number of other questions about hearing. As such the data does not enable analysis to differentiate between hyperacusis, phonophonia, and misophonia. Similarly, it is not possible to determine which children reported reduced sound tolerance as a component of other medical or syndromic conditions, or in isolation. Future research is needed in this area.

### *Behavioural correlates*

A substantial proportion (42.9%) of the children with hyperacusis were found to show avoidance behaviours. Such behaviours are

**Table 4.** Associations of otoacoustic emission amplitude with hyperacusis (OR for hyperacusis per 1 SD increase in OAE amplitude).

| *Ear* | *Stimulus level* | *Frequency (kHz)* | *N with hyper-acusis* | *N without hyper-acusis* | *Unadjusted OR [95% CI]* | *P* | *N with hyper-acusis* | *N without hyper-acusis* | *Adjusted OR* [95% CI]* | *P* |
|---|---|---|---|---|---|---|---|---|---|---|
| Right | High | Broadband | 175 | 4461 | 1.06 [0.91, 1.24] | 0.418 | 144 | 3544 | 1.23 [1.02, 1.48] | 0.023 |
| | | 1 | 175 | 4461 | 1.06 [0.90, 1.23] | 0.453 | 144 | 3544 | 1.19 [0.99, 1.43] | 0.063 |
| | | 2 | 175 | 4461 | 1.03 [0.88, 1.20] | 0.680 | 144 | 3544 | 1.23 [1.03, 1.48] | 0.021 |
| | | 3 | 175 | 4461 | 0.97 [0.84, 1.13] | 0.788 | 144 | 3544 | 1.12 [0.94, 1.34] | 0.196 |
| | | 4 | 175 | 4461 | 0.94 [0.81, 1.10] | 0.498 | 144 | 3544 | 1.03 [0.86, 1.22] | 0.713 |
| Left | | Broadband | 187 | 4735 | 1.01 [0.87, 1.17] | 0.852 | 155 | 3766 | 1.18 [0.99, 1.40] | 0.055 |
| | | 1 | 187 | 4735 | 0.98 [0.85, 1.14] | 0.847 | 155 | 3766 | 1.10 [0.93, 1.31] | 0.240 |
| | | 2 | 187 | 4735 | 1.03 [0.89, 1.20] | 0.607 | 155 | 3766 | 1.21 [1.02, 1.44] | 0.028 |
| | | 3 | 187 | 4735 | 0.99 [0.85, 1.14] | 0.923 | 155 | 3766 | 1.12 [0.95, 1.33] | 0.165 |
| | | 4 | 187 | 4735 | 1.01 [0.87, 1.17] | 0.837 | 155 | 3766 | 1.14 [0.97, 1.35] | 0.106 |
| Right | Low | Broadband | 139 | 3914 | 1.19 [1.00, 1.42] | 0.039 | 114 | 3140 | 1.46 [1.18, 1.79] | <0.001 |
| | | 1 | 139 | 3914 | 1.14 [0.96, 1.36] | 0.118 | 114 | 3140 | 1.32 [1.08, 1.62] | 0.006 |
| | | 2 | 139 | 3914 | 1.21 [1.01, 1.44] | 0.031 | 114 | 3140 | 1.53 [1.24, 1.89] | <0.001 |
| | | 3 | 139 | 3914 | 1.12 [0.95, 1.33] | 0.165 | 114 | 3140 | 1.33 [1.08, 1.63] | 0.006 |
| | | 4 | 139 | 3914 | 1.04 [0.88, 1.23] | 0.613 | 114 | 3140 | 1.20 [0.98, 1.46] | 0.068 |
| Left | | Broadband | 145 | 4064 | 1.13 [0.96, 1.34] | 0.133 | 120 | 3259 | 1.34 [1.11, 1.63] | 0.003 |
| | | 1 | 145 | 4064 | 1.14 [0.96, 1.35] | 0.113 | 120 | 3259 | 1.31 [1.08, 1.59] | 0.005 |
| | | 2 | 145 | 4064 | 1.17 [0.99, 1.39] | 0.055 | 120 | 3259 | 1.42 [1.17, 1.73] | <0.001 |
| | | 3 | 145 | 4064 | 1.04 [0.88, 1.23] | 0.568 | 120 | 3259 | 1.20 [0.99, 1.46] | 0.059 |
| | | 4 | 145 | 4064 | 1.03 [0.87, 1.22] | 0.687 | 120 | 3259 | 1.17 [0.96, 1.41] | 0.101 |

