

1 **Abstract**

2 **Objective:** There is currently no singularly accepted definition of hyperacusis. The aim of this
3 study was to determine a definition and description of hyperacusis by clinician consensus.

4 **Design:** A three-round Delphi survey involving hearing healthcare professionals built towards
5 clinical consensus on a definition of hyperacusis. Round 1 involved three open-ended questions
6 about hyperacusis. Seventy-nine statements were generated on descriptions, impact, sounds,
7 and potential features of hyperacusis. Agreement on the relevance of each statement to defining
8 or describing hyperacusis was then measured in Rounds 2 and 3. General consensus was
9 defined a priori as $\geq 70\%$ agreement, or ≥ 90 for clinical decision making.

10 **Study Sample:** Forty-five hearing healthcare professionals were recruited to take part in this
11 study. Forty-one completed Round 1, 36 completed Round 2, and 33 completed Round 3.

12 **Results:** Consensus was reached on 42/79 statements. From these a consensus definition
13 includes “A reduced tolerance to sound(s) that are perceived as normal to the majority of the
14 population or were perceived as normal to the person before their onset of hyperacusis”. A
15 consensus description of hyperacusis was also determined.

16 **Conclusions:** This consensus definition of hyperacusis will help to determine the scope of
17 clinical practice guidelines and influence needed research on hyperacusis.

18 **Key words:** Sound tolerance, sound sensitivity, uncomfortable loudness levels,
19 psychoacoustics/hearing science

20

21

22

23 **Introduction**

24 Hyperacusis, literally meaning excessive hearing, is a hearing disorder which affects how a
25 person perceives sound. It has been described in many different ways, including an “unusual
26 tolerance to ordinary environmental sounds” (Vernon, 1987), “an unusual hypersensitivity or
27 discomfort induced by sound” (Marriage and Barnes, 1995), and “an aversion to loud sounds”
28 (Baguley et al., 2013). It is often described in terms of altering the ‘tolerance’ or ‘sensitivity’
29 towards certain sounds (Auerbach et al., 2014, Phillips and Carr, 1998, Hébert et al., 2013,
30 Khalifa et al., 2004, Wagenaar et al., 2010).

31 Hyperacusis is sometimes conflated with other hearing disorders such as phonophobia or
32 misophonia. Phonophobia (originating from Greek words of ‘sound’ and ‘fear’) is an anxiety
33 disorder, which is characterised as being an unwarranted and persistent fear of sound
34 (Mathisen, 1969). Misophonia involves the experience of intense emotional reactions to sounds
35 (Taylor, 2017, Potgieter et al., 2019), and often presents with adverse reactions to specific
36 patterns of sounds, or sounds presented in certain situations. Thus, although separate
37 conditions, misophonia, phonophobia, and hyperacusis are not considered to be mutually
38 exclusive. Indeed, Jastreboff and Jastreboff (2014) used decreased sound tolerance (DST) as
39 an umbrella term to describe multiple disorders that affect the perception of sound, including
40 hyperacusis and misophonia. They described DST as being present when a person displays
41 negative reactions to sounds that would not cause such reaction in the average listener.
42 Jastreboff and Jastreboff (2014) categorised phonophobia as a subtype of misophonia, rather
43 than an independent condition. Tyler et al. (2014) also suggested that phonophobia and
44 misophonia were different characteristics of hyperacusis, having proposed that hyperacusis
45 should be subcategorised as loudness, annoyance (also considered as misophonia), fear (also
46 considered as phonophobia) and pain hyperacusis.

47 The relationship between loudness recruitment and hyperacusis is also unclear (Marriage and
48 Barnes, 1995). Loudness recruitment involves an abnormally rapid growth in perceived
49 loudness, often associated with outer hair cell (OHC) loss (Moore, 1998). Recruitment is a
50 phenomenon that occurs in people with hearing loss, in which low magnitude sounds cannot
51 be heard, but a small growth in stimulus intensity can be perceived as a large growth by the
52 listener. This is thought to be a separate phenomenon to hyperacusis, although it has been
53 reported that 59.1% of people with hyperacusis have some form of hearing impairment (Paulin
54 et al., 2016), many in the high frequency range (Sheldrake et al., 2015), suggesting OHC loss.
55 Recruitment and hyperacusis can take place at the same time (Baguley, 2003). Furthermore,
56 people with hearing loss can have hyperacusis without having recruitment issues (Sheldrake et
57 al., 2015).

58 Hyperacusis is frequently found comorbid with tinnitus (the perception of sound in the absence
59 of any external sound source; Levine and Oron, 2015). As many as 86% of people with
60 hyperacusis also report tinnitus (Anari et al., 1999, Sheldrake et al., 2015), and around 40% of
61 people who present with tinnitus as a primary complaint also report hyperacusis (Jastreboff and
62 Jastreboff, 2000). Indeed, hyperacusis has been described as a pre-tinnitus state (Jastreboff and
63 Hazell, 1993, Guimarães et al., 2014), thought to be caused by a central gain within auditory
64 pathways. Often, tinnitus and hyperacusis are treated within the same clinic or service, with
65 attention focussed primarily on tinnitus. Some studies have even pointed to aetiological
66 connections between tinnitus and hyperacusis, such as hyperactivity within the auditory
67 network (Chen et al., 2015) and loss of hearing threshold sensitivity (though not necessary for
68 onset; Dauman and Bouscau-Faure, 2005). There are also many other medical conditions that
69 show comorbidity with hyperacusis, some of which also present with increased sensitivity of
70 other senses including vision (reaction to light) and touch. Such conditions include Williams
71 syndrome (Klein et al., 1990, Gothelf et al., 2006), autism spectrum disorder (Danesh et al.,

72 2015), depression (Paulin et al., 2016), chronic fatigue syndrome, Lyme disease, Meniere's
73 disease and posttraumatic stress disorder (Goodson and Hull, 2015).

