Author Manuscript
Author Manuscript
Author Manuscript

HHS Public Access

Author manuscript

Ear Hear. Author manuscript; available in PMC 2022 September 01.

Published in final edited form as:

Ear Hear. 2021 ; 42(5): 1137–1150. doi:10.1097/AUD.0000000000001005.

Prevalence of Decreased Sound Tolerance (Hyperacusis) in Individuals with Autism Spectrum Disorder: A Meta-analysis

Zachary J. Williams, B.S.[1,2,3,4], **Evan Suzman, B.S.**[5], **Tiffany G. Woynaroski, Ph.D., CCC-SLP**[2,3,4,6]

[1]Medical Scientist Training Program, Vanderbilt University School of Medicine, Nashville, TN

[2]Department of Hearing and Speech Sciences, Vanderbilt University Medical Center, Nashville, TN

[3]Vanderbilt Brain Institute, Vanderbilt University, Nashville, TN

[4]Frist Center for Autism and Innovation, Vanderbilt University, Nashville, TN

[5]Graduate Program in Biomedical Sciences, Vanderbilt University, Nashville, TN

[6]Vanderbilt Kennedy Center, Vanderbilt University Medical Center, Nashville, TN

Abstract

Objective: Hyperacusis, defined as decreased tolerance to sound at levels that would not trouble most individuals, is frequently observed in individuals with autism spectrum disorder (ASD). Despite the functional impairment attributable to hyperacusis, little is known about its prevalence or natural history in the ASD population. The objective of this study was to conduct a systematic

Correspondence addressed to: Zachary J. Williams, 1215 21st Avenue South, Medical Center East, Room 8310, Nashville, TN 37232, (805)-729-6691, Zachary.j.williams@vanderbilt.edu.

Author Contributions

ZJW conceptualized the study, performed the literature search, extracted the data, performed and interpreted all statistical analyses, and drafted the manuscript. ES assisted in performing the literature search, extracting data, and critically revising the manuscript. TGW supervised the literature search, provided methodological guidance, and critically revised the manuscript. All authors have read, approved of, and taken responsibility for the final version of the manuscript.

Disclosures

ZJW serves on the family advisory committee of the Autism Speaks Autism Treatment Network site at Vanderbilt University and the autistic researcher review board of the Autism Intervention Network on Physical Health (AIR-P). He has also received consulting fees from Roche. The remaining authors have nothing to declare. No funding organization or source of support had a role in the study design, data collection, analysis, or interpretation, decision to publish, or preparation of this manuscript.

Data Access, Responsibility, and Analysis

No original data was generated in this study. Study-level data used in the meta-analysis is presented in the online supplement (Supplemental Tables S1-S2). Additional study materials, including data extraction protocols and code for statistical analyses are available from the first author upon request.

Additional References Cited in Meta-analysis (to be included in bibliography)

(Albores-Gallo et al., 2012; Allison et al., 2008; Azouz, Khalil, et al., 2014; Azouz, Kozou, et al., 2014; Bennett et al., 2019; Bhatara et al., 2013; Billstedt et al., 2007; Bishop et al., 2006, 2013; Canal-Bedia et al., 2011; Carrington et al., 2014, 2019; Cervantes et al., 2017; Chaste et al., 2015; Dai et al., 2020; Danesh et al., 2015; de Giambattista et al., 2019; Demopoulos & Lewine, 2016; Downs et al., 2005; Egelhoff, 2011; Gomes et al., 2004; Grapel et al., 2015; Green et al., 2016; Grzadzinski et al., 2016; Guo et al., 2019; Hall et al., 2016; Hattier et al., 2012; Hussein et al., 2019; Inada et al., 2011; Jussila et al., 2020; Kamio et al., 2015; Kara et al., 2014; Kientz & Dunn, 1997; Kim et al., 2016; Klintwall et al., 2011; Kopecky et al., 2013; Kozlowski et al., 2012; Law et al., 2016; Levitin et al., 2005; Lord et al., 1997; Matheis et al., 2019; Matson et al., 2013, 2017; Mercati et al., 2017; Nupur et al., 2016; Pahan, 2003; Porges et al., 2014; Richler et al., 2007; Robins et al., 2001; Rosenhall et al., 1999; Shardell, 2013; Silva & Schalock, 2012; Sipes et al., 2011; Srisinghasongkram et al., 2016; Stein et al., 2013; Stenberg et al., 2014; Tan et al., 2012; Tavassoli et al., 2016, 2019; Tharpe et al., 2006; Tomchek, 2005; Tomchek & Dunn, 2007; Troyb et al., 2014; Ventola et al., 2007; VerMaas-Lee, 1999; Wong et al., 2018)

review and meta-analysis estimating the current and lifetime prevalence of hyperacusis in children, adolescents, and adults with ASD. By precisely estimating the burden of hyperacusis in the ASD population, the current study aims to enhance recognition of this particular symptom of ASD and highlight the need for additional research into the causes, prevention, and treatment of hyperacusis in persons on the spectrum.

Design: We searched PubMed and ProQuest to identify peer-reviewed articles published in English after January 1993. We additionally performed targeted searches of Google Scholar and the gray literature, including studies published through May 2020. Eligible studies included at least 20 individuals with diagnosed ASD of any age and reported data from which the proportion of ASD individuals with current and/or lifetime hyperacusis could be derived. In order to account for multiple prevalence estimates derived from the same samples, we utilized three-level Bayesian random-effects meta-analyses to estimate the current and lifetime prevalence of hyperacusis. Bayesian meta-regression was used to assess potential moderators of current hyperacusis prevalence. In order to reduce heterogeneity due to varying definitions of hyperacusis, we performed a sensitivity analysis on the subset of studies that ascertained hyperacusis status using the Autism Diagnostic Interview-Revised, a structured parent interview.

Results: A total of 7783 nonduplicate articles were screened, of which 67 were included in the review and synthesis. Hyperacusis status was ascertained in multiple ways across studies, with 60 articles employing interviews or questionnaires and 7 using behavioral observations or objective measures. The mean (range) age of samples in the included studies was 7.88 (1.00-34.89) years. The meta-analysis of interview/questionnaire measures ($k_{(3)}$ = 103, n_{ASD}=13093) estimated the current and lifetime prevalence of hyperacusis in ASD to be 41.42% (95% CrI [37.23, 45.84]) and 60.58% [50.37, 69.76], respectively. A sensitivity analysis restricted to prevalence estimates derived from the ADI-R ($k_{(3)}$ = 25, n_{ASD} = 5028) produced similar values. The estimate of current hyperacusis prevalence using objective/observational measures ($k_{(3)}$= 8, n_{ASD} = 488) was 27.30% [14.92, 46.31]. Heterogeneity in the full sample of interview/questionnaire measures was substantial but not significantly explained by any tested moderator. However, prevalence increased sharply with increasing age in studies using the ADI-R (BF_{10} = 93.10, R^2_{Het} = 0.692).

