Decreased Sound Tolerance Disorders (Hyperacusis and Misophonia) in Cognitively Able Autistic Adults I: Prevalence and Clinical Features

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Abstract

Background: Decreased sound tolerance (DST; i.e., difficulty tolerating sounds in one's environment) is one of the most common sensory features of autism, with a lifetime prevalence between 50–70%. Multiple disorders, including hyperacusis (the perception of moderate-intensity sounds as excessively loud or physically painful) and misophonia (a neuropsychiatric condition in which specific "trigger" sounds cause excessive emotional reactions such as anger, irritation, and disgust) likely contribute to the burden of DST in the autistic population, but there has been little exploration of this symptom cluster in autism in a way that distinguishes these conditions from each other or that examines how these disorders potentially differ in autistic and non-autistic individuals.

Methods: A sample of 936 independent autistic adults was recruited from the Simons Foundation Powering Autism Research for Knowledge cohort. Hyperacusis and misophonia case status were operationalized using self-report survey items reflecting DST symptom endorsement and functional impairment. Descriptive statistics were derived to report on the prevalence of each DST category, as well as clinical features of individuals in both categories.

Results: Across the sample, 40.2% met case criteria for hyperacusis (sex-weighted prevalence: 33.5%), whereas 35.5% met case criteria for misophonia (sex-weighted prevalence: 29.1%), and comorbid hyperacusis and misophonia was found in 27.0% (sex-weighted prevalence: 21.9%). Both hyperacusis and misophonia were associated with tinnitus and tinnitus disorder, although neither condition was associated with (diagnosed) hearing loss. Sound-evoked pain was a commonly reported symptom of hyperacusis, with 64.7% of autistic adults experiencing pain at least once per month and 24.7% of autistic individuals with hyperacusis meeting criteria for "pain hyperacusis." Onset of hyperacusis was much earlier than previously reported in non-autistic samples, although most other features of both hyperacusis and misophonia did not differ from what is known from general population studies.

Conclusion: Both hyperacusis and misophonia (and their comorbidity) are highly prevalent in the autistic population and should be studied in tandem (rather than as a unified construct) in future investigations of autism-associated DST.

Keywords: hyperacusis, misophonia, epidemiology, prevalence, autism, comorbidity

Community Brief

Why is this an important issue?

It is well known that autistic people have "decreased sound tolerance" (sound sensitivity), but there are actually multiple different types of sound sensitivities that researchers usually don't study separately. This is one of the first large studies to look at two of these subtypes, *hyperacusis* (trouble putting up with everyday sounds because they're too loud or physically painful) and *misophonia* (getting extremely and irrationally angry, annoyed, or disgusted when you hear very specific "trigger" sounds like chewing, sniffing, or repetitive sounds) in autistic adults.

What was the purpose of this study?

The purpose of this study was to see how many autistic adults have hyperacusis and misophonia, how often these two types of sound sensitivity overlap, and whether the symptoms of hyperacusis and misophonia look the same in autistic people as they do in non-autistic people.

What did the researchers do?

The researchers used surveys to ask about sensory sensitivities, health, and quality of life in a large sample of over 900 legally independent autistic adults who were part of SPARK (an online participant pool). These surveys included questionnaires that specifically asked about hyperacusis and misophonia symptoms, which they then looked at.

What were the results and conclusions of the study?

A lot of autistic adults have hyperacusis and misophonia, even to the point where it's bad enough to get in the way of their lives (hyperacusis: 40.2%; misophonia 35.5%). Most of the autistic people who have one of these disorders seem to have the other, though it's not 100% overlap. Hyperacusis starts at an earlier age in autism than it does in the general population, but other than that, the ways these disorders look seem to be very similar in autistic and non-autistic people, as far as this study can tell.

What are potential weaknesses in the study?

This study only used surveys and not other things like clinical interviews or hearing tests to "diagnose" hyperacusis or misophonia, so it's possible that some classifications are wrong. More work with better diagnoses of these conditions (and their co-occurrence) is needed.

How will these findings help autistic adults now or in the future?

This study brings attention to the fact that autistic adults do indeed still have high rates of sound sensitivity disorders, and those disorders are the same ones seen in non-autistic people presenting to audiology clinics with these complaints. People considering sound sensitivity in the context of autism would do well to consider the ways in which these conditions are diagnosed and treated by audiologists and other allied health professionals already, as it is unlikely that autism-related sound sensitivity disorders are unique enough to warrant their own separate care pathways.

Introduction

Autistic individuals frequently find aspects of their sensory environment that do not bother most people to be very difficult to tolerate, and since the publication of the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5[1]), hyperreactivity to sensory stimuli has been among the core diagnostic criteria for autism. Sensory hyperreactivity can occur across all sensory modalities (e.g., vision, hearing, touch, taste), appears unrelated to intellectual functioning,[2] and remains present into adulthood.[3] Moreover, many in the adult autistic community appear very invested in addressing or even developing effective interventions for sensory hyperreactivity, as it constitutes a salient correlate of reduced quality of life for many autistic adults,[4] as well as a substantial contributor to distress and impairment in their daily lives.[5] Given its association with significant disability and acceptability as a treatment target by much of the autistic community, this domain of autism features constitutes one of the primary areas where the implementation of "neurodiversity-affirmative biomedical research" (i.e., community-infomed basic/translational science informing targeted biomedical intervention development[6] could provide immense benefit to autistic individuals.

Among the varying types of sensory hyperreactivity observed in autism, hyperreactivity to auditory stimuli, hereafter referred to as decreased sound tolerance (DST[7]), is among the most common and debilitating. A previous meta-analysis by our group [8] estimated that approximately 50–70% of autistic people experience some sort of clinically significant DST in their lifetime, with current prevalence rates varying by age and seeming to decrease slightly over time, consistent with findings in a later longitudinal study[9]. The prominence of DST, particularly the subtype of hyperacusis, in autism is well-known, and an abundance of review articles have been published on the topic in recent years[7,10–16] describing clinical characteristics, impacts on functioning, management strategies, putative correlates, and hypothesized mechanisms. However, despite growing interest in the topic, empirical research on autism and DST is extremely limited in scope and breadth, and many foundational questions about the nature of DST in autism (as well as methodological questions about how best to define, operationalize, and study it) remain to be investigated.

Notably, DST is not actually a unitary construct,[7,17,18] despite often being treated as one in the context of autism research.[7] In widely-accepted frameworks,[7,17–19] the taxonomy of DST disorders includes at least three major conditions, of which two (hyperacusis and misophonia) have been most frequently studied.[20] *Hyperacusis* is a disorder in which moderate-intensity sounds that are thought of as innocuous by most are perceived as excessively loud, distressing, or causing physical pain[18,21,22]; there is some evidence that "pain hyperacusis" (in which everyday sounds frequently cause sensations of ear/head pain) may represent a distinct condition or subtype.[23–25] *Misophonia* is another disorder of sound tolerance in which individuals have excessive and inappropriate emotional responses such as anger, extreme irritation, or disgust in response to specific "trigger" sounds (e.g., chewing, tapping, sniffling[26–28]), although unlike in hyperacusis, the strength of these reactions is unrelated to volume and instead primarily dependent on whether the source of the sound can be identified[29]. Compounding this issue, most work on DST in autism is based on caregiver reports of their (often young or minimally speaking) child's aversion to sounds, and behavioral indicators of DST such as covering one's ears or physically distancing oneself from a sound source could theoretically be indicators of multiple DST subtypes.

Given the potential conflation of DST subtypes, much of the literature regarding DST phenotypes and correlates in autism is difficult to interpret, and it leaves the field's phenomenological understanding of DST-related experiences in autistic individuals much more impoverished than previously realized (although see recent work by Scheerer et al.30,31 for notable exceptions that have adopted the multiple-disorder framework). Additionally, due to the lack of empirical studies examining DST at the level of its constituent disorders in the autistic population, we currently know little about (a) whether and how these disorders differ phenotypically or mechanistically from analogous DST disorders in non-autistic individuals,7,32 (b) whether there is a strong comorbidity between DST disorders in autistic individuals (for which evidence is mixed in the non-autistic population33–37), and (c) whether the categorical labels of specific DST disorders (i.e., a hyperacusis or misophonia "diagnosis") would be useful in parsing the heterogeneity in clinical outcomes for autistic individuals after accounting for more frequently-assessed individual differences.

The present study begins to fill some of these gaps in the literature by conducting an initial large-scale survey of a group of autistic individuals capable of easily self-reporting on their experiences of DST (i.e., cognitively and linguistically able adults), which can then be used as a starting place for phenomenological inquiry into more difficult-to-assess populations, such as autistic children and individuals with co-occurring cognitive/language impairments. By leveraging a large online sample of legally independent autistic adults, the current study sought to accomplish the following goals:

- Investigate self-reported symptoms of multiple DST disorders (hyperacusis and misophonia) and their dimensional associations in the adult autistic population.
- Estimate the prevalence of categorical "clinically significant hyperacusis" and "clinically significant misophonia" in autistic adults, as well as their co-occurrence with each other, tinnitus, and hearing loss.

- Describe the salient clinical features of hyperacusis and misophonia as they present in the autistic population.

This study is presented in two parts, with the current article focusing exclusively on DST prevalence and clinical features and a companion article[38] focusing on the demographic and clinical correlates of hyperacusis and misophonia in the current sample.