74 Hyperacusis, by its various descriptions, is thought to affect between 3.2 and 17.2% of the
75 population (Hannula et al., 2011, Coelho et al., 2007, Andersson et al., 2002, Fabijanska et al.,
76 1999). It can present itself in many ways, with common symptoms including headache,
77 discomfort, anxiety, and fatigue. Some people with hyperacusis report pain upon hearing
78 certain sounds. Hyperacusis can be extremely debilitating, causing people to avoid social
79 situations, heavily impacting on a person's quality of life. People with hyperacusis often wear
80 hearing protectors, such as ear plugs or headphones, in an attempt to protect themselves from
81 noises that cause discomfort (Blaesing and Kroener-Herwig, 2012, Jüris et al., 2014, Paulin et
82 al., 2016). Although this may seem intuitive to a person experiencing hyperacusis, prolonged
83 use of ear protection can cause the auditory system to become even more sensitive to noise,
84 thereby intensifying symptoms (Formby et al., 2003, Munro et al., 2014). Avoiding sound
85 sources can also lead to the same phenomenon (Baguley, 2003).

86 A range of different tools are currently used to assess hyperacusis. Multi-item questionnaires
87 include the hyperacusis questionnaire (HQ), which quantifies and evaluates different features
88 of hyperacusis (Khalifa et al., 2002), the Multiple-Activity Scale for Hyperacusis (MASH),
89 which rates the impact certain activities have on everyday life (Dauman and Bouscau-Faure,
90 2005), the German Questionnaire on Hypersensitivity to Sound (Geräuschüberempfindlichkeit;
91 GÜF), which assesses the subjective distress in patients with hypersensitive hearing (Blasing
92 et al., 2010, Nelting et al., 2002) and the Inventory of Hyperacusis Symptoms (IHS), which
93 assesses severity of subjective hyperacusis impact (Greenberg and Carlos, 2018). Another tool
94 is the Uncomfortable Loudness Levels (ULLs) test, which is used to determine the loudness
95 level that becomes uncomfortable for the listener at different pure tone frequencies. A key issue
96 with this measurement, despite some research on the topic (e.g. Aazh et al., 2018, Aazh and

97 Moore, 2017), is that there are no officially recommended cut-off levels that enable diagnosis
98 of hyperacusis. In typically hearing individuals with no known loudness tolerance problem,
99 ULLs vary from 86-98 dbHL (Knobel and Sanchez, 2006). In people reporting hyperacusis,
100 ULLs have been reported that range from 69.3 dB HL to 76dB HL (Juris et al., 2013). There is
101 mixed evidence for a relationship between HQ scores and ULLs (Meeus et al., 2010, Aazh and
102 Moore, 2017).

103 In terms of hyperacusis management, there is little in the literature (Fackrell et al., 2017), and
104 many unanswered questions. Recently, a set of 28 research priorities were defined using the
105 James Lind Alliance Priority Setting Partnership method (Fackrell et al., 2019a, Fackrell et al.,
106 2019b). As well as priorities relating to the physiology and treatment of hyperacusis, many
107 questions related to diagnostic criteria, and how to distinguish hyperacusis from other hearing-
108 related conditions.

109 The aim of this study was to determine a clinician consensus definition of hyperacusis, to
110 inform diagnosis and the scope of future clinical practice guidelines.

111 **Methods**

112 This study followed a standard Delphi methodology. The Delphi method is an iterative process
113 that seeks anonymous judgements and controlled feedback from experts until a consensus is
114 reached (Linstone and Turoff, 1975). A three round Delphi survey sought clinical consensus
115 on a definition and further description of hyperacusis, in an expert panel of hearing healthcare
116 professionals. The Delphi review constituted a service evaluation so ethical review board
117 approval was not required.

118 **Recruitment of expert panel**

119 Invitation emails were sent to a range of hearing healthcare clinicians with expertise in
120 hyperacusis within the UK. Emails were sent initially to those who had previously participated

121 in a hyperacusis prioritisation exercise (Fackrell et al., 2019) and expressed a wish to
122 participate in further hyperacusis-related activities. Some clinicians who agreed to take part in
123 the study also shared an invitation with appropriate colleagues within their networks. The
124 invitation email contained a description of the study aims and Delphi process, and an
125 approximate timeline for participation. Clinicians were eligible to take part if they (1) had
126 experience of managing adults and/or children with hyperacusis, and (2) were willing to share
127 their expert opinions in a three-part Delphi survey about hyperacusis. Initially 45 hearing
128 healthcare professionals agreed to take part. Only participants who completed Round 1 were
129 eligible to participate in Round 2, and only participants completing Round 2 were eligible to
130 participate in Round 3.

131 **Delphi survey**

132 The online Delphi survey was designed to include three rounds. Round 1 consisted of open-
133 ended questions on hyperacusis. Rounds 2 and 3 contained a series of statements which
134 members of the panel were asked to evaluate. All three rounds of the Delphi survey were
135 designed using Online Surveys (<https://www.onlinesurveys.ac.uk/>) and distributed electronically
136 with personalised links via email. Each member of the expert panel was automatically assigned
137 a randomly generated participant identification number, and all responses were anonymised.