Conclusions and Relevance: In this meta-analysis, we found a high prevalence of current and lifetime hyperacusis in individuals with ASD, with a majority of individuals on the autism spectrum experiencing hyperacusis at some point in their lives. The high prevalence of hyperacusis in individuals with ASD across the lifespan highlights the need for further research on sound tolerance in this population and the development of services and/or interventions to reduce the burden of this common symptom.

Introduction

Hyperacusis is a hearing disorder involving an increased sensitivity to or decreased tolerance of sound at levels that would not trouble most individuals (Fackrell et al., 2019). Individuals experiencing hyperacusis report that everyday sounds can be unpleasant, intense, frightening, painful, or overwhelming and can cause significant anxiety and distress (Fackrell et al., 2019; Jastreboff & Jastreboff, 2015; Tyler et al., 2014). In a pivotal review on this topic, Tyler et al. (2014) suggested that hyperacusis be divided into four subtypes based on clinical presentation: loudness hyperacusis, pain hyperacusis, annoyance hyperacusis, and fear hyperacusis. Notably, a more recent consensus definition of hyperacusis (Fackrell et al., 2019) states that this condition is distinct from misophonia (an anger-predominant reaction to specific sounds such as chewing) and phonophobia (a specific phobia of certain sounds or categories of sounds), which correspond approximately to Tyler's categories of "annoyance hyperacusis" and "fear hyperacusis," respectively. Thus, within the current study, hyperacusis refers specifically to *loudness hyperacusis*, wherein sounds of moderate intensity are perceived as excessively loud, and *pain hyperacusis*, wherein sounds evoke physical pain in the ear or head at levels far below those needed to cause pain in a typical listener (i.e., approximately 120 dB SPL). Hyperacusis can have profound effects on the lives of those who experience it, negatively affecting social and occupational functioning, emotional well-being, hearing, sleep, and concentration (Tyler et al., 2014).

The prevalence of hyperacusis in the general population is variable, depending both on the definition of hyperacusis used in a given study and the specific methods used to determine case status. Based on data from the 2014 National Health Interview Survey, an estimated 5.9% of U.S. adults report sensitivity to everyday sounds based the following question: "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 annoying to you?" (Zelaya et al., 2015). Additionally, in a large, population-based Swedish study, self-reported sound intolerance was endorsed by 9% of adults, and approximately 2% had been diagnosed with hyperacusis by a physician (Paulin et al., 2016). Hyperacusis in adults is also associated with a range of other disorders, including tinnitus, hearing impairment, traumatic brain injury, migraine, anxiety disorders, mood disorders, and functional somatic syndromes such as fibromyalgia and irritable bowel syndrome (Assi et al., 2018; Cederroth et al., 2020; Paulin et al., 2016; Sheldrake et al., 2015). Hyperacusis has been less well characterized in the general pediatric population (Potgieter et al., 2020), although a systematic review of prevalence studies in children and adolescents reported a range of prevalence rates from 3.2–17.1% (Rosing, Schmidt, et al., 2016). Pediatric hyperacusis appears to be more common in younger children, and the overwhelming majority of clinically-referred children are under 10 years of age (Amir et al., 2018; Baguley et al., 2013; Myne & Kennedy, 2018; Rosing, Kapandais, et al., 2016). Compared to adults, children with hyperacusis are substantially less likely to present with comorbid hearing loss or tinnitus and much more likely to have comorbid neurodevelopmental conditions such as autism spectrum disorder (ASD) or attention deficit-hyperactivity disorder (Amir et al., 2018; Hall et al., 2016; Myne & Kennedy, 2018; Potgieter et al., 2020). Notably, there has been little research on the developmental trajectory of hyperacusis across the lifespan, and it remains unknown how many individuals with childhood-onset hyperacusis continue to experience symptoms into adulthood.

Research on hyperacusis is still in its infancy; the condition remains medically unexplained with no definitive etiology or pathophysiology (Pienkowski et al., 2014; Radziwon et al., 2020; Roberts & Salvi, 2019), and no evidence-based recommendations exist to guide its diagnosis or treatment. While there are many unresolved questions in hyperacusis research (Baguley & Hoare, 2018; Fackrell et al., 2019), one area of particular interest is the overlap of hyperacusis with ASD (Fackrell et al., 2019). In addition to the social communication impairments and restricted, repetitive behaviors characteristic of ASD, many individuals on the autism spectrum exhibit hyper- or hyporeactivity to sensory stimuli, now considered a core feature of the condition in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; American Psychiatric Association, 2013; Ben-Sasson et al., 2009, 2019; Hazen et al., 2014). Among the sensory manifestations of ASD, decreased sound tolerance is common (Gomes et al., 2008; O'Connor, 2012; Stiegler & Davis, 2010), with studies reporting prevalence rates as high as 86% in this population (Hussein et al., 2019; Law et al., 2016). Furthermore, of 412 children seeking specialist care for hyperacusis in a single-center study, 60% had diagnoses of ASD (Amir et al., 2018). In children with ASD, hyperacusis is associated with reduced participation in family, school, and community activities, challenging behaviors, increased caregiver burden, and safety concerns for both children and family members (Hussein et al., 2019; Law et al., 2016). This symptom can also persist into adulthood (Kuiper et al., 2019; Landon et al., 2016; Robertson & Simmons, 2015; Tavassoli et al., 2014), contributing to workplace difficulties (Hayward et al., 2019; Hedley et al., 2018; Lorenz et al., 2016; Robertson & Simmons, 2015), anxiety (Landon et al., 2016; Simonoff, 2020), avoidance behavior (Landon et al., 2016), and general distress (Griffith et al., 2011; Jones et al., 2003; Smith & Sharp, 2013). While it remains unknown whether the hyperacusis observed in ASD is phenomenologically or physiologically distinct from hyperacusis in other populations, this symptom contributes substantially to the disability associated with ASD and may serve as a worthwhile target for clinical intervention.