Materials and Methods

Participants

Independent autistic adults above the age of 18 years (all of whom were their own legal guardians, and all of whom self-reported professional diagnoses of DSM-5 autism spectrum disorder or equivalent previously-used diagnoses such as Asperger's disorder) were recruited during Spring 2021 from the Simons Foundation Powering Autism Research for Knowledge (SPARK)[39] cohort using the SPARK Research Match service (Project No. RM0111Woynaroski_DST). These individuals, although not formally screened for intellectual disability beyond the SPARK medical history questionnaire (4.97% of sample positive for "*Intellectual disability, cognitive impairment, global developmental delay, or borderline intellectual functioning"*; 92.3% non-missing data), these and other prior results in the SPARK cohort[40] helped support the assertion that an overwhelming majority of our current sample was of average cognitive ability. A total of 1271 individuals consented to participate in the study, 936 of whom were included in the current analyses (894 with no missing data). Additional information on the survey exclusion parameters, data cleaning process, and justifications for various exclusions can be found in the Supplemental Methods. All participants gave informed consent, and all study procedures were approved by the institutional review board at Vanderbilt University Medical Center.

Measures

Hearing and Sound Tolerance Disorder Diagnosis Operationalization. As DST disorders have no consensus diagnostic criteria or widely-accepted "gold standard" for diagnosis, our research group developed a set of operational diagnostic criteria for hyperacusis, misophonia, and phonophobia, which have been published previously.[19,41] Instead of individuals self-reporting previous clinical diagnoses, which are rarely given for DST conditions in the autistic population, or self-identifying whether they met the criteria for a disorder based on a vignette or description, we opted to operationalize "clinically-significant hyperacusis" and "clinically-significant misophonia" status based on individual responses to survey questions that we felt could satisfy each of the individual diagnostic criteria listed (see Table 1 for full item text, source surveys, and endorsement frequencies). Participants were not categorized into similar binary groups for the related DST condition of *phonophobia* (a specific phobia of sound, often secondary to other DST conditions[18,20]) due to an inability to operationalize the key diagnostic criterion that an individual's fear or anxiety be *"out of proportion to the actual danger caused by the specific sound or situation (including the potential to provoke other symptoms such as hyperacusis pain).*"[19(p3)] Thus, only the DST disorders of hyperacusis and misophonia were assessed categorically in the current study.

Hyperacusis was screened for using questions adapted from (a) the single-question operationalization of the condition in the 2014 National Health Interview Survey (NHIS[42]): "*Some people are bothered by everyday sounds or noises that don't bother most people. Do everyday sounds, such as from a hair dryer, vacuum cleaner, lawnmower, or siren, seem too loud or painful to you?"*, and (b) the single hyperacusis screening item from the European School for Interdisciplinary Tinnitus Research Screening Questionnaire (ESIT-SQ[43]): *"Over the* <u>*last*</u>

month, *have external sounds been a problem, being too loud, uncomfortable, or painful for you when they seemed normal to others around you?"* Item scores from one of the dimensional DST questionnaires explored in the companion article[38] [the Inventory of Hyperacusis Symptoms (IHS[44,45]) and a newly-written item asking about the frequency of sound-induced pain over the past year (based on a previous question analyzed in a phenotypic study of pain hyperacusis[25])] were also included in the hyperacusis "caseness" algorithm.

In order to be classified as having clinically-significant hyperacusis, autistic adults in the current study were required to meet four operational criteria of (a) endorsing the NHIS-like hyperacusis screening question, (b) clinically significant amounts of loudness intolerance and/or sound-induced pain, (c) difficulty being in loud places, and (d) associated impairment (Table 1). Criteria of the algorithm (e.g., criterion *b.* "clinically significant loudness intolerance or sound-induced pain") were mapped to certain response options of the IHS or another measure (e.g., "*Compared to most people, common everyday sounds seem excessively loud to me"* [hyperacusis response: "Very much so"]; "*Sound can cause me pain and/or physical discomfort"* [hyperacusis responses: "Somewhat" / "Very much so"]) based on their theoretical correspondence to clinical severity. Additionally, a subset of the individuals who were classified as having hyperacusis were further classified as having "pain hyperacusis" if they reported experiencing sound-induced pain more days than not (i.e., at least 4 times per week).

Misophonia status was ascertained in the sample using the Duke-Vanderbilt Misophonia Screening Questionnaire (DVMSQ[46]), a questionnaire developed by our research group that has been previously published and found to be invariant in its latent structure between autistic and non-autistic individuals. The DVMSQ includes a theory-based diagnostic algorithm, based on the misophonia consensus definition[26] and our group's criteria for misophonia,[19,41] which can be operationalized using a combination of DVMSQ questionnaire items.[46] Individuals meeting DVMSQ screening criteria for "clinical misophonia" were categorized as such in the current study (see Table 1 for individual items and endorsement frequencies).

An additional condition of interest for the current investigation was tinnitus, which is commonly comorbid with many forms of DST, particularly hyperacusis.[23,47] To assess whether an individual met criteria for tinnitus, we used the single-item tinnitus screening question from the ESIT-SQ: "*Tinnitus refers to the perception of noise in your head or ears (such as ringing or buzzing) in the absence of any corresponding source of sound external to your head. Over the past year, have you had tinnitus in your head or in one or both ears that lasts for more than five minutes at a time?*" Responses indicating episodic or chronic tinnitus in the past year (lasting more than 5 minutes at a time or longer) were used to classify an individual as meeting criteria for tinnitus. Furthermore, we sought to determine which subset of autistic individuals with tinnitus met criteria for *tinnitus disorder* (i.e., tinnitus that is associated with emotional distress, cognitive dysfunction, and/or autonomic arousal, leading to behavioral changes and functional disability[48]). To define tinnitus disorder, we administered individuals meeting criteria for tinnitus the Tinnitus Handicap Inventory–Screening Version (THI-S[49]); individuals scoring ≥6 on this measure (the recommended cutoff on the THI-S for "clinically significant tinnitus") were classified as having tinnitus disorder.

Lastly, as hyperacusis is also frequently associated with some hearing loss in the general population,[50] participants reported hearing loss status using previous diagnoses of any of the following: "presbycusis," "sudden sensorineural hearing loss," "other hearing loss," or "Ménière's disease". A selection of any of the aforementioned conditions was sufficient to classify an individual as meeting case criteria for hearing loss in the current study.

Measures of DST Symptoms and Clinical Features. As part of the study survey, participants completed multiple previously developed and psychometrically tested self-report measures of DST symptomatology, including the Inventory of Hyperacusis Symptoms,[44,45] Duke-Vanderbilt Misophonia Screening Questionnaire,[46] Duke Misophonia Questionnaire (DMQ[51]), and DSM-5 Specific Phobia Severity Scale–Modified for Phonophobia (DSM-Phono[52]). With the exception of the DMQ (which was presented in a modified form[46]), additional information about these measures (and the remaining measures assessing clinical correlates) can be found in the companion article,[38] in which those instruments are analyzed dimensionally in the same sample described here. In the present study, individual items from the DVMSQ and DMQ were incorporated into the analysis of the clinical features of misophonia (for additional detail, see "Descriptive Analyses" section).

Duke Misophonia Questionnaire (DMQ). The Duke Misophonia Questionnaire (DMQ[51]) is an 86-item modular self-report questionnaire that assesses a large number of misophonia-related constructs (i.e., specific triggers, trigger frequency, symptoms in response to misophonic triggers [affective, physical, cognitive], specific coping strategies [before, during, and after being triggered], misophonia-related impairment, and dysfunctional beliefs about misophonia). This measure was rigorously developed using a stakeholder-informed design to generate a comprehensive item pool, and a preliminary psychometric study has established the latent structure, reliability, and convergent validity of the DMQ subscales in a sample of general-population adults,[51] although it has not been formally evaluated psychometrically in the autistic population. To reduce participant burden, the current study administered an abbreviated version of the DMQ[46] that included: (a) the trigger list (16 Yes/No items, including a "none of the above" option), (b) the "frequency of being triggered" item (6-point Likert scale from 1="*Once per* *month or less*" to 6="*6 or more times per day*"), (c) a novel "global impairment" item ("*Please rate the overall impact of ALL bothersome sounds on your life over the past month.*") that was rated on a visual analog scale (VAS) ranging from 0="*No Effect"* to 100="*Extreme Effect*" [presented after the "Frequency" item], (d) the 23 DMQ symptom scale (DMQ-SS) items (5 affective, 8 physiological, 10 cognitive; rated on a 5-point Likert scale from 0="*Not at all*" to 4="*Always/Almost always*"), and (e) the DMQ Impairment Scale (DMQ-IMP; 12 items). Two summary scores from the DMQ were derived in the current study: the total number of triggers (range 0–15) and the DMQ-IMP total score (12 items; range 0–4 [mean item score]; current sample [Cronbach's] α=0.955). Remaining items were included for descriptive/item-level analyses only. Notably, the DMQ-IMP total score allows individuals to be categorized into "misophonia impairment categories" based on level of reported impairment[51]—0="none" (includes individuals who did not endorse any triggers on the DMQ and, therefore, did not complete the remainder of the questionnaire] 0.01–1.08="minimal–mild"; 1.09–3.17="moderate"; 3.18–4.00="severe"—which were also described in the current study for individuals meting DVMSQ misophonia criteria.