138 *Design of Round 1 Open-Ended Questionnaire*

139 The first round consisted of three open-ended questions designed to broadly extract what
140 participants considered did and did not constitute hyperacusis. Questions were: (1) What is
141 your current understanding/definition of hyperacusis?; (2) What do you observe to be the main
142 presenting features of hyperacusis?; and (3) What characteristics or conditions do you think
143 are commonly mistaken for hyperacusis but are not?. There was no limit on the length of
144 responses.

145 Additional demographic information collected included: job role, whether working in the
146 public/private sector, years in current profession, and an approximate number of hyperacusis
147 patients seen in the previous 3 months.

148 *Design of Round 2 Closed Questionnaire*

149 The responses provided by panellists in Round 1 were organised into statements and then
150 grouped into themes using a thematic analysis approach (Boyatzis, 1998). Analysis was carried
151 out independently by two authors who then agreed a final dataset. The protocol of thematic
152 analysis was predefined in five steps.

- 153 1. Familiarisation process. Each author immersed themselves with answers to all
154 questions, allowing themselves to become acquainted with each response.
- 155 2. The author began to look for recurring themes or ideas within responses to each
156 individual question.
- 157 3. The author began to generate codes which identify a feature of the original response,
158 taking the most meaningful element(s).
- 159 4. Codes that were considered to be equivalent were grouped together into themes. After
160 this was completed, authors joined to discuss and agree the codes and themes.
- 161 5. All themes and codes were reviewed, and a consensus was reached by all authors on
162 each theme.

163 Eight themes were identified and formed sections of the Round 2 questionnaire. The first two
164 themes were ‘tolerance’ and ‘sensitivity’ and included statements related to how hyperacusis
165 is defined. Under these themes, participants were asked to consider whether each statement
166 provided the best (score 1) to worst (score 4) description of hyperacusis.

167 The next three themes included general descriptions of hyperacusis, the impact of hyperacusis,
168 and the sounds involved in hyperacusis. Participants were asked to rate the extent to which they

169 agreed with each statement with the response options: 1 = Always; 2, = Almost Always; 3 =
170 Sometimes; 4 = Almost Never; 5 = Never, or 6 = Not sure.

171 The remaining themes provided a list of potential features of hyperacusis; conditions, emotions,
172 and ‘other’ features that may relate to hyperacusis. Participants were asked to indicate whether
173 they considered each feature a (1) defining feature of hyperacusis; (2) common feature of
174 hyperacusis; (3) occasional feature of hyperacusis; (4) not a feature of hyperacusis; or (5) not
175 sure. Participants had the option to provide any comments at the end of the questionnaire. Some
176 conditions in this round could be considered as comorbid with hyperacusis. Participants were
177 reminded to rate these as features of hyperacusis, and not as comorbidities.

178 *Design of Round 3 Closed Questionnaire*

179 Round 3 consisted of the same list of closed statements used in Round 2. For each statement,
180 summary results (percentages of panellists who chose each response option) from Round 2
181 were reported back to the panellists. In Round 3 participants were given the opportunity to
182 revise their responses, and to provide comments at the end of the questionnaire.

183 *Interpretation/Consolidation and Final Consensus*

184 To generate percentage agreement, the number of participants responding ‘Always’ and
185 ‘Almost Always’ on each statement was summed, as were the number of respondents
186 answering ‘Never’ and ‘Almost Never’. This gave three categories: ‘Always’, ‘Sometimes’
187 and ‘Never’. For this Delphi, the general consensus level was set a priori to be 70% agreement,
188 i.e. 70% or more respondents answered within the same category. However, for the purpose of
189 a consensus definition of hyperacusis that can inform clinical decision making, the more
190 stringent agreement level of 90% for statements was required.

191

192 **Results**

193 The Delphi survey was conducted between January 2019 and April 2019. In Round 1, 41/45
194 participants completed the survey (91.1% response rate). Demographics of those who
195 completed Round 1 are given in Table 1. The response rate in Round 2 was 36/41 (87.8%), and
196 in Round 3 was 33/36 (91.7%).

197

198 **Levels of agreement**

199 Statements that reached the highest level of agreement to be the best descriptors of hyperacusis
200 were ‘*A reduced tolerance to sound(s)*’ (90.9%) and ‘*An increased sensitivity to sound(s)*’
201 (78.8%). Many statements that could either ‘Always’ or ‘Sometimes’ describe hyperacusis
202 reached consensus. There was 81.8% agreement that ‘*Hyperacusis can always cause an*
203 *abnormal response to normal sound(s)*’. Eight further statements on which consensus was
204 reached were that hyperacusis can *sometimes*

- 205 • Impact a person’s relationship (93.9% agreement)
- 206 • Cause a person to use ear protection, such as ear plugs or earphones (97% agreement)
- 207 • Reduce a person’s confidence (93.9% agreement)
- 208 • Cause poor concentration (87.9% agreement)
- 209 • Cause a person to withdraw from social situations (81.9% agreement)
- 210 • Limit the daily activities of a person (84.8% agreement)
- 211 • Cause a person to avoid social situations (78.8% agreement)
- 212 • Disrupt daily functioning (78.8%)

213 ‘*Sounds perceived as normal to the majority of the population*’ and ‘*Sounds perceived as*
214 *normal to the person before their onset of hyperacusis*’ were both recognised as *always*

215 involved with hyperacusis, with agreement of 100% and 91% respectively. Sounds that were
216 voted to *sometimes* involve hyperacusis were ‘*moderate sounds*’ and ‘*annoying sounds*’, both
217 with an agreement of 90%.