Although decreased sound tolerance has been recognized as part of the ASD phenotype since Kanner's first description of the condition (Kanner, 1943), relatively few studies have explicitly attempted to assess the proportion of individuals with ASD with hyperacusis. In one of the only reviews on the topic, Gomes and colleagues (2008) cited prevalence estimates between 15–100% based on an unsystematic literature search. Another commonly cited figure is that of Rimland and Edelson (1995), who reported that 40% of children with ASD exhibit "some symptoms of sound sensitivity" based on an unpublished sample of over 17,000 parent questionnaires. A more recent online survey study by Law and colleagues (2016) reported the current and lifetime prevalence of "auditory hypersensitivity" to be 77.6% and 86.6%, respectively, in a sample of 814 children with ASD. Prevalence estimates tend to be lower when hyperacusis is assessed using psychoacoustic tests and behavioral observations, with Rosenhall et al. (1999) reporting a prevalence of 18% based on the inability to behaviorally tolerate an 80 dB nHL broadband click and Demopoulos & Lewine (2016) reporting a hyperacusis prevalence of 37% based on speech LDLs three standard deviations below a control group's mean score. Given the wide-ranging estimates of hyperacusis prevalence in the ASD literature, there is a substantial need for more information regarding the prevalence of this particular symptom in persons on the autism spectrum. More precise estimates regarding hyperacusis prevalence in this population will provide crucial background information for adequate medical and audiological evaluation and service delivery. Additionally, such estimates are necessary to accurately define the burden of hyperacusis in the ASD population, increasing the recognition of this condition among the research community and providing a justification for policymakers and funding agencies to allocate additional resources to understanding and ameliorating this reportedly common symptom (Gillum et al., 2011).

Although there has been no quantitative synthesis of the ASD hyperacusis prevalence literature to date, estimates of hyperacusis prevalence in individuals with ASD are present in a large number of published studies. Questions assessing decreased sound tolerance are included in several widely used clinical tools used to assess ASD (Lord et al., 1994; Robins et al., 2001, 2014; Tomchek & Dunn, 2007), and item-level data for these measures is often reported, even in cases when hyperacusis is not the focus of a given study. Thus, any study reporting item-level data for any of these measures can be utilized as an estimate of the prevalence of hyperacusis in ASD. However, clinical questionnaires and interviews assess hyperacusis symptoms in variable ways, and these measures differ significantly in the aspects of sound tolerance that they address and the degree to which they are able to differentiate hyperacusis from other forms of decreased sound tolerance, such as misophonia and phonophobia.

One particularly common ASD questionnaire assessing hyperacusis is the Modified Checklist for Autism in Toddlers (M-CHAT; Robins et al., 2001), which consists of 23 Yes/No questions answered by the parent of a young child. The M-CHAT is widely used to screen for ASD in children 18–24 months of age (Yuen et al., 2018), and this tool was recommended by the American Academy of Pediatrics for autism screening in general pediatric practice (Johnson et al., 2007). Notably, item 11 of the M-CHAT assesses decreased sound tolerance in the child, stating, "*Does your child ever seem oversensitive to noise? (e.g., plugging ears).*" A successor of this form, the M-CHAT Revised with Follow-up (M-CHAT R/F; Robins et al., 2014), contains a similar item, "*Does your child get upset by everyday noises? (For example, does your child scream or cry to noise such as a vacuum cleaner or loud music?),*" and affirmative answers to this item are followed up with more detailed questioning regarding (a) the specific sounds that a child reacts to (two or more common noises required for credit) and (b) the specific reactions the child has to these sounds (for the item to be endorsed, child must scream, cry, and/or cover ears in response to noise and do so most of the time). Items assessing hyperacusis are also present on questionnaires that assess sensory features of ASD across multiple modalities, such as the (Short) Sensory Profile (McIntosh et al., 1999; Z. J. Williams, 2020; Z. J. Williams et al., 2018), which contains the item, "*Responds negatively to unexpected or loud noises (for example, cries or hides from vacuum cleaner, dog barking, hair dryer).*" Notably, while multiple self-report hyperacusis questionnaires are available for use in the general population (Fackrell & Hoare, 2018), only one study to date has employed a validated hyperacusis self-report (the Hyperacusis Questionnaire [HQ]; Khalfa et al., 2002) in an ASD sample, with 69% of the sample exceeding the HQ cutoff for hyperacusis (Danesh et al., 2015).

Of the clinical tools commonly used to assess hyperacusis across the ASD research literature, perhaps the most accurate tool is the Autism Diagnostic Review–Revised (ADI-R; Lord et al., 1994), a 111-item semi-structured parent interview covering the areas of reciprocal social interaction, communication and language, and restricted and repetitive behaviors (including sensory abnormalities). Each item is scored separately for current and lifetime presence/severity. One item of the ADI-R (item 72) specifically addresses the presence of hyperacusis, with the experimenter initially asking "*Has [child] ever seemed oversensitive to noise?*" and following up that question with more specific prompts such as: *Has s/he ever deliberately and regularly put her/his hands over her/his ears in response to ordinary sounds? Does s/he do this now? To what kinds of sounds?* and *Have you ever had to adjust what you do because [child] was so upset by noises?* As described in the ADI-R manual (Rutter et al., 2013), the question is specifically designed to assess "predictable, generally increased sensitivity to everyday sounds, such as household appliances or traffic, rather than a reaction to a sudden, harsh, or unexpected noise such as thunder or a loudspeaker." Moreover, this question differentiates hyperacusis from other forms of decreased sound tolerance, as idiosyncratic responses to highly specific sounds (as would occur in misophonia or phonophobia) are excluded from this item and are instead covered by item 73 (which assesses hyper-reactivity to specific sensory stimuli in any modality and includes a prompt assessing misophonia-like features: "*Does [child] ever get unusually upset or irritated by particular sounds such as people coughing or a baby crying?*"). Lastly, in order to receive a score of "1" on item 72 (indicating mild hyperacusis) or greater, the child must exhibit regular negative reactions to sounds (other than sounds that are considered excessively loud by most people) for a period of at least three months. As the ADI-R is considered one of the diagnostic "gold-standard" measures for ASD (Falkmer et al., 2013), data from this interview are frequently reported in the ASD literature, resulting in a large number of high-quality assessment of hyperacusis available for synthesis.

In the current study, we aimed to leverage the large amount of published literature on the prevalence of hyperacusis in ASD to produce quantitatively robust estimates of hyperacusis prevalence in this population. The current investigation is the first meta-analysis of this literature, as well as the first quantitative synthesis of hyperacusis prevalence in any population. In addition to simply calculating the proportion of individuals with current and lifetime hyperacusis, we also sought to assess the factors responsible for the high variability in prevalence estimates across studies. As there are few investigations in the literature systematically examining the relations between hyperacusis prevalence and age, sex, intellectual ability, or other sociodemographic factors in the ASD population, meta-analytic information regarding study-level moderators of hyperacusis could potentially inform research and clinical practice. Thus, the current study employed advanced statistical approaches to provide pooled estimates of hyperacusis prevalence in individuals with ASD and test whether a number of methodological and sociodemographic factors significantly influence prevalence estimates across studies.