*Adjusted for maternal education, housing tenure, parental social class, sex of child, birthweight, gestational age, admission to SCBU, readmission to hospital and tympanometric status.



common amongst hyperacusic individuals and have previously been documented in both adults and children (Klein et al, 1990; Andersson et al, 2002). For many parents, ear protection might appear the logical attempt at amelioration as most are unlikely to be unaware of the potential for reduced auditory sensitivity to increase the gain of the central auditory system. However, in ALSPAC, only 8% of the children reporting sound sensitivity had used ear protection. The present study found that only a small proportion of the children with hyperacusis also had other sensory hypersensitivities (the most common of these being over-sensitivity to light and colours). Similar findings have been reported elsewhere with children with ASD (Gomes et al, 2004) and with hyperacusic adults (Andersson et al, 2002).

*Non-auditory risk factors*

Male gender, higher maternal education level, and poor neonatal health were found to be associated with a higher risk of hyperacusis. The male gender bias is consistent with Sattar (2009) who found a 2:1 male to female ratio in a clinic-based sample of children. However, it is at variance with Coelho et al (2007a), who with a smaller population-based sample from Brazil found hyperacusis to be slightly more common in girls (significance levels not reported). Further studies would be needed to determine whether the presentation of hyperacusis does vary between populations or whether this disparity is just a reflection of the differences in methodology between the two studies.

Hyperacusis was reported by around 40% of the children with ASD who attended the research clinic, showing it is a common condition in ASD consistent with other studies (Rosenhall et al, 1999; Gomes et al, 2004). However around two thirds of the children in ALSPAC with ASD diagnosed by age 11 did not attend the age 11 research clinic (Williams et al, 2008), and so the results should be interpreted with caution. This is also likely to have biased the population prevalence value, resulting in an underestimation.

Maternal education level was included in the analyses as a measure of socio-economic status (SES). Higher maternal education levels were associated with children reporting hyperacusis. However, neither of the other two measures of SES used (housing tenure, parental social class) were associated in this way. It is possible that the children of educated mothers were more articulate than the other children and were therefore better able to recognize and describe their symptoms. These children might also have had a greater awareness of health issues or had parents who were more attentive to their health needs. The authors are not aware of any other health conditions that are similarly socially patterned, most being associated with lower SES (including lower parental education levels).

Adverse early neonatal health was found to be associated with the children reporting hyperacusis. The most plausible causal explanation for this is that neonatal illness has a detrimental effect of on auditory neurodevelopment; the association between time in the Neonatal Intensive Care Unit (NICU) and hearing loss is well-established (Davis et al, 1997), although in this study we did not find a higher risk of hyperacusis in babies admitted to NICU at birth but rather in those still in hospital at four weeks or who had been readmitted. Readmission to hospital after discharge may be due to suspected infection, feeding problems, life threatening events, or jaundice, although in many cases there is no clear organic diagnosis (Oddie et al, 2005). Alternatively, it is possible that adverse experiences in the neonatal period might cause changes in cognitive development and predispose the child to later behavioural or emotional problems (Anand & Scalzo, 2000) which might contribute to any hyperacusis or phonophobia. A possible non-causal explanation is that parents of a child who has had a difficult

start in life might be hyper-vigilant with respect to their child's health with the result that their child is more likely to report minor health concerns too.

*Associations with auditory risk factors*

A strong association was found between hyperacusis and tinnitus in these children. Such associations have been well documented (Anari et al, 1999; Coelho et al, 2007a,b; Baguley et al, 2013b), the common mechanism of increased central gain being postulated (Jastreboff & Hazell, 1993). No associations were found between hyperacusis and hearing threshold level, in contrast to Coelho et al (2007a) who found minimum hearing loss (but not moderate/profound loss) in the left ear to be a risk factor for hyperacusis (although the wide confidence interval found by Coelho et al indicates a lack of precision in their results).

We did not find an association between middle-ear function at age 11 and report of hyperacusis. This is contrary to previous suggestions that OME may be a risk factor for decreased sound tolerance (Sun et al, 2011). We did not examine the early middle-ear history of the cohort, and so cannot rule out early otitis media with effusion as a contributory factor.