218

219 Potential features of hyperacusis identified from Round 1 were categorised as conditions,
220 emotions, or other (Supplementary Material 1).

221 *Conditions:* No conditions reached consensus for being a *defining* or *common* feature of
222 hyperacusis. As an occasional feature of hyperacusis, ‘Superior canal dehiscence’ had 78.8%
223 agreement, and ‘Sensory disorder’ had 72.7% agreement.

224 *Emotions:* There was high levels of agreement that the following were *common* features of
225 hyperacusis: avoidance (90.9% agreement), withdrawal (93.9% agreement), annoyance (93.9%
226 agreement), sensitivity (90.9% agreement), intolerance (93.9% agreement), upset (81.8%
227 agreement), aversion (87.9% agreement), irritability (81.8% agreement), altered behaviour
228 (84.8% agreement), anxiety (87.9% agreement), high stress (81.8% agreement), distress
229 (87.9% agreement), fear (78.8% agreement), loss of confidence (75.8% agreement) and
230 frustration (78.8% agreement). Emotions considered *occasionally* presented in hyperacusis
231 were anger (81.8% agreement) and depression (81.8% agreement).

232 Although emotions avoidance, withdrawal, annoyance, sensitivity and intolerance reached a
233 90% consensus, they were voted as *common* and not *defining* features of hyperacusis. For this
234 reason, it was decided that they will be placed within the consensus-based description of
235 hyperacusis, rather than definition.

236 *Other:* Other features considered to be *common* presenting features in hyperacusis were
237 hypervigilant/hyperalert hearing (81.8% agreement), and discomfort towards sounds (75.8%
238 agreement). Headaches were as agreed by 75.8% of participants to be an *occasional* feature in

239 hyperacusis. Incorrect programming of hearing aids as well as a normal reaction to loud sounds
240 were both established as not being a feature of hyperacusis (87.9% agreement on both).

241 **A consensus-based definition and description of hyperacusis**

242 Four statements reached consensus (90% agreement level) to form a consensus definition of
243 hyperacusis suitable for clinical decision making. Based on these, hyperacusis was defined as

244 *A reduced tolerance to sound(s) that are perceived as normal to the majority of the*
245 *population or were perceived as normal to the person before their onset of hyperacusis,*
246 *where 'normal' refers to sounds that are generally well tolerated.*

247 A consensus-based (70% agreement level) description of hyperacusis is that it is an increased
248 sensitivity to sounds(s). Hyperacusis always involves an abnormal response to normal
249 sound(s), and some people with hyperacusis can also be affected by moderately loud or
250 annoying sounds. Hyperacusis can sometimes impact a person's relationship, reduce a person's
251 confidence, and cause them to withdraw from or avoid social situations, limiting their daily
252 activity and disrupting daily functioning. Poor concentration is also sometimes present with
253 hyperacusis. Hyperacusis can lead a person to use ear plugs, ear phones, or sound defenders in
254 an attempt to protect themselves from noise.

255 By consensus, avoidance, withdrawal, annoyance, sensitivity, intolerance, upset, aversion,
256 irritability, altered behaviour, anxiety, high stress, distress, fear, and frustration were
257 considered commonly present in people with hyperacusis. Depression and anger were
258 considered occasionally present. Other features considered common in hyperacusis were
259 hypervigilant/hyperalert hearing and discomfort towards sounds. Headaches were also
260 considered an occasional feature of hyperacusis.

261 **Discussion**

262 This study is the first to establish a clinical consensus-based definition and description of
263 hyperacusis. This can usefully inform diagnostics and the scope of clinical practice guidelines.
264 Practice guidelines are needed to ensure consistent clinical examination of patients with
265 suspected hyperacusis. This will also allow comparisons to be made across clinical populations.

266 Though this is the first study to establish a consensus-based definition of hyperacusis, previous
267 definitions/descriptions have been used within literature. For example, in a review on clinical
268 interventions for hyperacusis in adults, Fackrell et al. (2017) described hyperacusis as “...the
269 perception of everyday environmental sound as being overwhelmingly loud or intense”. They
270 also report terminology such as ‘reduced’, ‘decreased’, or ‘collapsed sound tolerance’ as being
271 used to describe hyperacusis. Our Delphi review provides consensus-based support for the use
272 of ‘reduced’ and ‘tolerance’ when describing hyperacusis, but not for ‘environmental sound
273 as being overwhelmingly loud or intense’. Tyler et al. (2014) described loudness hyperacusis
274 as “[...] moderately intense sounds are judged to be very loud compared with what a person
275 with normal hearing would perceive.”. This Delphi review has provided a consensus-based
276 definition of hyperacusis as one condition, whereas Tyler et al. (2014) proposed sub-categories
277 of hyperacusis, including loudness, annoyance, pain and fear hyperacusis. In this review, the
278 terms ‘annoyance’ and ‘fear’ reached consensus as common but not defining features of
279 hyperacusis. Furthermore, ‘pain’ did not reach a consensus as a feature of hyperacusis so was
280 not included within the definition.