Materials and Methods

Search Strategy and Selection Criteria

A systematic review and meta-analysis was conducted in accordance with MOOSE (Stroup et al., 2000) and PRISMA (Moher et al., 2009) guidelines. We searched PubMed and 83 databases available through the ProQuest search engine (e.g., PsycINFO/PsycARTICLES) for publications on ASD that reported on decreased sound tolerance, as defined using a combination of keywords and filters (see Supplemental Methods file for full search procedures). Results were limited to peer-reviewed, English-language studies published between January 1, 1993 and September 29, 2019. Following a previous meta-analysis of ASD comorbidity (Lai et al., 2019), we restricted our analysis to studies conducted after the publication and widespread adoption of ICD-10/DSM-IV, when the definition of ASD was expanded dramatically (Volkmar & McPartland, 2014). Additionally, targeted searches of Google Scholar using terms such as *("autism diagnostic interview" OR "ADI-R") AND ("sensitivity to noise" OR "item 72")* were utilized to identify studies in which item-level data from commonly used ASD clinical measures with indices of hyperacusis were reported. These searches were supplemented with a targeted search of the gray literature, including ProQuest Dissertations and Theses and abstracts from the International Society for Autism Research annual meeting. All articles included in the meta-analysis were then subjected to forward/backward citation tracing using Google Scholar to identify additional relevant studies (last search conducted September 18, 2020). References were managed using Papers software (Readcube, Cambridge, USA).

Author ZJW screened titles/abstracts of all potentially eligible publications, using predetermined criteria to select studies for full-text review. All studies selected at this stage reported cross-sectional data, recruited a sample of at least 20 individuals with ASD, and included some mention of sensory processing, auditory function, or a clinical instrument known to contain items related to decreased sound tolerance. We excluded case reports, review articles, and studies focused exclusively on sensory modalities other than audition. The threshold of $n \geq 20$ was chosen to maximize the number of included studies while still limiting small-sample bias (Borenstein et al., 2009; Lai et al., 2019).

Full-text articles were assessed for inclusion by authors ZJW, with author ES independently reviewing 20% of the articles to evaluate inter-rater reliability of the inclusion criteria (quantified using percent agreement and Cohen's (1960) kappa). All disagreements regarding inclusion were discussed by the two authors until consensus was reached. Included studies met all of the following criteria: (a) they were peer-reviewed journal articles, dissertations/theses, or conference presentations, (b) they included at least 20 individuals with diagnosed ASD, confirmed by clinical assessment, parent report, medical records, or databases with a stated diagnosis, and (c) they reported data from which the proportion of ASD individuals with current and/or lifetime hyperacusis could be derived (see Supplemental Methods file for more details). Studies in which individuals screened positive for ASD but did not receive formal diagnostic evaluations were excluded. Corresponding authors of studies published since 2015 that met criteria for inclusion but did not report the proportion of the sample with hyperacusis (e.g., studies reporting item-level data for ADI-R item 72, but not endorsement frequencies) were contacted via email to request additional data.

Data Extraction and Assessment

Author ZJW extracted data from all articles according to a standardized protocol (available on request). Author ES independently extracted data from 20% of included studies, and reliability of data extraction was assessed using intra-class correlation coefficients (ICC(3,1); Shrout & Fleiss, 1979). For each included study, we extracted n_{ASD} and

$n_{\text{Hyperacusis}}$ for all eligible measures and subgroups. Potential study-level moderators were also extracted, including publication year, sample size (log transformed), measure timeframe (current vs. lifetime prevalence), proportion of females in the sample, proportion of sample with IQ/DQ<70, mean age, and UN's 2019 Human Development Index (HDI; Çilingirtürk & Koçak, 2018) of the study's country of origin. Additional information on each of these potential moderators is presented in the Supplemental Methods.

In addition to the above moderators, we graded all studies on an ad-hoc eight-item measure of study quality derived in part from the criteria used in a previous systematic review of pediatric hyperacusis prevalence (Nemholt et al., 2015; Rosing, Schmidt, et al., 2016) and criteria used to grade ASD sample characterization in a prior meta-analysis by our group (Z. J. Williams, 2020). Studies were assessed on the following eight criteria (see Supplemental Table S1 for full scoring guidelines): (1) age and sex of the ASD sample are reported, (2) cognitive ability of the ASD sample is reported, (3) diagnoses of ASD are confirmed using rigorous criteria, (4) hearing impairment is assessed in the study participants, (5) participants recruited from an epidemiological population or large-scale registry (hence being more representative than a typical clinical or community sample; Lai et al., 2019), (6) method used to define hyperacusis is reported in sufficient detail, (7) method used to define hyperacusis is likely to have relatively high reliability (i.e., an interview or structured observation rather than a single questionnaire item), and (8) the hyperacusis measure in question attempts to differentiate hyperacusis from other forms of decreased sound tolerance (e.g., by focusing on the loudness of the sound as the aversive quality). Overall quality scores were calculated as the mean of all eight items, ranging from 0 to 1 with higher scores reflecting relatively higher study quality. Scores for each item and overall scores for each included study sample are presented in (Supplemental Tables S2).

Statistical Analysis

All statistical analyses were performed in R (R Core Team, 2020). To model the proportion of individuals with hyperacusis in each study, we utilized a binomial-normal meta-analysis model with logit link function (Hamza et al., 2008; Schwarzer et al., 2019; Stijnen et al., 2010). Multiple independent samples of individuals with ASD reported within the same article were treated as separate studies in our model. As multiple effect sizes (e.g., both a current and lifetime prevalence) were often reported for a single study, we utilized a three-level random-effects meta-analysis model (Cheung, 2014; Pastor & Lazowski, 2017; Van den Noortgate et al., 2012), treating effect size (level 3) as a random effect nested within study (level 2). As we chose not to conflate prevalence estimates derived from subjective measures (i.e., questionnaires and interviews) and objective measures (i.e., behavioral observations and psychoacoustic tests), separate meta-analysis models were constructed to analyze prevalence for these two measure categories (see Gomes et al., 2004; Tavassoli et al., 2019 for further discussion about discrepancies between these measures). In addition, we performed a sensitivity analysis by analyzing only the studies that used item 72 of the ADI-R as their measure of hyperacusis prevalence. In addition to the systematic and standardized way that the ADI-R characterizes hyperacusis in ASD, another strength of this sensitivity analysis is the fact that none of the included ADI-R studies had the explicit goal of analyzing hyperacusis prevalence, reducing the chances of ADI-R prevalence estimates being affected by publication bias. The meta-analysis models for the subjective measure category (and the ADI-R sensitivity analysis) also included a fixed effect of measure timeframe (i.e., current vs. lifetime) in order to account for the necessarily higher estimates reported by lifetime measures.

Model estimation was performed in a Bayesian framework using the R package *brms* (Bürkner, 2017, 2018). Weakly informative priors were employed, with a Normal(0,1) prior on logit-transformed regression coefficients (including intercept) and a Half-Cauchy(0.3) prior on the standard deviations of the random intercept terms (D. R. Williams et al., 2018). We utilized the posterior mode and the 95% highest density credible interval (CrI) to summarize parameter distributions. Additional information on model estimation and choice of priors can be found in the Supplemental Methods file.