Ad-hoc Clinical History Questionnaire. In addition to the validated measures of DST symptoms, we asked participants a number of newly written survey questions designed to assess their first-person experiences of DST, primarily hyperacusis (e.g., associated symptoms, onset, functional impact). These questions were presented together with the demographics form and relevant questions about medical/psychiatric history as a combined "Demographics and Health History Form." Although several of these novel questions were analyzed in a structured manner (e.g., those used to help determine hyperacusis caseness), the majority of questions were only analyzed descriptively. See Supplemental Table S1 for content and response options of the ad-hoc questions analyzed in the present study.

Data Analysis

All statistical analyses were conducted in the R statistical computing environment, version 4.3.0,[53] with many of the models based on functions from the openly-available *ZPack.Bayes* collection of functions written by the first author.[54]

Descriptive Analyses. We first described the sample in terms of self-reported demographics, as well as several clinical background variables of interest (e.g., self-reported psychiatric diagnoses, tinnitus/tinnitus disorder status, hearing loss status, prior traumatic brain injury, migraine). We also calculated the prevalence of "clinically-significant hyperacusis," "clinically-significant misophonia," and their overlap in our sample using the case ascertainment algorithms described previously, additionally reporting the frequency of individuals at each "impairment level" (for hyperacusis based on the ESIT-SQ hyperacusis question,[43] and for misophonia based on the DMQ-IMP subscale.[51]

Within the subsample of autistic adults meeting criteria for clinically significant hyperacusis, we specifically analyzed the phenotype of sound-induced pain, examining its frequency and intensity, as well as the proportion of autistic adults who would meet operational criteria for "pain hyperacusis." We also broadly queried about other sound-evoked physical symptoms of hyperacusis using the following question, "*When you hear a loud sound, do you ever experience any of the following physical sensations? (Select all that apply)*". Additional clinical features of hyperacusis, including the age of onset, laterality/asymmetry of symptoms, and presence/context of "setbacks" or temporary symptom flare-ups[55] were also explored.

Within the subsample of autistic adults meeting criteria for clinically significant misophonia, we reported on the number of triggers endorsed, as well as the frequency of each individual trigger endorsed (based on the DMQ trigger list). Frequency of being triggered, global impact, and emotional and physical responses to being triggered (based on DVMSQ and DMQ item responses, respectively), as well as the frequency of finding one's emotional responses to being triggered excessive/unreasonable [46] were also specifically explored in the misophonia subsample.

Categorical Associations. Although there were very few inferential statistics utilized in the current study, we did evaluate some associations of categorical variables using a Bayesian analog of the Pearson chi-squared test (Gûnel-Dickey contingency table tests[56,57]). This was implemented with the *bayes.proptest* function from *ZPack.Bayes*.[54] An independent multinomial sampling plan was used for 2×2 contingency tables, allowing us to conduct a test of the equality of two proportions (i.e., does the odds ratio of endorsement [*OR*] equal 1); Dirichlet priors (a_1, a_2=1) were placed on cell counts in each row. The *OR* was considered the quantity of interest, and 15,000 Monte Carlo posterior samples were calculated using the joint posterior distribution of all model parameters.[25] Notably, as the null hypothesis of complete equivalence is always false at the population level,[58] this posterior was then tested against the interval null hypothesis that the *OR* would fall within a region of values that we deemed *a priori* to be trivially small (i.e., the region of practical equivalence [ROPE[59]]). The ROPE was defined in this case to be an *OR* interval of [0.833, 1.20], which is approximately equal to Cohen's *d* values of [-0.1, 0.1] and therefore constitutes half the magnitude of a prototypical "small" effect size.[60] A ROPE Bayes factor (BF_{ROPE}[61–63]) was calculated to quantify evidence for or against the hypothesis that the *OR* falls within the ROPE. $BF_{\text{ROPE}}>3$ suggests values are outside the ROPE (proportions are meaningfully different), $BF_{ROPE}<1/3$ suggests they are within the ROPE (proportions are practically equivalent), and $1/3<BF_{ROPE}<3$ is inconclusive [64]. The Bayesian probability of direction [Pd; 61] was also derived as an analog to the p-value. P_d is approximately the inverse of a one-tailed *p*-value, with values of $P_d>.975$ suggesting "statistical significance" at the $p<.05$ level.

Results

Basic demographic and clinical data for individuals in the current sample (as well as hyperacusis, misophonia, and hyperacusis+misophonia subgroups) are reported in Table 2. As with the previous study drawn from this sample,[65] autistic adults in the present study (n=936, mean age 37.49 years, age range 18–80 years) were predominantly non-Hispanic White (80.1%; 77.1–78.3% of hyperacusis/misophonia groups), assigned female at birth (63.0%), college-educated (48.2% completed a 4-year college degree or more), and diagnosed with autism in adulthood (median age of diagnosis=23.21 years; 38.7% diagnosed before 18 years of age; 6.6% diagnosed before 4 years of age). Participants reported a mean of 1.78 lifetime psychiatric/neurodevelopmental diagnoses in addition to autism (range 0–7; 81.2% reporting at least one), as well as a mean of 0.67 functional somatic syndrome diagnoses, medical conditions such as fibromyalgia, irritable bowel syndrome, and interstitial cystitis that are symptom-based syndromes and difficult to conclusively associate with any specific pathology[66,67] [range 0–7; 36.5% reporting at least one]. Notably, a sizable portion of the sample (43.6%) reported tinnitus at the level ascertained by the ESIT-SQ, with nearly one third of autistic adults (32.8%) meeting criteria for *tinnitus disorder* as operationalized using the THI-S. Conversely, the prevalence of (professionally diagnosed) hearing loss was relatively low (7.7%).

Prevalence, Comorbidity, and Impact of Decreased Sound Tolerance Disorders

Using the hyperacusis and misophonia case ascertainment criteria outlined previously, 456 autistic adults (48.7% of the sample) met criteria for some form of clinically significant DST disorder (either hyperacusis, misophonia, or both). Of these individuals, 377 autistic adults (40.3% of the whole sample) met the criteria for hyperacusis, 332 (35.5% of the whole sample) met the criteria for misophonia, and 253 (27.0% of the whole sample) met criteria for both disorders simultaneously. Although the correlates of hyperacusis are explored more fully in the companion paper,[38] it is worth noting that the prevalence of both DST disorders varied substantially according to sex at birth (Hyperacusis: 49.7% of females vs. 24.3% of males; Misophonia: Hyperacusis: 44.2% of females vs. 20.5% of males). Given the sex-imbalanced nature of our sample, we also calculated sex-weighted prevalence estimates using the estimated 3:1 ratio of males to females in the autistic population [68], which were 33.5% for hyperacusis, 29.1% for misophonia, and 21.9% for both conditions.

Additionally, as categorically defined hyperacusis and misophonia were highly comorbid in the current sample (67.1% of the hyperacusis group and 76.2% of the misophonia group met criteria for the other condition; OR=12.26, $\mathrm{CrI}_{95\%}$ [9.08, 16.94], $P_{d>}$.999, $BF_{\mathrm{ROPE}=}8.49\times10^{15}$). Notably, the approximately half of individuals meeting criteria for either hyperacusis or misophonia was much larger than the number of autistic individuals who reported ever seeing a medical professional for help with DST-related symptoms (n=158; 16.9% of the sample), although having a clinical DST condition and seeking medical help for DST conditions were strongly associated (25.7% of those with clinically significant DST sought medical help vs. 8.5% of those without clinically significant DST; OR=3.73, $\mathrm{CrI}_{95\%}$ [2.56, 5.50], P_d>.999, $BF_{\mathrm{ROPE}}=1.17\times10^5$).

Comorbidity of hyperacusis with tinnitus was significant, with 56.0% of autistic adults with hyperacusis meeting tinnitus criteria as opposed to 35.2% of those without hyperacusis (OR=2.33, $\mathrm{CrI}_{95\%}$ [1.78, 3.03], P_d>.999, $BF_{\mathrm{ROPE}}=4.80\times10^3$). The relative increase was even higher for the association between hyperacusis and *tinnitus disorder* (47.2% vs. 23.1%; OR=2.98, $\mathrm{CrI}_{95\%}$ [2.25, 3.92], P_d>.999, $BF_{\mathrm{ROPE}}=2.46\times10^5$). Misophonia was also similarly associated with higher rates of both tinnitus (55.1% vs. 37.3%; OR=2.07, $\mathrm{CrI}_{95\%}$ [1.58, 2.72], P_d>.999, BF_{ROPE}=594) and tinnitus disorder (47.6% vs. 24.7%; OR=2.77, $\mathrm{CrI}_{95\%}$ [2.08, 3.67], P_d>.999, $BF_{\mathrm{ROPE}}=1.01\times10^5$). Notably, both hyperacusis and misophonia were unassociated with (diagnosed) hearing loss in the current sample (Hyperacusis: OR=1.14, $\mathrm{CrI}_{95\%}$ [0.704, 1.86], P_d=.699, BF_{ROPE}=0.070; Misophonia: OR=0.855, $\mathrm{CrI}_{95\%}$ [0.508, 1.40], P_d=.730, BF_{ROPE}=0.081).

In the hyperacusis group (n=377), ESIT-SQ impairment level was rated as "*a moderate problem*" in 44.0%, "*a big problem*" in 29.7%, and *"a very big problem"* in 26.3%. Using the DMQ-IMP cutoff scores for impairment in the misophonia group (n=332), 3.0% were rated as having no impairment, 23.2% as mild impairment, 62.7% as moderate impairment, and 11.1% as severe impairment.