281 Hyperacusis is a growing field of research with many unanswered research questions recently
282 identified. A clear accepted definition of hyperacusis is an essential step forward in hyperacusis
283 research, and will be valuable in working towards answering the 28 priority research questions
284 as determined by professionals and patients with hyperacusis (Fackrell et al., 2019a). One of
285 the top 10 priority questions asks “Are there different meaningful types of hyperacusis?”
286 Interestingly, categorising hyperacusis was not mentioned by the expert panel at any stage of

287 this Delphi review, as proposed by Tyler et al. (2014). This simple terminology may assist in
288 the diagnosis and management of patients; however, further research is needed before these
289 conditions can be formally classified as sub-types of hyperacusis, rather than separate
290 phenomena such as phonophobia or misophonia. However, research into the mechanisms
291 underlying different symptoms of hyperacusis are sparse, and more is needed to determine
292 whether there are indeed different subtypes and potentially different therapeutic targets.

293 Within the Delphi, the statement “Hyperacusis can *sometimes* be described as a reduction in
294 the Uncomfortable Loudness Levels (ULL) in Pure Tone Audiometry (PTA)” reached a 78.8%
295 agreement. Despite this, The British Society of Audiology recommended procedure for the
296 Determination of ULLs (BSA, 2011) recommended that ULLs should not be performed
297 routinely within clinical situations as it involves exposing patients to high levels of sounds,
298 which can be especially distressing to a patient with hyperacusis. It is also important to note
299 that the pure tones used in ULLs may not meaningfully represent the types of sound causing
300 distress in certain individuals (Fackrell and Hoare, 2019). Where not clinically indicated, the
301 British Society of Audiology (Hoare et al., 2019) recommend use of multi-item questionnaires
302 such as the HQ (Khalifa et al., 2002), the MASH (Dauman and Bouscau-Faure, 2005) and the
303 GUF (Nelting and Finlayson, 2004, Nelting et al., 2002) to assess for hyperacusis. This newly
304 formed definition and description of hyperacusis may inform the creation of a new assessment
305 scale for hyperacusis, which may help with the development of new practice guidance
306 development.

307 Interestingly, 57.8% of respondents in this Delphi review said that hyperacusis can be described
308 as an emotional reaction to sound(s), yet 66.7% of people agreed that misophonia was only an
309 occasional feature of hyperacusis. This highlights the potential ambiguity in questions relating
310 to conditions that are associated with hyperacusis, but not comorbid. Furthermore emotions
311 that reached consensus as being common features of hyperacusis included annoyance,

312 frustration, and irritability, which typically feature in misophonia (Potgieter et al., 2019), and
313 fear, avoidance, withdrawal, aversion, and anxiety, which typically feature in phonophobia
314 (Asha'ari et al., 2010) . In this Delphi, 66.7% of people also agreed that phonophobia was an
315 occasional feature of hyperacusis. Results from this Delphi suggest that phonophobia and
316 misophonia are indeed separate conditions to hyperacusis.

317 A limitation of this Delphi review is that a small number of experts felt that some questions
318 were slightly unclear or ambiguous, especially those in relation to conditions that may be
319 included as a feature of hyperacusis, but not as a comorbid condition. In completing Round 3
320 one participant noted *“I found some ambiguity in the questions relating to other conditions
321 (not including comorbidities) this time that I don't think occurred to me last time. I have
322 answered the Qs ‘as a feature of hyperacusis’ e.g. tinnitus: although hyperacusis is very often
323 present alongside tinnitus, tinnitus is not a feature of hyperacusis per se.”* Around 86% of
324 people with hyperacusis as a primary complaint also complain of tinnitus (Anari et al., 1999).
325 This shows that tinnitus is commonly comorbid with hyperacusis but does not necessarily mean
326 that tinnitus is a feature of hyperacusis, although studies have pointed to similar aetiologies
327 between the conditions, such as cochlear damage causing altered brain responses or central
328 gain (Knipper et al., 2013). It has also been demonstrated that people with tinnitus have
329 enhanced auditory sensitivity, compared to a non-tinnitus population (Hebert et al., 2013).
330 Consensus was not met for any conditions as a defining, common, or occasional features of
331 hyperacusis. They therefore were not included in the final definition or description.

332 One participant wrote *“In my experience, clinicians don’t often understand the differences
333 [between hyperacusis, phonophobia and misophonia], plus in online searches these terms are
334 often used interchangeably when in fact they are different from one another”*. This highlights
335 the need for a consensus derived definition of hyperacusis as well as clear clinical guidelines
336 outlining the difference between each condition involving sound tolerance. Another participant

337 stated that it was difficult to say whether items were ‘defining’ features of hyperacusis on their
338 own, saying that hyperacusis is a collection of symptoms rather than individual symptoms. The
339 consensus-based description should serve to highlight the many potential symptoms that
340 clinicians should be alert to in supporting people who have hyperacusis.

341 This Delphi process was limited to UK participants, to help inform diagnostics and the scope
342 of UK clinical practise guidelines. However, this means the consensus-based definition is also
343 limited to UK audiences.

344 **Conclusion:**

345 This study used the Delphi review process to find a consensus-based definition and description
346 of hyperacusis. A series of statements reached consensus and a definition of hyperacusis with
347 an accompanying description has been suggested. This is an essential starting point to
348 determine the scope of clinical practice guidelines, and will also support and fuel further
349 research into the mechanisms and management of hyperacusis.