To assess the heterogeneity of studies in our analysis, we calculated a multilevel I^2 statistic (Higgins & Thompson, 2002), quantifying heterogeneity from the study and effect size levels combined. In addition, we reported the *ICC*(2) coefficient proposed by Cheung (2014), reflecting the proportion of I^2 attributable to between-study (level 2) variance. As a more intuitive measure of study heterogeneity, we also calculated a model-based 95% prediction interval (PI; IntHout et al., 2016). Conditional on the observed data and prior information, there is a 95% chance that a future estimate of hyperacusis prevalence in ASD will lie within the 95% PI, and thus this interval is helpful to visualize the potential distribution of effects to be expected from future studies on this topic.

As the majority of studies in our analysis did not report hyperacusis prevalence as a primary outcome (and in many cases reported it incidentally), we did not expect to encounter substantial publication bias in this body of literature. Furthermore, as a prevalence figure is not typically tested for statistical significance, we felt it unlikely that the estimates in our meta-analysis would be subject to the “significance chasing” publication bias often seen in the biomedical and social sciences (Ware & Munafò, 2015). Nevertheless, we sought to mitigate publication bias in our prevalence estimates by performing a comprehensive gray-literature search and including a large number of unpublished theses and conference abstracts in our analyses. As standard quantitative measures of publication bias have not been validated for use in a three-level meta-analysis framework, we chose to graphically assess publication bias in each meta-analytic model via the examination of funnel plots.

Potential moderators of hyperacusis prevalence were analyzed using Bayesian meta-regression. Moderators of interest included publication year (see Rødgaard et al., 2019), log-transformed sample size (included as a supplementary test of “small-study” publication bias; Sterne et al., 2000), study-level demographic variables (i.e., mean age of the study sample, proportion of females in the sample, and the proportion of individuals in the sample with cognitive abilities in the intellectually disabled range [IQ or DQ < 70]; Lai et al., 2019), 2019 UN HDI, and total study quality score.

We chose only to analyze moderators of *current* hyperacusis prevalence in order to eliminate unmodeled interactions between measure timeframe and other potential moderator variables. Meta-regression analyses were only performed for the subjective prevalence model and the ADI-R only model, as the objective model contained fewer than 15 studies (Borenstein et al., 2009; Lai et al., 2019). Missing moderator values were handled via 10-fold random-forest imputation, implemented using the *missForest* R package (Shah et al., 2014; Stekhoven, 2013; Stekhoven & Bühlmann, 2012). Each meta-regression model was compared to its respective baseline (intercept-only) model using a Bayes factor (BF_{10}) calculated via bridge sampling (Gronau et al., 2017, 2020). BF_{10} values greater than 3 provide significant evidence in favor of the tested moderator effect, whereas values less than 1/3 provide significant evidence *against* that effect, and values between 1/3 and 3 are deemed inconclusive (Wagenmakers et al., 2011). Bayes factors derived from multiply imputed data were defined as the arithmetic mean of the individual Bayes factors computed from each imputed dataset (Hoijtink et al., 2019).

Results

The initial database search identified 10255 records, with an additional 506 records being identified via our gray literature search and forward/backward citation tracing. After removing duplicate entries, titles and abstracts from the remaining 7783 articles were screened to determine eligibility for full-text review. Of these, 641 articles were subjected to full-text review, and 67 were found to meet criteria for inclusion in the study (Supplemental Table S1-S2), including one study for which eligible data were obtained via author contact (Carrington et al., 2019). Inter-rater agreement was excellent for both study inclusion decisions (95% agreement, κ = 0.905) and data extraction [$ICC(3,1) = 1.00$]. A full PRISMA flow diagram is presented in Figure 1.

In total, we extracted 111 estimates of hyperacusis prevalence (103 subjective [25 ADI-R], 8 objective) from 91 independent samples, representing data from 13433 individuals with ASD (see Supplemental Tables S2-S3 for details of included studies). Estimates of hyperacusis prevalence ranged from 4–86% (median 43.75%). Individuals included in these studies were predominantly young children (mean age = 7.88 years), 78.3% were male, and 60.2% had IQ/DQ > 70, although demographics varied widely between studies. Study quality scores varied substantially between studies, ranging from 0.125 to 0.938, with a mean of 0.506 and standard deviation of 0.203 (Supplemental Table S2). Studies using objective measures had slightly higher quality scores ($M = 0.632$, $SD = 0.143$) than studies using subjective measures ($M = 0.496$, $SD = 0.204$), although the subset of subjective studies that used the ADI-R to ascertain hyperacusis status scored highly on our quality metric ($M = 0.780$, $SD = 0.06$).

Based on our meta-analytic models, the pooled prevalence of current hyperacusis was 41.42% (95% CrI [37.23, 45.84]) when assessed with an interview/questionnaire (Table 1; Figure 2) and 27.30% [14.92, 46.31] when assessed using an observational/objective measure (see Supplemental Figure S1 for a graphical representation of the objective model). The pooled lifetime prevalence of hyperacusis in ASD (subjective measures only) was substantially higher at 60.58% [50.37, 69.76]. When restricting analyses to just those studies utilizing the ADI-R, point estimates of both current prevalence (48.78% [42.48, 54.91]) and lifetime prevalence (64.25% [53.81, 72.60]) were slightly higher than the full sample of studies using subjective measures. However, there was inconclusive evidence to suggest that prevalence rates derived from the ADI-R were different from those obtained using other subjective measures (BF_{10} = 0.53, R^2_{Het} = 0.017). All meta-analytic models displayed substantial heterogeneity (I^2 > 90%), resulting in wide 95% PIs (Table 1; see Supplemental Table S4 for additional heterogeneity metrics). Funnel plots were largely symmetric (see Supplemental Figure S2), reflecting minimal publication bias in the included studies.

None of the tested moderator variables significantly influenced the prevalence of current hyperacusis across the full sample of subjective studies (Table 2). While the majority of moderator analyses were inconclusive, Bayes factors provided substantial evidence *against* the moderating role of publication year. Additionally, there was slight evidence *in favor* of a moderating role of age (BF_{10} = 1.528), with 10.1% of the heterogeneity in prevalence estimates attributable to age. Exploratory post-hoc plotting of this relationship revealed an inverted-U shaped trend, with hyperacusis prevalence in ASD increasing from childhood to adolescence, peaking at approximately age 15–16, and declining thereafter (Supplemental Figure S3). Bayes factors were also inconclusive regarding the moderating effects of sex ratio (BF_{10} = 0.404), the proportion of the sample with intellectual disability (BF_{10} = 0.466), and study quality (BF_{10} = 0.462); however, these variables explained negligible amounts of between-study heterogeneity (R^2_{Het} < 0.01), indicating that they were unlikely to be meaningful determinants of hyperacusis prevalence in the ASD population. When analyzing studies reporting current hyperacusis prevalence using the ADI-R (k = 17, n_{ASD} = 3363), the mean age of study participants was a significant moderator of prevalence, explaining the majority of between-study heterogeneity (BF_{10} = 93.10, R^2_{Het} = 0.692). Estimated ADI-R hyperacusis prevalence increased substantially with age, rising from 35.12% at 2 years to 71.50% at 14 years (Figure 3). Bayes factors provided significant evidence against the moderation of ADI-R prevalence estimates by sample size (BF_{10} = 0.25), and all other moderator analyses were inconclusive, including sex ratio, proportion of the sample with intellectual disability, publication year, and study quality.