Clinical Features of Hyperacusis

Sound-induced ear pain was a common symptom in the current sample, being endorsed at least once in the past year by 484 individuals (51.7%) and by 296 of the individuals with hyperacusis (78.5%). Within the subset of 377 autistic individuals with hyperacusis, nearly two thirds of individuals (64.7%) experienced sound-induced pain at least once per month, and for 93 individuals (24.7% of the hyperacusis group; 9.9% of the full sample), pain occurred more days than not, meeting case criteria for pain hyperacusis. Autistic adults with hyperacusis also endorsed a wide range of other sound-evoked symptoms (Figure 1), with the most common non-painful symptoms being tinnitus, palpitations, and ear fullness/pressure (each endorsed by over 40% of the sample). Intriguingly, nearly three quarters of the hyperacusis group (72.1%) endorsed temporary symptom “flare-ups” or “setbacks” caused by loud sounds at some point in their lives. Lastly, the onset of hyperacusis symptoms in this sample was quite early, occurring before age 6 in 59.9% of the sample and before age 12 in an additional 12.2%. Additional findings related to hyperacusis are reported in the Supplemental Results.

Clinical Features of Misophonia

Endorsement of each DMQ trigger in the misophonia group is displayed in Table 3. At least one “classic” misophonia trigger was endorsed by the vast majority of autistic adults judged to have clinically significant misophonia on the DVMSQ (oral/nasal sounds: 85.5%; repetitive sounds: 88.0%; any “classic trigger”: 94.9%), suggesting very few autistic individuals with clinical misophonia were un-triggered by all sounds in these categories. Primary emotions in response to triggers included intense irritation/annoyance, anger, disgust, and fear/panic, in that order. Additional findings related to misophonia are reported in the Supplemental Results.

Discussion

The current study presents one of the largest and most comprehensive investigations of DST phenomenology, prevalence, and clinical features in the autistic population to date, specifically focusing on the major DST disorders of hyperacusis and misophonia in a sample of predominantly cognitively able autistic adults. As one of the first studies to differentiate hyperacusis from misophonia in the autistic population (rather than grouping them together under the single label of “auditory hyperreactivity”), our results allow us to examine these two distinct yet related disorders, providing a foundation for understanding their separate etiologies, correlates, natural histories, and pathophysiological mechanisms. The present investigation focused primarily on exploring the number of autistic individuals meeting operational criteria for hyperacusis and misophonia, as well as specific clinical features associated with each condition. Although largely descriptive in nature, our findings provide some insight into fundamental questions about the nature of DST disorders in autistic individuals, providing a critical step toward advancing basic and applied research that can support clinically meaningful outcomes such as improved diagnosis, supports, and targeted interventions for these often-debilitating conditions.

Prevalence and Comorbidity of Hyperacusis and Misophonia

An important contribution of the current study was estimation of the prevalence of each DST disorder. Based on operational diagnostic criteria and self-reported symptoms, 48.7% of our autistic adult sample presented with either hyperacusis or misophonia, with clinically significant hyperacusis observed in 40.2% (sex-weighted prevalence: 33.5%), clinically significant misophonia was observed in 35.5% (sex-weighted prevalence: 29.1%), and comorbid hyperacusis and misophonia observed in 27.0% (sex-weighted prevalence: 21.9%). This research represents one of the first estimates of misophonia prevalence in the autistic population, and is the only study to assess this quantity using a measure that has been psychometrically validated in autistic individuals.[46] Compared to 7.3% of general population adults in the original DVMSQ validation study (*n*=1403) meeting case criteria for misophonia,[46] a prevalence of 29.1–35.5% clearly indicates that rates of clinically significant misophonia are substantially elevated in autistic relative to non-autistic adults, as has been previously assumed [7]. Estimates by Scheerer et al.[31], notably derived from a cohort of autistic adults that all self-reported some degree of (unspecified and possibly subclinical) DST, have also indicated that 56% of autistic adults met criteria for clinically significant hyperacusis and 30% met criteria for clinically significant misophonia, compared to 17% and 13% of non-autistic adults with self-reported DST who met criteria for these same conditions.

When considering clinically significant hyperacusis, the sex-weighted prevalence obtained in the current study was slightly lower than most "current prevalence" estimates in our previous meta-analysis of (largely pediatric) studies (meta-analytic estimate: 37–46%[8]). This is likely due to the previously-observed age effects on hyperacusis prevalence in autistic individuals,[8,9] which demonstrate that the prevalence of *clinically significant* hyperacusis decreases with age in the autistic population (though whether hyperacusis persists in these individuals to some subclinical extent remains unclear and in need of additional study). Nevertheless, this study demonstrates that rates of both hyperacusis and misophonia remain high in adulthood, with each persisting to a clinically significant extent in approximately 30% of autistic individuals. Lastly, approximately 16% of autistic individuals had previously sought care from a physician or allied health professional for their DST symptoms, a rate substantially lower than the number with clinically significant DST symptoms in the sample. Although reasons for this gap between clinically significant DST and help-seeking behavior were not formally explored, it is possible that clinicians' generally poor awareness of DST and limited ability to treat it in current clinical settings[69] may contribute to reduced motivation among patients to seek medical care at this time.

Within the current sample, diagnostic overlap (i.e., comorbidity) between hyperacusis and misophonia was exceptionally high, on par with comorbidity between mood and anxiety disorders.[70] Although some general population studies have indicated that these conditions may have substantial overlap,[34,71] this is to our knowledge the highest estimate of hyperacusis-misophonia comorbidity published to date and may potentially be a unique characteristic of the autistic DST phenotype. It is, however, notable that the one other survey to characterize the overlap of clinically significant hyperacusis and misophonia in a sample of autistic adults (all of whom self-reported some degree of clinical or subclinical DST) found rates of clinically significant hyperacusis, misophonia, and hyperacusis+misophonia in 56%, 30%, and 26% of the sample, respectively.[31] Given that the present study was conducted solely with self-report survey instruments, it may also be the case that at least some of this covariation is methodological (e.g., some individuals with hyperacusis meet DVMSQ screening criteria due to ambiguities in wording, or indicators for hyperacusis and misophonia, when used together, select cases where one diagnosis meets the clinical threshold, but the other is subclinical). However, unpublished data from our research group,[41] which include structured clinical interviews to characterize autistic and non-autistic adults with DST, suggest that the co-occurrence between DST conditions is indeed quite frequent, particularly in autistic adults,[73] and an explanation of these results as purely an artifact of the survey methodology is highly unlikely. As hyperacusis and misophonia have predominantly been studied in isolation up until this point, it is imperative, particularly in the autistic population, that studies of one of these constructs attempt to include measures of the other. It is additionally incumbent upon developers of psychometric tools to ensure that any putative measure of "hyperacusis symptoms" or "misophonia symptoms" demonstrates sufficient specificity for the DST domain of interest and can discriminate between individuals with hyperacusis and misophonia. Such transdiagnostic investigation of DST symptoms will ensure that the likely substantial population of individuals with both hyperacusis and misophonia can be further characterized, and that investigators will be able to isolate the effects of each DST dimension more confidently by incorporating adequate statistical controls for confounding by the other dimension/diagnosis.

Associations of hyperacusis and misophonia with hearing disorders, namely tinnitus, tinnitus disorder, and (diagnosed) hearing loss, were also assessed. As demonstrated in prior general population literature,[47] hyperacusis was strongly related to tinnitus, although the prevalence was somewhat lower in the autistic population (56.0% using a common epidemiologic screening question) than in some general population samples (e.g., 86% in two large samples[25,50]). Tinnitus disorder is a more recently established condition and given the lack of standardized operationalization of this disorder or similar "clinically significant tinnitus" definitions, it is difficult to determine whether the prevalence in the current sample is substantially higher or lower than other samples. Nevertheless, hyperacusis is typically believed to be a poor prognostic factor in tinnitus patients,[74] and a significant relationship with tinnitus *disorder* in the present sample is consistent with prior literature.

Interestingly, there was no significant relationship between hyperacusis and diagnosed hearing loss in autistic adults, despite such a relationship being previously reported for the general population.[50,75] This may be due to several reasons, including the fact that hyperacusis in autism is more likely to be neurodevelopmental than (putatively) post-traumatic in origin.[7] Additional contributors could be the relatively low prevalence of diagnosed hearing impairments in the current sample or the fact that associations between hyperacusis and hearing loss in previous studies was demonstrated using audiometric thresholds rather than self-report of professional diagnoses. Additionally, there were no meaningful differences in associations between misophonia, tinnitus, or hearing loss between autistic individuals with loudness and pain hyperacusis, mirroring general-population findings.[25]

Misophonia was also significantly associated with tinnitus and tinnitus disorder (though not hearing loss), replicating a prior association in the general population.[34] Notably, the association between misophonia and tinnitus in the current study was nearly as large as between hyperacusis and tinnitus (which was not the case in the general population study examining the same associations[34]), leading us to believe that at least some of the strength of this association could be explained by the strong overlap of misophonia and hyperacusis in the current sample. Genetic correlations have also been found between misophonia symptoms and tinnitus in other work,[76] suggesting that hyperacusis is unlikely to fully mediate this relationship. Nevertheless, additional investigations controlling for confounding by hyperacusis status are necessary to isolate the effect of misophonia when examining overlap with tinnitus and other hearing disorders.