350 **Acknowledgments**

351 Julie Brady, Julie Carr, David Baguley, Richard Nicholson (Nottingham University Hospitals
352 NHS Trust), Anirvan Banerjee (South Tees Hospital NHS Foundation Trust), Clare Marris
353 (Sheffield Teaching Hospital NHS Foundation Trust), Pam Comiskey (NHS Fife), Georgina
354 Wilkinson (Manchester University NHS Foundation Trust), Peter Byrom (Peter Byrom
355 Audiology, Sheffield), Laura Turton (South Warwickshire NHS Foundation), Bernadette
356 Parker (University Hospitals Coventry and Warwickshire NHS Trust), Mary-Louise Montague
357 (NHS Lothian), Leza Munro (NHS Forth Valley), Joanne Goss (GIG Cymru NHS Wales), Ali
358 Hilali (Bridgewater Community Healthcare NHS Foundation Trust), Veronica Kennedy
359 (Bolton NHS Foundation Trust), Beth Claesen (Bath and North East Somerset – Virgin Care),
360 Tony Kay (Aintree University Hospital NHS Foundation Trust), Zarina Naeem (Manchester

361 University NHS Foundation Trust), Alan Hopkirk (Invizear) are thanked for their contributions
 362 in this study.

363

364 *Table 1. Demographics of the expert panel.*

	Frequency
Job Role:	
Audiologist	17
Audiologist/Hearing Therapist	12
ENT Specialist	3
Audiovestibular physician	3
Clinical Scientist	3
Paediatrician	2
Academic/CBT Psychotherapist	1
Sector:	
Public (NHS)	37
Private	4
Years in current profession:	
0 - 9	6
10 -19	8
20 -29	16
30+	11
Hyperacusis patients seen in last 3 months:	
0-4	13
5-9	15
10-14	8
15-19	2
20	2
40	1

365

366 **References**

367 AAZH, H., MCFERRAN, D. & MOORE, B. C. J. 2018. Uncomfortable loudness levels among children
368 and adolescents seeking help for tinnitus and/or hyperacusis. *Int J Audiol*, 57, 618-623.

369 AAZH, H. & MOORE, B. C. J. 2017. Factors related to uncomfortable loudness levels for patients seen
370 in a tinnitus and hyperacusis clinic. *Int J Audiol*, 56, 793-800.

371 ANARI, M., AXELSSON, A., ELIASSON, A. & MAGNUSSON, L. 1999. Hypersensitivity to sound:
372 Questionnaire data, audiometry and classification. *Scandinavian Audiology*, 28, 219-230.

373 ANDERSSON, G., LINDVALL, N., HURSTI, T. & CARLBRING, P. 2002. Hypersensitivity to sound
374 (hyperacusis): a prevalence study conducted via the Internet and post. *Int J Audiol*, 41, 545-
375 54.

376 ASHA'ARI, Z. A., MAT ZAIN, N. & RAZALI, A. 2010. Phonophobia and hyperacusis: practical points
377 from a case report. *The Malaysian journal of medical sciences : MJMS*, 17, 49-51.

378 AUERBACH, B. D., RODRIGUES, P. V. & SALVI, R. J. 2014. Central gain control in tinnitus and
379 hyperacusis. *Frontiers in neurology*, 5, 206-206.

380 BAGULEY, D., MCFERRAN, D. & HALL, D. 2013. Tinnitus. *The Lancet*, 382, 1600-1607.

381 BAGULEY, D. M. 2003. Hyperacusis. *Journal of the Royal Society of Medicine*, 96, 582-585.

382 BLAESING, L. & KROENER-HERWIG, B. 2012. Self-reported and behavioral sound avoidance in tinnitus
383 and hyperacusis subjects, and association with anxiety ratings. *Int J Audiol*, 51, 611-7.

384 BLASING, L., GOEBEL, G., FLOTZINGER, U., BERTHOLD, A. & KRONER-HERWIG, B. 2010.
385 Hypersensitivity to sound in tinnitus patients: an analysis of a construct based on
386 questionnaire and audiological data. *Int J Audiol*, 49, 518-26.

387 BOYATZIS, R. E. 1998. *Transforming qualitative information: Thematic analysis and code
388 development*, sage.

389 BSA 2011. Recommended procedure: Determination of uncomfortable loudness levels. British
390 Society of Audiology Reading, UK.

391 CHEN, Y.-C., LI, X., LIU, L., WANG, J., LU, C.-Q., YANG, M., JIAO, Y., ZANG, F.-C., RADZIOW, K., CHEN,
392 G.-D., SUN, W., KRISHNAN MUTHAIAH, V. P., SALVI, R. & TENG, G.-J. 2015. Tinnitus and
393 hyperacusis involve hyperactivity and enhanced connectivity in auditory-limbic-arousal-
394 cerebellar network. *eLife*, 4, e06576-e06576.

395 COELHO, C. B., SANCHEZ, T. G. & TYLER, R. S. 2007. Hyperacusis, sound annoyance, and loudness
396 hypersensitivity in children. *Prog Brain Res*, 166, 169-78.

397 DANESH, A. A., LANG, D., KAF, W., ANDREASSEN, W. D., SCOTT, J. & ESHRAGHI, A. A. 2015. Tinnitus
398 and hyperacusis in autism spectrum disorders with emphasis on high functioning individuals
399 diagnosed with Asperger's Syndrome. *Int J Pediatr Otorhinolaryngol*, 79, 1683-8.