Discussion

In this study, we performed the first quantitative synthesis of the literature on hyperacusis in ASD, meta-analyzing the prevalence of co-occurring hyperacusis in over 13000 individuals on the autism spectrum. Based on a heterogeneous mixture of interview and questionnaire measures, our meta-analytic models estimated that the current prevalence of hyperacusis in the ASD population is between 37–45%, with approximately 50–70% of individuals with ASD exhibiting signs of hyperacusis at some point in their lives. These estimates were not appreciably altered when only considering studies utilizing the ADI-R (a “gold-standard” structured ASD symptom interview), although the few studies assessing hyperacusis via behavioral observations or objective tests reported modestly lower (current) prevalence rates (15–46%). Remarkably, the current prevalence estimate from our study is very similar to the oft-cited 40% figure reported by Rimland and Edelson (1995). Considering that ASD affects approximately 2% of the American population (Dietz et al., 2020; Maenner et al., 2020), these results suggest that between 2.4–3 million individuals with ASD in the U.S. currently experience symptoms of hyperacusis.

Like other meta-analyses of ASD comorbidity (Lai et al., 2019), the included prevalence estimates in the present study were extremely heterogeneous, with estimated I^2 values greater than 90% in all three of our models and extremely wide 95% PIs. Interestingly, none of the proposed moderator variables were found to explain a significant portion of heterogeneity in (interview/questionnaire) prevalence estimates, suggesting that the vast majority of between-study heterogeneity is not attributable to sample demographics or easily quantified study-level variables. As the diverse questionnaire and interview methods used to ascertain hyperacusis status surely contributed to observed heterogeneity, we performed a sensitivity analysis, restricting our sample to prevalence estimates based on the ADI-R, a well-validated clinical interview that includes rigorous assessment of hyperacusis severity. Using Bayesian meta-regression, we determined that nearly 70% of the between-study heterogeneity in ADI-R current prevalence estimates was due to the age of the study sample, with older individuals on average tending to report substantially higher rates of hyperacusis than younger individuals. Notably, the age range of the current ADI-R studies (2–14 years) did not include adults, potentially accounting for the inconclusive moderation by age seen in the full sample. Although data from adult samples were relatively scarce ($k_{(3)} = 6$), post-hoc examination of the association between current hyperacusis prevalence and age across all subjective studies revealed an inverted U-shaped relationship, with prevalence peaking in adolescence and declining in adulthood (see Supplemental Figure S3 for a graphical representation of this pattern). Additional studies are necessary to more precisely determine the prevalence of hyperacusis in older adolescents and adults with ASD, providing fundamental information about the trajectory of hyperacusis symptoms over the lifespan.

Strengths of the current study include the large and diverse body of literature collected for analysis via extensive search procedures, sample demographics similar to the broader ASD population (cf. Maenner et al., 2020), the use of statistically robust Bayesian hierarchical models, the simultaneous assessment of current and lifetime prevalence, and the inclusion of a sensitivity analysis to eliminate heterogeneity caused by measurement instrument. However, our study is not without limitations. Much like the prevalence study of Lai et al. (2019), we chose to only extract prevalence estimates from individuals with ASD, preventing us from calculating odds ratios comparing the prevalence of hyperacusis in ASD to that in the general population. The estimates included in our study were also extremely heterogeneous ($I^2 > 0.90$). Although we used meta-regression to test a number of potential moderators of prevalence across studies and found age to be a meaningful predictor of hyperacusis prevalence, much between-study heterogeneity was nevertheless left unexplained. Thus, a number of still unexplained variables, such as the particular inclusion/exclusion criteria and study questions of each sample, may exert substantial influence over reported prevalence estimates. We were also unable to rule out a number of putative moderators of hyperacusis prevalence estimates, including sex ratio, proportion of the sample with intellectual disability, HDI, and study quality (for which findings were inconclusive). Future research on this topic should therefore attempt to further define specific sociodemographic and methodological factors that affect estimates of hyperacusis prevalence in the ASD population.

One major contributor to the heterogeneity between studies was the criteria used to assess hyperacusis. As no clinical consensus has been reached regarding the optimal diagnostic procedure for hyperacusis in ASD, we chose to include studies in which authors made the determination of hyperacusis status based on their own criteria, as well as studies that employed face-valid single item measures of decreased sound tolerance. Notably, many items used to assess hyperacusis (such as the item *Does your child ever seem oversensitive to noise? (e.g., plugging ears)* on the M-CHAT) were worded to assess sound intolerance broadly, possibly conflating the presence of hyperacusis with phonophobia, misophonia, or other constructs such as inability to focus in the presence of background noise. However, this is not the case for the ADI-R, which includes a great deal of additional questioning and interviewer guidance to specifically differentiate hyperacusis from related sound tolerance conditions. As prevalence estimates did not meaningfully change when restricting our analyses to only those studies assessing hyperacusis using the ADI-R, we are relatively confident that the prevalence figures reported are not substantially inflated by the inclusion of additional non-hyperacusis sound tolerance complaints.

While broad inclusion criteria allowed us to gather a large and diverse sample of studies, it also resulted in high between-study heterogeneity, limiting our conclusions about the prevalence of hyperacusis that can be expected in later studies. Future research should attempt to define a diagnostic “gold-standard” for hyperacusis (e.g., based on loudness discomfort levels, as suggested by Aazh & Moore, 2017), which can then be used to provide more reliable and valid prevalence estimates of this condition in ASD and other populations of interest. An additional limitation of our study was the inability to determine the severity of hyperacusis symptoms. A sizable portion of individuals with ASD and hyperacusis may have relatively mild symptoms (Bennett et al., 2019; Law et al., 2016), and thus the number of individuals with hyperacusis severe enough to warrant clinical intervention remains unclear. Lastly, although we examined the association between age and hyperacusis prevalence, all studies included in our meta-analysis were cross-sectional, limiting the conclusions we are able to draw about the developmental trajectories of hyperacusis symptoms. Longitudinal evaluations of hyperacusis severity in ASD are thus needed to further understand these trajectories and identify the factors that best predict the remission of hyperacusis symptoms with age.