Clinical Features of Hyperacusis

This study also provided an in-depth investigation of the clinical features of hyperacusis within the autistic population. Within our sample of autistic adults with hyperacusis, sound-induced ear pain was a frequently reported symptom, a finding that has never been empirically demonstrated in the literature until this point (although autistic individuals selected for DST appear to have some degree of sound-induced "pain or discomfort" per the IHS at high rates[31]). Though *pain hyperacusis* (i.e., hyperacusis with sound-induced pain more days than not) was only endorsed by approximately one in four autistic adults with hyperacusis, the majority of individuals with hyperacusis reported experiencing sound-induced pain at less regular intervals (78.5% within the past year, 64.7% in the past month). In addition to excessive loudness and sound-induced pain, autistic adults with hyperacusis endorsed many other sound-evoked symptoms, including tinnitus, palpitations, ear-fullness/pressure, and headaches, as well as temporary sound-induced symptom flare-ups similar to the "setbacks" seen in many non-autistic patients with hyperacusis.[25] Although the survey questions querying whether individuals *ever* experienced these symptoms (and therefore cannot speak to their frequency), it is notable that they do occur in a sizable minority of autistic individuals with hyperacusis (or in the case of symptom flare-ups, the majority) and therefore may play a role in the overall clinical picture.

Questions regarding the onset of hyperacusis provided insight into the nature and course of the condition and whether it is likely central or peripheral in origin. The majority of autistic adults with hyperacusis reported either a very early age of onset (<6 years) or not knowing when symptoms began (which, based on the clinical experience of the author interviewing autistic adults with DST,[41] is frequently associated with never remembering a time in one's life without DST), and fewer than 10% reported an age of hyperacusis onset after 12 years of age. This is in contrast to a general population sample reported by our group,[25] where the onset of hyperacusis was well into adulthood for the vast majority of participants. Such substantial differences suggest potentially different etiologies for autism-associated hyperacusis (theorized to be neurodevelopmental in origin, secondary to autism-linked alterations in synaptogenesis and other neurodevelopmental processes[7,77]) and the more "classic" acquired hyperacusis observed in non-autistic individuals with noise trauma, which is thought to be the result of central gain after peripheral auditory damage[75,78] but may also be a functional neurological symptom in at least a subset of cases.[79]

Clinical Features of Misophonia

Lastly, we investigated the clinical features of misophonia in autistic adults, in the first study to our knowledge to do so. Notably, autistic adults with misophonia endorsed many of the "classic" misophonia triggers (i.e., oronasal and repetitive sounds) with a very high frequency, with almost 95% of individuals in the misophonia group reported being triggered by at least one of these sounds. However, the single trigger category endorsed the most in the misophonia group was, "*People talking in the background (e.g., phone calls in public, many people talking at once).*" It is unclear whether the DMQ directions around triggers, which state to endorse all that "bother you much more intensely than they do other people," could play a role in this outside endorsement, as many autistic people report being *overwhelmed*, but perhaps not necessarily "triggered" in terms of a misophonic reaction, by many overlapping conversations.[31,80] Aside from this potential artifact, all other triggers appeared similar in rank-order to those selected by individuals in the general population in prior work.[51] Additional clinical features of misophonia observed were that: (a) the predominant emotions were irritation followed by anger and disgust (plus fear/panic to a lesser extent); (b) physical reactions predominantly included muscle tension, reflexively jumping, and heart racing; (c) a majority of participants (but not all) viewed their reactions as excessive or unreasonable; and (d) the degree of trigger avoidance and functional impairment from misophonia was substantial. All of these appear in line with the consensus definition of misophonia,[26] which was largely based on clinical descriptions of non-autistic patients, and suggest that the clinical features of misophonia do not substantially differ in autistic and non-autistic adults.

Limitations

This study is not without limitations. First and foremost, the DST constructs under study are themselves without consensus-based diagnostic criteria; as such, there are no "gold standard" diagnoses of hyperacusis or misophonia or instruments that can be used to make such diagnoses. The operational diagnostic criteria employed in the current study attempt to provide a standard for the field to follow, but they are clearly just a start and warrant modification with further input by experts and stakeholders. Beyond hyperacusis and misophonia, we also were unable to classify individuals for the disorder of *phonophobia* due to poor operationalization of some phonophobia criteria in our survey. Thus, it remains unclear how many autistic adults demonstrate DST due to phonophobia (with or without pre-existing hyperacusis or misophonia). Additionally, all symptoms were ascertained using self-report surveys; thus, the sensitivity and specificity of any symptom or classification may have been lower than if assessed using a clinician interview or more objective measure (e.g., audiometric testing in the case of hearing loss). Particularly in the case of prevalence estimates, future study utilizing validated clinical interviews would produce much more valuable data.

Moreover, the study did not assess or characterize subclinical manifestations of hyperacusis or misophonia, despite these likely corresponding to the majority of DST observed in autistic individuals.[31] However, when DST is rated as a minor problem or not significantly impacting one's life, it becomes less clinically relevant to characterize for the purposes of treatment and service provision, and characterization of subclinical DST was deemed out of the scope of the current study. More research is nevertheless needed to determine the phenomenology of the full DST spectrum present in autistic youth and adults. Another limitation is the lack of a non-autistic control group. A number of conclusions were drawn with regard to the general population prevalence or features of some of these conditions, though no general population sample actually completed this exact survey protocol in the present study. Though it may be reasonable to make cross-group comparisons with consideration of historical data, particularly as pronounced as these differences appear to be, additional studies that utilize formal control groups of non-autistic individuals will be particularly helpful in assessing between-group differences. Lastly, this study was limited in its demographics to a fairly unrepresentative group of autistic adults (i.e., predominantly cognitively-able adults who skewed highly educated, late diagnosed, and female). Sex-weighted prevalence estimates sought to correct for the most significant effect of sample composition on prevalence, but the remaining covariate effects could still have had an outsized effect on the sample. In the future, improved sampling schemes could be employed to better investigate the prevalence of these conditions in the broader autistic population.

Conclusion

Decreased sound tolerance represents one of the most prevalent and impactful subtypes of sensory reactivity in autistic individuals, and the disorders of hyperacusis and misophonia both appear to contribute significantly to the overall burden of DST in the autistic population. In a sample of 936 autistic adults recruited from SPARK, we descriptively analyzed the prevalence and clinical features of both DST conditions, finding that 48.7% of the sample demonstrated clinically significant DST of some type. Examining these results by DST condition, we found rates of hyperacusis in 40.2% of the sample (sex-weighted prevalence: 33.5%), misophonia in 35.5% of the sample (sex-weighted prevalence: 29.1%), and comorbid hyperacusis/misophonia in 27.0% of the sample (sex-weighted prevalence: 21.9%). Pain hyperacusis (sound-induced pain more days than not) was reported by approximately one fourth of individuals with hyperacusis (9.9% of the whole sample), and many other sound-evoked symptoms were also reported by a substantial minority of autistic hyperacusis patients. There were few differences in clinical features between the DST seen in autism and analogous disorders in the general population, although autism-associated hyperacusis appears to have a much earlier onset due to its neurodevelopmental origin. Misophonia in particular appeared to have a clinical picture very similar to that seen in the general population, suggesting that underlying mechanisms may not meaningfully differ in autistic and non-autistic individuals. This work highlights the relevance of both hyperacusis and misophonia (as well as their substantial overlap) to DST in the autistic population and demonstrates the need to study both disorders in tandem when investigating symptoms or mechanistic factors related to sound tolerance in future studies of autistic individuals.

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CRediT Author Statement

Zachary J. Williams: Conceptualization, Methodology, Software, Formal analysis, Data Curation, Visualization, Writing - Original Draft, Funding acquisition; **D. Jonah Barrett:** Software, Validation, Data Curation; **Carissa J. Cascio:** Writing - Review & Editing, Funding acquisition; **Tiffany G. Woynaroski:** Conceptualization**,** Writing - Review & Editing**,** Supervision**,** Funding acquisition

Disclosures

ZJW has received consulting fees from Roche and Autism Speaks. He is also vice-chair of ANSWER (Autistic and Neurodivergent Researchers Working for Equity in Research), a division of the Autism Intervention Research Network on Physical Health. He additionally serves on the scientific advisory boards of Simons Foundation Powering Autism Research for Knowledge (SPARK) and Hyperacusis Research Ltd., both unpaid positions. None of the other authors declare any conflicts of interest.