400 DAUMAN, R. & BOUSCAU-FAURE, F. 2005. Assessment and amelioration of hyperacusis in tinnitus
401 patients. *Acta Oto-Laryngologica*, 125, 503-509.

402 FABIJANSKA, A., ROGOWSKI, M., BARTNIK, G. & SKARZYNSKI, H. Epidemiology of tinnitus and
403 hyperacusis in Poland. Proceedings of the sixth international tinnitus seminar, 1999.
404 Citeseer, 569-571.

405 FACKRELL, K. & HOARE, D. J. 2019. Scales and questionnaires for decreased sound tolerance.
406 *Hyperacusis and disorders of sound intolerance: clinical and research perspectives*. Plural
407 Publishing, San Diego.

408 FACKRELL, K., POTGIETER, I., SHEKHAWAT, G. S., BAGULEY, D. M., SEREDA, M. & HOARE, D. J. 2017.
409 Clinical Interventions for Hyperacusis in Adults: A Scoping Review to Assess the Current
410 Position and Determine Priorities for Research. *Biomed Res Int*, 2017, 2723715.

411 FACKRELL, K., STRATMANN, L., GRONLUND, T. A. & HOARE, D. J. 2019a. Top ten hyperacusis research
412 priorities in the UK. *The Lancet*, 393, 404-405.

413 FACKRELL, K., STRATMANN, L., KENNEDY, V., MACDONALD, C., HODGSON, H., WRAY, N., FARRELL, C.,
414 MEADOWS, M., SHELDRAKE, J., BYROM, P., BAGULEY, D. M., KENTISH, R., CHAPMAN, S.,
415 MARRIAGE, J., PHILLIPS, J., POLLARD, T., HENSHAW, H., GRONLUND, T. A. & HOARE, D. J.
416 2019b. Identifying and prioritising unanswered research questions for people with

417 hyperacusis: James Lind Alliance Hyperacusis Priority Setting Partnership. *BMJ Open*, 9,
418 e032178.

419 FORMBY, C., SHERLOCK, L. P. & GOLD, S. L. 2003. Adaptive plasticity of loudness induced by chronic
420 attenuation and enhancement of the acoustic background (L). *The Journal of the Acoustical*
421 *Society of America*, 114, 55-58.

422 GOODSON, S. & HULL, R. 2015. Hyperacusis. *American Speech-Language-Hearing Association.*
423 *Audiology Information Series: Hyperacusis.*

424 GOTHELF, D., FARBER, N., RAVEH, E., APTER, A. & ATTIAS, J. 2006. Hyperacusis in Williams syndrome:
425 characteristics and associated neuroaudiologic abnormalities. *Neurology*, 66, 390-5.

426 GREENBERG, B. & CARLOS, M. 2018. Psychometric Properties and Factor Structure of a New Scale to
427 Measure Hyperacusis: Introducing the Inventory of Hyperacusis Symptoms. *Ear Hear*, 39,
428 1025-1034.

429 GUIMARÃES, A. C., DE CARVALHO, G. M., MONTEIRO ZAPPELINI, C. E., MEZZALIRA, R., STOLER, G.,
430 PASCHOAL, J. R. & MÁRCIA MARIA DE FREITAS DIAS, V. 2014. Study of the relationship
431 between the degree of tinnitus annoyance the presence of hyperacusis☆☆Please cite this
432 article as: Guimarães AC, Carvalho GM, Voltolini MM, Zappellini CE, Mezzalira R, Stoler G, et
433 al. Study of the relationship between the degree of tinnitus annoyance and the presence of
434 hyperacusis. *Braz J Otorhinolaryngol*. 2014;80:24-8. *Brazilian Journal of Otorhinolaryngology*,
435 80, 24-28.

436 HANNULA, S., BLOIGU, R., MAJAMAA, K., SORRI, M. & MAKI-TORKKO, E. 2011. Self-reported hearing
437 problems among older adults: prevalence and comparison to measured hearing impairment.
438 *J Am Acad Audiol*, 22, 550-9.

439 HEBERT, S., FOURNIER, P. & NORENA, A. 2013. The auditory sensitivity is increased in tinnitus ears. *J*
440 *Neurosci*, 33, 2356-64.

441 HÉBERT, S., FOURNIER, P. & NOREÑA, A. 2013. The Auditory Sensitivity is Increased in Tinnitus Ears.
442 *The Journal of Neuroscience*, 33, 2356-2364.

443 HOARE, D. J., BYROM, P. & SHADIS, A. 2019. *Determination of uncomfortable loudness levels*
444 [Online]. British Society of Audiology (BSA). Available: [http://www.thebsa.org.uk/wp-](http://www.thebsa.org.uk/wp-content/uploads/2019/07/ULLs-recommended-procedure-Public-Consultation-Copy.pdf)
445 [content/uploads/2019/07/ULLs-recommended-procedure-Public-Consultation-Copy.pdf](http://www.thebsa.org.uk/wp-content/uploads/2019/07/ULLs-recommended-procedure-Public-Consultation-Copy.pdf)
446 [Accessed 01/03/2020].

447 JASTREBOFF, P. J. & HAZELL, J. W. 1993. A neurophysiological approach to tinnitus: clinical
448 implications. *British journal of audiology*, 27, 7-17.

449 JASTREBOFF, P. J. & JASTREBOFF, M. M. 2000. Tinnitus Retraining Therapy (TRT) as a method for
450 treatment of tinnitus and hyperacusis patients. *J Am Acad Audiol*, 11, 162-77.