Of interest were the higher amplitude OAEs observed in children with hyperacusis in response to low, but not high level stimuli. The protocol for measuring OAE in this study used two different stimulus levels (both of which were lower than levels used for hearing screening purposes), based on the hypothesis that OAEs generated to lower stimulus levels are more sensitive to differences in cochlear function than those generated at higher levels (Lapsley Miller et al, 2004). OAE amplitude is influenced both by the conduction of sound through the external/middle ear and the activity of the outer hair cells in the cochlea; an effect related to the conduction of sound would not be expected to vary with stimulus level, and indeed there were no differences in middle-ear function observed between the groups. The larger differences in OAE amplitude measured at the lower but not the higher stimulus level suggest an active, physiological effect originating from the cochlea rather than the external or middle ear. Possible explanations could be the presence of more spontaneous OAEs in the hyperacusis group, which are known to be associated with larger amplitude transient evoked OAE (Moulin et al, 1993); however the amplitude differences would be expected for both high and low level stimulus levels. An alternative explanation could be increased gain of the cochlear amplifier resulting from abnormal efferent function, which has been proposed as a mechanism of tinnitus (Eggermont, 2012) and specifically in tinnitus and hyperacusis following closed head injury (Attias et al, 2005); the ipsilateral medial olivary complex (MOC) efferent system is stimulated by the sound used to generate the TEOAE (Guinan, 2006).

To our knowledge these findings have not been observed in any other study of hyperacusis and OAE. Studies of children with autism (in whom hyperacusis is highly prevalent) have shown either no difference in OAE amplitude when compared to a control group (Gravel et al, 2006), or have shown *lower* amplitude OAE (Katzenell & Segal, 2001). One explanation for the contradictory results of our study may be the different stimuli levels used as well as recording in the 'linear' mode, ensuring that both the linear and nonlinear components of the OAE are recorded. In studies of efferent function on OAE, it is not known how the MOC response is altered by use of the nonlinear rather than the linear mode to record the OAE (Guinan, 2006).

*Study limitations*

As discussed previously, the cases with hyperacusis in this study were identified through a single question as part of a general examination of hearing, which could have led to misclassification of cases with or without hyperacusis. This would result in a biased prevalence estimate if there was systematic misclassification of one group compared to the other. Children with ASD in ALSPAC were less likely to attend the research clinics than other children; given that children with ASD are more likely to report hyperacusis it is possible that the prevalence value of this study is an under estimate.

Our sample of children was socially advantaged compared to the remainder of the ALSPAC cohort and therefore to the UK population as a whole. Such a phenomenon is not uncommon in large-scale epidemiological studies where there will be a 'volunteer effect' (Delgado & Llorca, 2004). Given the association between maternal education and hyperacusis, it is possible that this socioeconomic bias might have affected our prevalence estimate: more educated mothers might have been more likely to bring their child to the assessment clinic resulting in an over-estimate of the prevalence of hyperacusis. This could have occurred despite the fact that the children attended the assessment clinic for a variety of health measures other than just those relating to auditory function and performance. It is, however, unlikely that this over-representation of socially advantaged families would have affected the associations that we describe.

This study was not able to examine early OME history of the children, or any family history of tinnitus or hyperacusis. Future studies could examine this.

The finding that the prevalence of hyperacusis at age 11 years is such that on average one child in each primary school class will experience this symptom has some implications. Specifically, teachers and teaching assistants could usefully be made aware of hyperacusis, the burden that places upon the child, and how classroom behaviours could be modified to accommodate this. One might hypothesize that these recommendations would also apply to children with hearing loss and tinnitus.

## Conclusions

We have estimated the prevalence of hyperacusis in 11-year-old children in the UK to be 3.68%, (95% CI 3.25, 4.14), with nearly half of these children exhibiting behavioural avoidance. Hyperacusis in this age group is therefore not uncommon: we would expect approximately one child out of a class of 30 to be troubled by this. Hyperacusis is more common in boys and about 42% of hyperacusic children will also experience tinnitus. Hearing loss does not appear to be a risk factor for hyperacusis but there is an association with increased otoacoustic emission amplitude which requires further investigation.

## Acknowledgements

We are extremely grateful to all the families who took part in this study, the midwives for their help in recruiting them, and the whole ALSPAC team which includes interviewers, computer and laboratory technicians, clerical workers, research scientists, volunteers, managers, receptionists, and nurses. The UK Medical Research Council and the Wellcome Trust (Grant ref: 092731) and the University of Bristol provide core support for ALSPAC. This publication is the work of the authors, and Amanda Hall will serve as guarantor for the contents of this paper.

***Declaration of interest:*** The authors declare no conflicts of interest.

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