In conclusion, our meta-analysis suggests that hyperacusis is extremely common in individuals with ASD, with prevalence rates greater than those of most major mental disorders that commonly co-occur with autism (Lai et al., 2019). The prevalence of hyperacusis was similar across a range of questionnaire and interview measures, and despite a large amount of heterogeneity, prevalence estimates were relatively unaffected by study-level factors including sex ratio and cognitive ability. Based on ADI-R data, the prevalence of hyperacusis appears to increase with age until adolescence, but preliminary evidence from older samples suggests that hyperacusis symptoms may remit in adulthood for some individuals. Additional research in older individuals on the spectrum is necessary for a high level of confidence in this finding. By demonstrating the frequency of comorbid hyperacusis in ASD, our study highlights the need for health care providers to assess, monitor, and accommodate decreased sound tolerance in individuals with ASD across the lifespan. Furthermore, as there is currently insufficient evidence to recommend any behavioral or pharmacological treatment for hyperacusis in ASD (Fung et al., 2012; Sandbank et al., 2020; Schoen et al., 2019; Sinha et al., 2006; Weitlauf et al., 2017), additional research is desperately needed to develop and evaluate clinical interventions targeting this common symptom (Fackrell et al., 2019).

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

This work was supported by National Institute of General Medical Sciences grant T32-GM007347 (ZJW) and the Nancy Lurie-Marks Family Foundation (ZJW, TW). The authors would like to thank the research teams who conducted all primary studies summarized herein, as well as all of the individuals with autism and their families who have contributed to this body of research.

Conflicts of Interest and Sources of Funding

This work was supported by National Institute of General Medical Sciences grant T32-GM007347 (ZJW) and the Nancy Lurie-Marks Family Foundation (ZJW, TW). ZJW serves on the Family Advisory Committee of the Autism Speaks Autism Treatment Network site at Vanderbilt University and the autistic researcher review board of the Autism Intervention Research Network on Physical Health (AIR-P). He has also received consulting fees from Roche. The remaining authors have nothing to declare. No funding agency or source of support had a role in the study design, data collection, analysis, or interpretation, decision to publish, or preparation of this manuscript.

References

Aazh H, & Moore BCJ (2017). Factors related to uncomfortable loudness levels for patients seen in a tinnitus and hyperacusis clinic. International Journal of Audiology, 56(10), 793–800. 10.1080/14992027.2017.1335888 [PubMed: 28622055]

Albores-Gallo L, Roldán-Ceballos O, Villarreal-Valdes G, Betanzos-Cruz BX, Santos-Sánchez C, Martínez-Jaime MM, Lemus-Espinosa I, & Hilton CL (2012). M-CHAT Mexican version validity and reliability and some cultural considerations. ISRN Neurology, 2012, 1–7. 10.5402/2012/408694

Allison C, Baron-Cohen S, Wheelwright S, Charman T, Richler J, Pasco G, & Brayne C (2008). The Q-CHAT (Quantitative CHecklist for Autism in Toddlers): A normally distributed quantitative measure of autistic traits at 18–24 months of age: Preliminary report. Journal of Autism and Developmental Disorders, 38(8), 1414–1425. 10.1007/s10803-007-0509-7 [PubMed: 18240013]

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (DSM-5®) (5th ed.). American Psychiatric Association Publishing.

Amir I, Lamerton D, & Montague M-L (2018). Hyperacusis in children: The Edinburgh experience. International Journal of Pediatric Otorhinolaryngology, 112, 39–44. 10.1016/j.ijporl.2018.06.015 [PubMed: 30055737]

Assi H, Moore RD, Ellemberg D, & Hébert S (2018). Sensitivity to sounds in sport-related concussed athletes: A new clinical presentation of hyperacusis. Scientific Reports, 8(1), 9921. 10.1038/s41598-018-28312-1 [PubMed: 29967340]

Azouz HG, Khalil M, Abd El Ghani HM, & Hamed HM (2014). Somatosensory evoked potentials in children with autism. Alexandria Journal of Medicine, 50(2), 99–105. 10.1016/j.ajme.2013.07.002

Azouz HG, Kozou H, Khalil M, Abdou RM, & Sakr M (2014). The correlation between central auditory processing in autistic children and their language processing abilities. International Journal of Pediatric Otorhinolaryngology, 78(12), 2297–2300. 10.1016/j.ijporl.2014.10.039 [PubMed: 25468467]

Baguley DM, Bartnik G, Kleinjung T, Savastano M, & Hough EA (2013). Troublesome tinnitus in childhood and adolescence: Data from expert centres. International Journal of Pediatric Otorhinolaryngology, 77(2), 248 – 251. 10.1016/j.ijporl.2012.11.009 [PubMed: 23245492]

Baguley DM, & Hoare DJ (2018). Hyperacusis: Major research questions. HNO, 66(5), 358–363. 10.1007/s00106-017-0464-3 [PubMed: 29392341]

Bennett A, Marvin AR, Coury D, Malow BA, & Lipkin PH (2019, May). Sensitivity to sounds and touch and sleep concerns in children with ASD. International Society for Autism Research Annual Meeting. International Society for Autism Research Annual Meeting, Montreal, QC. https://insar.confex.com/insar/2019/webprogram/Paper30361.html

Ben-Sasson A, Gal E, Fluss R, Katz-Zetler N, & Cermak SA (2019). Update of a meta-analysis of sensory symptoms in ASD: A new decade of research. Journal of Autism and Developmental Disorders, 49(12), 4974–4996. 10.1007/s10803-019-04180-0 [PubMed: 31501953]

Ben-Sasson A, Hen L, Fluss R, Cermak SA, Engel-Yeger B, & Gal E (2009). A meta-analysis of sensory modulation symptoms in individuals with autism spectrum disorders. Journal of Autism and Developmental Disorders, 39(1), 1–11. 10.1007/s10803-008-0593-3 [PubMed: 18512135]

Bhatara A, Quintin E-M, Fombonne E, & Levitin DJ (2013). Early sensitivity to sound and musical preferences and enjoyment in adolescents with autism spectrum disorders. Psychomusicology: Music, Mind, and Brain, 23(2), 100–108. 10.1037/a0033754

Billstedt E, Gillberg IC, & Gillberg C (2007). Autism in adults: Symptom patterns and early childhood predictors. Use of the DISCO in a community sample followed from childhood. Journal of Child Psychology and Psychiatry, 48(11), 1102–1110. 10.1111/j.1469-7610.2007.01774.x [PubMed: 17995486]

Bishop SL, Hus V, Duncan A, Huerta M, Gotham K, Pickles A, Kreiger A, Buja A, Lund S, & Lord C (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. Journal of Autism and Developmental Disorders, 43(6), 1287–1297. 10.1007/s10803-012-1671-0 [PubMed: 23065116]

Bishop SL, Richler J, & Lord C (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. Child Neuropsychology, 12(4–5), 247–267. 10.1080/09297040600630288 [PubMed: 16911971]

Borenstein M, Hedges LV, Higgins JPT, & Rothstein HR (2009). Introduction to meta-analysis. Wiley.