451 JURIS, L., ANDERSSON, G., LARSEN, H. C. & EKSELIUS, L. 2013. Psychiatric comorbidity and
452 personality traits in patients with hyperacusis. *Int J Audiol*, 52, 230-5.

453 JÜRIS, L., ANDERSSON, G., LARSEN, H. C. & EKSELIUS, L. 2014. Cognitive behaviour therapy for
454 hyperacusis: A randomized controlled trial. *Behaviour Research and Therapy*, 54, 30-37.

455 KHALFA, S., BRUNEAU, N., ROGE, B., GEORGIEFF, N., VEUILLET, E., ADRIEN, J. L., BARTHELEMY, C. &
456 COLLET, L. 2004. Increased perception of loudness in autism. *Hear Res*, 198, 87-92.

457 KHALFA, S., DUBAL, S., VEUILLET, E., PEREZ-DIAZ, F., JOUVENT, R. & COLLET, L. 2002. Psychometric
458 normalization of a hyperacusis questionnaire. *Orl*, 64, 436-442.

459 KLEIN, A. J., ARMSTRONG, B. L., GREER, M. K. & BROWN, F. R., 3RD 1990. Hyperacusis and otitis
460 media in individuals with Williams syndrome. *J Speech Hear Disord*, 55, 339-44.

461 KNIPPER, M., VAN DIJK, P., NUNES, I., RUTTIGER, L. & ZIMMERMANN, U. 2013. Advances in the
462 neurobiology of hearing disorders: recent developments regarding the basis of tinnitus and
463 hyperacusis. *Prog Neurobiol*, 111, 17-33.

464 KNOBEL, K. A. & SANCHEZ, T. G. 2006. [Loudness discomfort level in normal hearing individuals]. *Pro*
465 *Fono*, 18, 31-40.

466 LEVINE, R. A. & ORON, Y. 2015. Tinnitus. *Handb Clin Neurol*, 129, 409-31.

467 LINSTONE, H. A. & TUROFF, M. 1975. *The Delphi Method*, Addison-Wesley Reading, MA.

468 MARRIAGE, J. & BARNES, N. M. 1995. Is central hyperacusis a symptom of 5-hydroxytryptamine (5-
469 HT) dysfunction? *The Journal of Laryngology & Otology*, 109, 915-921.

470 MATHISEN, H. 1969. Phonophobia After Stapedectomy. *Acta Oto-Laryngologica*, 68, 73-77.

471 MEEUS, O. M., SPAEPEN, M., RIDDER, D. D. & HEYNING, P. H. 2010. Correlation between hyperacusis
472 measurements in daily ENT practice. *Int J Audiol*, 49, 7-13.

473 MOORE, E. 1998. *Cochlear Hearing Loss*, London, Whurr.

474 MUNRO, K. J., TURTLE, C. & SCHAETTE, R. 2014. Plasticity and modified loudness following short-
475 term unilateral deprivation: evidence of multiple gain mechanisms within the auditory
476 system. *J Acoust Soc Am*, 135, 315-22.

477 NELTING, M. & FINLAYSON, N. K. 2004. *Geräuschüberempfindlichkeits-Fragebogen: GÜF; Manual*,
478 Hogrefe.

479 NELTING, M., RIENHOFF, N. K., HESSE, G. & LAMPARTER, U. 2002. Die Erfassung des subjektiven
480 Leidens unter Hyperakusis mit einem Selbstbeurteilungsbogen zur
481 Geräuschüberempfindlichkeit (GÜF). *Laryngo-Rhino-Otol*, 81, 327-334.

482 PAULIN, J., ANDERSSON, L. & NORDIN, S. 2016. Characteristics of hyperacusis in the general
483 population. *Noise & health*, 18, 178-184.

484 PHILLIPS, D. P. & CARR, M. M. 1998. Disturbances of loudness perception. *J Am Acad Audiol*, 9, 371-
485 9; quiz 399.

486 POTGIETER, I., MACDONALD, C., PARTRIDGE, L., CIMA, R., SHELDRAKE, J. & HOARE, D. J. 2019.
487 Misophonia: A scoping review of research. *Journal of Clinical Psychology*, 75, 1203-1218.

488 SHELDRAKE, J., DIEHL, P. U. & SCHAETTE, R. 2015. Audiometric Characteristics of Hyperacusis
489 Patients. *Frontiers in Neurology*, 6.

490 TAYLOR, S. 2017. Misophonia: A new mental disorder? *Med Hypotheses*, 103, 109-117.

491 TYLER, R. S., PIENKOWSKI, M., RONCANCIO, E. R., JUN, H. J., BROZOSKI, T., DAUMAN, N., DAUMAN,
492 N., ANDERSSON, G., KEINER, A. J., CACACE, A. T., MARTIN, N. & MOORE, B. C. 2014. A review
493 of hyperacusis and future directions: part I. Definitions and manifestations. *Am J Audiol*, 23,
494 402-19.

495 VERNON, J. A. 1987. Pathophysiology of tinnitus: a special case--hyperacusis and a proposed
496 treatment. *Am J Otol*, 8, 201-2.

497 WAGENAAR, O., WIERINGA, M. & VERSCHUURE, H. 2010. A cognitive model of tinnitus and
498 hyperacusis; a clinical tool for patient information, appeasement and assessment. *Int*
499 *Tinnitus J*, 16, 66-72.

500