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Table 1: Criteria and Questions Used to Operationalize Hyperacusis and Misophonia Caseness

Hyperacusis Criteria			
Criterion	**Item [answers constituting endorsement]**	**Source Measure**	**Percent Endorsing**
Screening *[loudness OR pain from everyday sounds]*	*Some people are bothered by everyday sounds or noises that don't bother most people. Do everyday sounds, such as from a hair dryer, vacuum cleaner, lawnmower, or siren, seem too loud or painful to you?* [“Yes”]	NHIS [Adapted]	70.4%
Clinically significant loudness OR pain	*Compared to most people, common everyday sounds seem excessively loud to me:* [“Very much so”]	IHS	23.6%
	Sound can cause me pain and/or physical discomfort: [“Somewhat” / “Very much so”]	IHS	53.5%
	Over the past twelve (12) months, on average, approximately how often have you experienced physical pain in one or both ears as a result of sounds in your environment? [“One to three times per week” / “Four to seven times per week” / “Two to five times per day” / “Six or more times per day”]	Novel	20.7%
Avoidance of loud places	*I find the challenges of being exposed to loud sounds can make it difficult to be in loud places:* [“Somewhat” / “Very much so”]	IHS	75.1%
Hyperacusis-related impairment	*Over the last month, have external sounds been a problem, being too loud, uncomfortable, or painful for you when they seemed normal to others around you?* [“Yes, a moderate problem” / “Yes, a big problem” / “Yes, a very big problem”]	ESIT-SQ [Adapted]	51.5%
Misophonia Criteria			
Criterion	**Item [answers constituting endorsement]**	**Source Measure**	**Percent Endorsing**
Screening *[one or more trigger sounds]*	*Are there specific sounds that you are extremely bothered by, even if they are not loud? Examples include: chewing, slurping, crunching, throat clearing, finger tapping, foot shuffling, keyboard tapping, rustling, nasal sounds, pen clicking, appliance humming, clock ticking, and animal sounds.* [“Yes”]	DVMSQ	69.1%
Disproportionate feelings of irritation, anger/rage, or disgust	*When you are exposed to the bothersome sounds listed above, how often do you experience... Intense feelings of irritation or annoyance?* [“Often” / “Very often”]	DVMSQ	56.0%
	When you are exposed to the bothersome sounds listed above, how often do you experience... Feelings of anger or rage? [“Often” / “Very often”]	DVMSQ	25.2%
	When you are exposed to the bothersome sounds listed above, how often do you experience... Feelings of disgust? [“Often” / “Very often”]	DVMSQ	27.1%
Avoidance of triggers, or endurance with intense discomfort	*How often do you avoid situations where you may potentially hear these bothersome sounds?* [“Sometimes” / “Often” / “Very often”]	DVMSQ	58.7%
	When you are exposed to the bothersome sounds listed above, how often do you experience... Urges to cover your ears or block out the sound in some other way? [“Often” / “Very often”]	DVMSQ	53.8%
	When you are exposed to the bothersome sounds listed above, how often do you experience... Urges to run away from the sound? [“Often” / “Very often”]	DVMSQ	44.1%
Loss of Behavioral Control	*When you are exposed to the bothersome sounds listed above, how often do you experience... Feeling like you cannot control your response to the sound?* [“Sometimes” / “Often” / “Very often”]	DVMSQ	49.1%
	When you are exposed to the bothersome sounds listed above, how often do you experience... Urges to lash out violently at the person or object making the sound? [“Sometimes” / “Often” / “Very often”]	DVMSQ	30.6%

Misophonia-related impairment *[Requires at least two of seven items to be endorsed]*	*In the past 7 days, how much did your sound sensitivities interfere with... Your ability to interact with other people?* ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	29.3%
	In the past 7 days, how much did your sound sensitivities interfere with... Your ability to be productive at work or school? ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	25.6%
	In the past 7 days, how much did your sound sensitivities interfere with... Your ability to take care of your household responsibilities? ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	22.9%
	In the past 7 days, how much did your sound sensitivities interfere with... Your ability to participate in community activities (for example, festivities, religious, or other activities)? ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	24.7%
	To what degree have your sound sensitivities negatively impacted your mental or emotional health? ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	41.7%
	To what degree do you believe that your sound sensitivities have created problems for you? ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	43.4%
	To what degree do you believe that your sound sensitivities have made your entire life worse? ["A moderate amount" / "Very much" / "An extreme amount"]	DVMSQ	31.5%

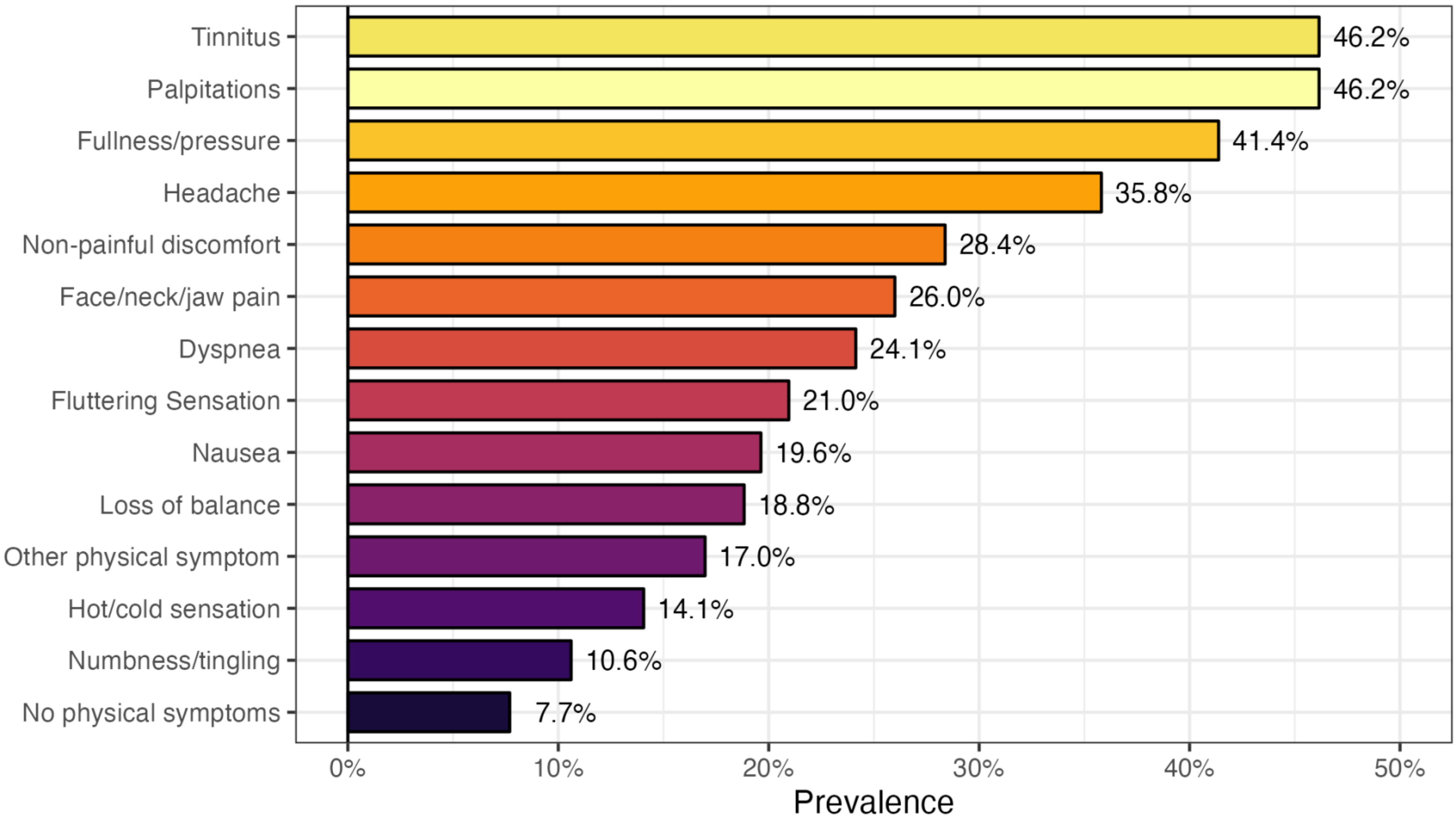
"Novel" indicates a newly written item. NHIS=2014 National Health Interview Survey[42]; ESIT-SQ=European School for Interdisciplinary Tinnitus Research Screening Questionnaire[43]; IHS=Inventory of Hyperacusis Symptoms[44,45]; DVMSQ=Duke-Vanderbilt Misophonia Screening Questionnaire[46]. Non-adapted questionnaire items are reprinted with permission from the copyright holders.

Table 2: Demographic and Clinical Characteristics of the Sample and Hyperacusis/Misophonia Subgroups

	Full Sample (*n*=936)	Hyperacusis (*n*=377)	Misophonia (*n*=332)	Hyperacusis + Misophonia (*n*=253)
Age (years)	37.49 (13.28)	37.49 (12.30)	37.19 (12.40)	37.41 (12.36)
Male	346 (37.0%)	84 (22.3%)	71 (21.4%)	52 (20.6%)
Female	590 (63.0%)	293 (77.7%)	261 (78.6%)	201 (79.4%)
Gender minority (transgender/non-binary)	122 (13.0%)	72 (19.1%)	74 (22.3%)	59 (23.3%)
Hispanic/Latino ethnicity	67 (7.2%)	31 (8.2%)	27 (8.1%)	21 (8.3%)
Race [“choose all that apply” format]				
White	854 (91.2%)	346 (91.8%)	307 (92.5%)	233 (92.1%)
American Indian/Alaska Native	57 (6.1%)	32 (8.5%)	30 (9.0%)	25 (9.9%)
Asian	38 (4.1%)	15 (4.0%)	16 (4.8%)	12 (4.7%)
Black/African American	40 (4.3%)	18 (4.8%)	17 (5.1%)	16 (6.3%)
Middle Eastern/North African	8 (0.85%)	4 (1.1%)	5 (1.5%)	4 (1.6%)
Native Hawaiian/Pacific Islander	4 (0.43%)	3 (0.8%)	3 (0.9%)	3 (1.2%)
Other Race	17 (1.8%)	8 (2.1%)	5 (1.5%)	4 (1.6%)
Educational Attainment				
No high school diploma	15 (1.6%)	6 (1.59%)	6 (1.81%)	5 (1.98%)
High school diploma/general education development	123 (13.1%)	35 (9.28%)	36 (10.8%)	23 (9.09%)
Vocational certificate	34 (3.63%)	20 (5.31%)	13 (3.92%)	11 (4.35%)
Some college	217 (23.2%)	100 (26.5%)	93 (28.0%)	73 (28.9%)
Associate degree	96 (10.3%)	46 (12.2%)	34 (10.2%)	30 (11.9%)
Bachelor's degree	221 (23.6%)	80 (21.2%)	71 (21.4%)	53 (20.9%)
Some graduate/professional school	53 (5.66%)	24 (6.37%)	19 (5.72%)	15 (5.93%)
Graduate/professional degree	177 (18.9%)	66 (17.5%)	60 (18.1%)	43 (17.0%)
Age of autism diagnosis (years)	25.43 (16.45)	26.55 (14.95)	26.21 (15.12)	26.41 (14.94)
Psychiatric diagnoses (other than autism)	1.78 (1.40)	2.13 (1.43)	2.18 (1.44)	2.25 (1.44)
Functional somatic syndrome diagnoses	0.67 (1.14)	0.97 (1.33)	0.97 (1.32)	1.03 (1.37)
Putative risk factors for sound tolerance disorders				
Tinnitus	408 (43.6%)	211 (56.0%)	183 (55.1%)	146 (57.7%)
Tinnitus disorder	307 (32.8%)	178 (47.2%)	158 (47.6%)	130 (51.4%)
Hearing loss (diagnosed)	72 (7.69%)	31 (8.22%)	23 (6.93%)	18 (7.11%)
Sudden hearing loss (not necessarily diagnosed)	61 (6.52%)	33 (8.75%)	29 (8.73%)	22 (8.7%)
Migraine (at least one episode per week)	142 (15.2%)	81 (21.5%)	73 (22.0%)	61 (24.1%)
Traumatic brain injury (with loss of consciousness)	220 (23.5%)	107 (28.4%)	96 (28.9%)	74 (29.2%)
Ruptured eardrum	89 (9.51%)	41 (10.9%)	36 (10.8%)	31 (12.3%)
History of pressure equalization tubes	125 (13.4%)	46 (12.2%)	38 (11.4%)	29 (11.5%)

Continuous/count variables are presented as *M* (*SD*), whereas other variables are presented as *n* (%) within each group. All data were self-reported on the study-specific demographics survey, with the exception of age of autism diagnosis, which was gathered from SPARK demographics. Functional somatic syndromes refer to diagnoses such as fibromyalgia, irritable bowel syndrome, interstitial cystitis, and myalgic encephalomyelitis/chronic fatigue syndrome.

Figure 1: Sound-evoked Symptoms in Hyperacusis Group (*n*=377)



Symptoms were endorsed in response to the question *"When you hear a loud sound, do you ever experience any of the following physical sensations? (Select all that apply).*" Symptom names are shortened versions of the names participants endorsed (e.g., "dyspnea" was "Sensations of shortness of breath or smothering").

Table 3: Endorsement of Common Misophonia Triggers in the Misophonia Group

Duke Misophonia Questionnaire Trigger Category	Endorsement [*n* (%)]
People talking in the background (e.g., phone calls in public, many people talking at once)	256 (77.1%)
People making mouth sounds while eating or drinking (e.g., chewing, crunching, slurping)	235 (70.8%)
People making repetitive sounds (e.g., typing, tapping nails on table, pen clicking, writing, construction work, using machinery)	228 (68.7%)
People making mouth sounds when not eating (e.g., making the "tsk" sound, heavy breathing, snoring, whistling)	213 (64.2%)
People making nasal/throat sounds (e.g., sniffing, sneezing, nose-whistling, coughing, throat-clearing)	199 (59.9%)
Repetitive or continuous sounds not made by a person (e.g., clock ticking, air conditioner humming, water running)	199 (59.9%)
Muffled sounds (e.g., voices separated by a wall, TV/music in another room)	177 (53.3%)
Animals making repetitive sounds (e.g., licking, chirping, barking, eating, drinking)	177 (53.3%)
Rubbing sounds (e.g., hands on pants, hands against one another, styrofoam rubbing together)	151 (45.5%)
Stomping or loud walking (e.g., heels clicking, flip flops, etc.)	147 (44.3%)
Speech sounds (e.g., "p" sounds, hissing "s" sounds, someone speaking with a lisp, high-pitched voices)	138 (41.6%)
Rustling or tearing objects (e.g., paper, plastic)	137 (41.3%)
Seeing someone making or about to make a sound that bothers you, even if you can't hear it (e.g., seeing someone reach into a bag of chips, seeing someone eating on TV with the volume off)	129 (38.9%)
Body or joint sounds (e.g., snapping fingers, cracking joints, jaw clicking)	112 (33.7%)
Other (please describe)	46 (13.9%)

Items are reprinted from the Duke Misophonia Questionnaire and Duke-Vanderbilt Misophonia Screening Questionnaire Trigger Modules[51,72] and used under a CC-BY-SA 4.0 license.

Decreased Sound Tolerance Disorders (Hyperacusis and Misophonia) in Cognitively Able Autistic Adults I: Prevalence and Clinical Features

Zachary J. Williams *et al.* (2024)

Supplemental Information

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Supplemental Methods

Participants

Independent autistic adults above the age of 18 years (all of whom were their own legal guardians, and all of whom self-reported professional diagnoses of DSM-5 autism spectrum disorder or equivalent previously-used diagnoses such as Asperger's disorder) were recruited during Spring 2021 from the Simons Foundation Powering Autism Research for Knowledge (SPARK; The SPARK Consortium, 2018) cohort using the SPARK Research Match service (Project No. RM0111Woynaroski_DST). Of note, authors ZJW and TGW are SPARK participants, and they and their families were excluded *a priori* from study participation. Importantly for the assessment of DST prevalence, the study was not advertised as a specific study of hearing disorders or "sound sensitivity" and was instead given a vague title (*"Investigating Relationships Between Sensory Sensitivities, Health, and Quality of Life in Autistic Adults")* to ideally appeal to a wider range of autistic adult participants. Notably, the validity of self-reported autism diagnoses in the SPARK cohort is high (Fombonne et al., 2022). Although we did not independently assess whether adults in the current cohort had current or historical diagnoses of intellectual disability, SPARK records indicated that for those with data on the variable indicating "*Intellectual disability, cognitive impairment, global developmental delay, or borderline intellectual functioning*" (an even broader category than intellectual disability; data available for 92.3% of current sample), only 4.97% of individuals reported a positive history of any of these conditions. Thus, combined with prior work in a similar SPARK cohort suggesting that over 95% of independent adults do not report prior diagnoses of intellectual disability (Williams, Everaert, et al., 2021), it is reasonable to assume that the overwhelming majority of autistic adults in the current sample (i.e., over 90%) lacked intellectual impairment. Due to DST being dependent on the audibility of the sound stimulus, individuals with self-reported cochlear implants (as a proxy for severe/profound hearing loss) were excluded from the sample, but we still allowed individuals with mild or moderate hearing loss to participate in the study. Notably, a very small minority of participants (*n*=28; 2.99% of the final sample) reported using hearing aids, and hearing aid status was not significantly related to hyperacusis or misophonia classification.

Participants completed a series of online surveys assessing demographics; medical, audiological, and psychiatric history; core features of autism; DST symptoms; co-occurring psychopathology; somatic symptom burden; and quality of life. Participants received a $10 USD Amazon gift code for the completion of all surveys. A total of 1271 individuals consented to participate in the study, 936 of whom were included in the current analyses (894 with no missing data). Data underwent rigorous quality checks, and survey participants were excluded from analyses if they: (a) met the SPARK definition of a possibly invalid autism diagnosis (e.g., age of diagnosis reportedly below 1 year of age; diagnosis rescinded by a professional; *n*=134), (b) did not self-report a professional diagnosis of autism on the study-specific demographics form (*n*=45), (c) reported demographic variables (e.g., age, sex at birth, receipt of special education services in childhood) that were inconsistent with those originally reported to SPARK (*n*=34), (d) reported the use of a cochlear implant (*n*=3), or (e) endorsed a professional diagnosis of either Alzheimer's disease or dissociative identity disorder on two "infrequency" questions within the psychiatric history (indicating either careless/random responding or a true diagnosis that could compromise the validity of self-report; *n*=23). In addition, individuals who dropped out of the study before completing all relevant DST questionnaires (*n*=108) were not included in the current analyses.

Supplemental Results

Clinical Features of Hyperacusis

Individuals with pain hyperacusis typically rated their level of impairment as more severe than those without pain hyperacusis on average (19.4% "*a moderate problem*", 33.3% "*a big problem*", and 47.3% *"a very big problem")*. The average severity of their pain was rated as 60.0 points (*SD*=17.1) on a 100-point visual analog scale. Within the hyperacusis group, there was an inconclusive association between pain hyperacusis and misophonia (74.2% [Pain hyperacusis] vs. 64.8% [Loudness hyperacusis]; OR=1.54, $CrI_{95\%}$ [0.92, 2.60], P_d=.954, BF_{ROPE}=0.340). Pain hyperacusis was also not meaningfully associated with tinnitus (55.9% vs. 56.0%; OR=0.998, $CrI_{95\%}$ [0.629, 1.59], P_d=.503, BF_{ROPE}=0.053), tinnitus disorder (49.5% vs. 46.5%; OR=1.13, $CrI_{95\%}$ [0.721, 1.81], P_d=.701, BF_{ROPE}=0.066), or hearing loss (6.5% vs. 8.8%; OR=0.774, $CrI_{95\%}$ [0.310, 1.83], P_d=.722, BF_{ROPE}=0.171).

Autistic adults with hyperacusis also endorsed a wide range of other sound-evoked symptoms, with the most common non-painful symptoms being tinnitus, palpitations, and ear fullness/pressure (each endorsed by over 40% of the sample). Most autistic adults with hyperacusis (72.1%) also reported experiencing temporary symptom "flare-ups" or "setbacks" caused by loud sounds at some point in their lives. Hyperacusis was further reported to be exacerbated by a wide range of non-sound factors in the majority of autistic adults, including *stress* (88.9%), *feeling tired/lack of sleep* (77.7%), *headache* (69.2%), *experiencing strong emotions* (65.8%), *uncomfortable non-auditory stimuli* (65.5%), and *pain elsewhere in my body* (50.4%). Autistic adults reported that their hyperacusis symptoms were exacerbated by a median of 5 (IQR [3, 6]) of the 7 pre-specified non-sound factors.

The onset of hyperacusis was predominantly reported to be in early childhood, with 226 individuals (59.9%) reporting an onset before 6 years of age and an additional 46 individuals (12.2%) reporting onset between 6–12 years. Most of the remaining individuals (n=76 [20.2%]) indicated that they did not know the onset of their hyperacusis, consistent with a lack of sudden or recent onset of symptoms in these individuals. Individuals reporting onset in the teenage years (3.5%) or in adulthood (4.2%) were relatively uncommon in the sample. When assessing the laterality of hyperacusis, only three cases of unilateral hyperacusis were reported (0.9%), although 23.6% of participants reported asymmetric symptoms favoring one ear over the other; responses indicated that asymmetric symptoms were worse in the left and right ears approximately evenly across participants. Strikingly, participants who reported a diagnosed hearing loss were substantially more likely to indicate that their hyperacusis symptoms were not symmetric (OR=4.91, $\mathrm{CrI}_{95\%}$ [2.29, 11.03], P_d>.999, BF_{ROPE}=150).

Clinical Features of Misophonia

Participants with misophonia reported a median of 7 triggers (IQR [5, 10]), with the most frequently endorsed trigger being "*People talking in the background (e.g., phone calls in public, many people talking at once)*" (77.1% endorsement), followed by *"People making mouth sounds while eating or drinking (e.g., chewing, crunching, slurping)*" (70.8% endorsement), and *"People making repetitive sounds (e.g., typing, tapping nails on table, pen clicking, writing, construction work, using machinery)"* (68.7% endorsement). When considering *any* oronasal trigger (endorsement of either "*mouth sounds – eating,*" "*mouth sounds – not eating,"* or "*nasal/throat sounds"* as a trigger), 85.5% of the misophonia group reported experiencing these "classic" triggers. Similarly, another compound trigger "classically" associated with misophonia (*repetitive* sounds, made up of "*people making repetitive sounds,*" "*objects making repetitive* *sounds,"* and "*animals making repetitive sounds*") was endorsed by 88% of the misophonia group, and 94.9% of individuals with clinically significant misophonia reported at least one oronasal or repetitive trigger sound on the DMQ.

Among autistic adults with misophonia, 93.7% reported experiencing trigger sounds at least once per week over the past month, with 55.1% indicating that they hear trigger sounds multiple times daily. On a 0–100 visual analog scale, autistic adults with misophonia noted that the overall impact of misophonia triggers on their life was a mean of 62.2 (*SD*=23.2). Regarding the emotions encountered when triggered, intense irritation/annoyance was most common (mean item score 3.64/4), followed by anger (mean item score 2.66/4), disgust (mean item score 2.39/4), and least commonly, fear/panic (mean item score 2.06/4). Physical responses when triggered included becoming rigid/stiff (mean item score 2.35/4), reflexively jumping (mean item score 2.24/4), feeling one's heart race (mean item score 2.19/4), breathing more intensely (mean item score 1.89/4), and trembling/shaking (mean item score 1.64/4). Notably, 75.3% of individuals with misophonia felt that their reactions when triggered were "*often*" or "*very often*" excessive, unreasonable, or out of proportion to how most other people would respond. Additionally, 72.6% of the misophonia group reported that they "*often*" or "*very often*" proactively avoid situations where they may potentially hear trigger sounds.

Supplemental Discussion

Clinical Features

Autistic adults also endorsed many different *non-sound* factors that appeared to frequently exacerbate hyperacusis symptoms, with stress, tiredness/lack of sleep, headache, strong emotions, and aversive non-sensory stimuli all causing such exacerbations in 65% or more of the hyperacusis sample. These appear to be similar to factors that are associated with hyperacusis symptom provocation in the general population (Williams, Suzman, et al., 2021), although non-auditory sensory stimuli worsening hyperacusis may be a factor that is more commonly or uniquely observed in autism and should be potentially examined further.

Similar to findings in the general population (Williams, Suzman, et al., 2021), the vast majority of hyperacusis seen in autism was bilateral and symmetric, suggesting a "site of lesion" in the central, rather than peripheral, auditory system. However, unilateral hyperacusis was reported by a small number of individuals (n=3), and a sizable minority of those with bilateral hyperacusis felt that their loudness tolerance was asymmetric (with the proportion noting a right vs. left ear symptom predominance approximately equal). Notably, this finding of asymmetric hyperacusis was strongly associated with a physician diagnosis of prior hearing loss, which is consistent with the associations between hyperacusis and asymmetric hearing loss found by Jahn and Polley (2023). It is likely that in such cases, one ear is perceived to tolerate loud sounds better because of additional cochlear damage on one side, rather than differential "gain" applied to each of the two ears. Thus, although asymmetric hyperacusis is indeed a genuine finding in both autistic and non-autistic adults (and may indeed be reflected in inter-aural asymmetries in loudness discomfort levels), these asymmetries are possibly driven by differences in (peripheral) hearing sensitivity rather than (central) loudness growth *per se*.

Supplemental Table S1: Items from Ad-hoc Clinical Questionnaire Used for Descriptive Phenotyping

Construct	Item Content	Response Options	Source Measure
Tinnitus	Tinnitus refers to the perception of noise in your head or ears (such as ringing or buzzing) in the absence of any corresponding source of sound external to your head. Over the past year, have you had tinnitus in your head or in one or both ears that lasts for more than five minutes at a time?	Yes, most of the time / Yes, a lot of the time / Yes, some of the time / No, not in the past year / No, never	ESIT-SQ [Adapted]
Sound-induced ear pain	Over the past twelve (12) months, on average, approximately how often have you experienced physical pain in one or both ears as a result of sounds in your environment?	Never / Less than once per month / One to three times per month / One to three times per week / Four to seven times per week / Two to five times per day / Six or more times per day	Novel
Sound-induced pain severity	Please rate the severity of your sound-induced pain over the past month, on average.	0–100 Visual Analog Scale	Novel
Additional symptoms of hyperacusis	When you hear a loud sound, do you ever experience any of the following physical sensations? *(Select all that apply)*	A fluttering sensation in your ear(s) / Other non-painful uncomfortable sensations in your ear(s) / A migraine or other headache / Pain in your neck, face, or jaw / Tinnitus (ringing or buzzing noise in your ears) / Loss of balance or postural control / Palpitations, pounding heart, or accelerated heart rate / Sensations of shortness of breath or smothering / Nausea or abdominal distress / Numbness or tingling sensations / Chills or hot flushes / Other physical sensations / None of the above	Novel
Presence of flare-ups or “setbacks”	When you hear a loud sound, does it ever cause your sound sensitivity[a] to temporarily worsen (i.e., sounds become louder, more painful, or more difficult to tolerate)?	Yes / No	Novel

Non-sound causes of flare-ups	Does your sound sensitivity[a] ever get worse (i.e., sounds become even louder or more difficult to tolerate) in any of the following situations?	When you are tired or did not sleep well / When you are experiencing a strong emotion / When you are feeling stressed / When you are experiencing pain somewhere else in your body / When you are experiencing discomfort from one of your other senses (sight, touch, smell, etc.) / When you are under the influence of drugs or alcohol / When you have a headache (including migraines) / Other / None of the above	Novel
Hyperacusis onset	At what age did your sound sensitivity[a] start?	Before age 6 / Age 6–12 / Age 13–17 / Age 18 or older / Don't know	Novel
Hyperacusis laterality	Are you sensitive to sound in one ear or both ears?	Left ear only / Right ear only / Both ears, but worse on left / Both ears, but worse on right / Both ears, same on both sides / Don't know	Novel
Received medical care for DST	Have you seen a doctor or other health care professional (e.g., an audiologist, psychologist, counselor, or occupational therapist) for help with problems tolerating sounds in your environment?	Yes / No	Novel

[a] Items were only administered to individuals who screened positive for hyperacusis and were embedded in a series of questions on hyperacusis (i.e., reduced tolerance to loud sounds and pain from sound). Thus, despite the vague language, these questions are expected to refer primarily to hyperacusis rather than sound tolerance symptoms in general.

Supplemental References

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