Bürkner P-C (2017). brms: An R package for Bayesian multilevel models using Stan. Journal of Statistical Software, 80(1). 10.18637/jss.v080.i01

Bürkner P-C (2018). Advanced Bayesian multilevel modeling with the R package brms. The R Journal, 10(1), 395–411.

Canal-Bedia R, García-Primo P, Martín-Cilleros MV, Santos-Borbujo J, Guisuraga-Fernández Z, Herráez-García L, del Mar Herráez-García M, Boada-Muñoz L, Fuentes-Biggi J, & Posada-de la Paz M (2011). Modified Checklist for Autism in Toddlers: Cross-cultural adaptation and validation in Spain. Journal of Autism and Developmental Disorders, 41(10), 1342–1351. 10.1007/s10803-010-1163-z [PubMed: 21161677]

Carrington SJ, Barrett SL, Sivagamasundari U, Fretwell C, Noens I, Maljaars J, & Leekam SR (2019). Describing the profile of diagnostic features in autistic adults using an abbreviated version of the Diagnostic Interview for Social and Communication Disorders (DISCO-Abbreviated). Journal of Autism and Developmental Disorders. 10.1007/s10803-019-04214-7

Carrington SJ, Kent RG, Maljaars J, Le Couteur A, Gould J, Wing L, Noens I, Van Berckelaer-Onnes I, & Leekam SR (2014). DSM-5 autism spectrum disorder: In search of essential behaviours for diagnosis. Research in Autism Spectrum Disorders, 8(6), 701–715. 10.1016/j.rasd.2014.03.017

Cederroth CR, Lugo A, Edvall NK, Lazar A, Lopez-Escamez J-A, Bulla J, Uhlen I, Hoare DJ, Baguley DM, Canlon B, & Gallus S (2020). Association between hyperacusis and tinnitus. Journal of Clinical Medicine, 9(8), 2412. 10.3390/jcm9082412

Cervantes PE, Matson JL, & Peters WJ (2017). An abbreviated scoring algorithm for the Baby and Infant Screen for Children with Autism Traits. Developmental Neurorehabilitation, 20(5), 287–293. 10.1080/17518423.2016.1211189 [PubMed: 27715370]

Chaste P, Klei L, Sanders SJ, Hus V, Murtha MT, Lowe JK, Willsey AJ, Moreno-De-Luca D, Yu TW, Fombonne E, Geschwind D, Grice DE, Ledbetter DH, Mane SM, Martin DM, Morrow EM, Walsh CA, Sutcliffe JS, Lese Martin C, … Devlin B (2015). A genome-wide association study of autism using the Simons Simplex Collection: Does reducing phenotypic heterogeneity in autism increase genetic homogeneity? Biological Psychiatry, 77(9), 775–784. 10.1016/j.biopsych.2014.09.017 [PubMed: 25534755]

Cheung MWL (2014). Modeling dependent effect sizes with three-level meta-analyses: A structural equation modeling approach. Psychological Methods, 19(2), 211–229. 10.1037/a0032968 [PubMed: 23834422]

Çilingirtürk AM, & Koçak H (2018). Human Development Index (HDI) rank-order variability. Social Indicators Research, 137(2), 481–504. 10.1007/s11205-017-1605-5

Cohen J (1960). A coefficient of agreement for nominal scales. Educational and Psychological Measurement, 20(1), 37–46. 10.1177/001316446002000104

Dai YG, Porto KS, Skapek M, Barton ML, Dumont-Mathieu T, Fein DA, & Robins DL (2020). Comparison of the Modified Checklist for Autism in Toddlers, Revised with Follow-Up (M-CHAT-R/F) positive predictive value by race. Journal of Autism and Developmental Disorders. 10.1007/s10803-020-04428-0

Danesh AA, Lang D, Kaf W, Andreassen WD, Scott J, & Eshraghi AA (2015). Tinnitus and hyperacusis in autism spectrum disorders with emphasis on high functioning individuals diagnosed with Asperger's syndrome. International Journal of Pediatric Otorhinolaryngology, 79(10), 1683–1688. 10.1016/j.ijporl.2015.07.024 [PubMed: 26243502]

de Giambattista C, Ventura P, Trerotoli P, Margari M, Palumbi R, & Margari L (2019). Subtyping the Autism Spectrum Disorder: Comparison of Children with High Functioning Autism and Asperger Syndrome. Journal of Autism and Developmental Disorders, 49(1), 138–150. 10.1007/s10803-018-3689-4 [PubMed: 30043350]

Demopoulos C, & Lewine JD (2016). Audiometric profiles in autism spectrum disorders: Does subclinical hearing loss impact communication? Autism Research, 9(1), 107–120. 10.1002/aur.1495 [PubMed: 25962745]

Dietz PM, Rose CE, McArthur D, & Maenner M (2020). National and State Estimates of Adults with Autism Spectrum Disorder. Journal of Autism and Developmental Disorders. 10.1007/s10803-020-04494-4

Downs D, Schmidt B, & Stephens TJ (2005). Auditory behaviors of children and adolescents with pervasive developmental disorders. Seminars in Hearing, 26(4), 226–240. 10.1055/s-2005-922445

Egelhoff K (2011). Auditory processing abilities of children diagnosed with autism spectrum disorder [PhD Thesis, The Ohio State University]. http://rave.ohiolink.edu/etdc/view?acc_num=osu1306864962

Fackrell K, & Hoare DJ (2018). Scales and questionnaires for decreased sound tolerance. In Fagelson MA & Baguley DM (Eds.), Hyperacusis and disorders of sound intolerance (pp. 43–58). Plural Publishing.

Fackrell K, Stratmann L, Kennedy V, MacDonald C, Hodgson H, Wray N, Farrell C, Meadows M, Sheldrake J, Byrom P, Baguley DM, Kentish R, Chapman S, Marriage J, Phillips J, Pollard T, Henshaw H, Gronlund TA, & Hoare DJ (2019). Identifying and prioritising unanswered research questions for people with hyperacusis: James Lind Alliance Hyperacusis Priority Setting Partnership. BMJ Open, 9(11), e032178. 10.1136/bmjopen-2019-032178

Falkmer T, Anderson K, Falkmer M, & Horlin C (2013). Diagnostic procedures in autism spectrum disorders: A systematic literature review. European Child & Adolescent Psychiatry, 22(6), 329–340. 10.1007/s00787-013-0375-0 [PubMed: 23322184]

Fung LK, Chahal L, Libove RA, Bivas R, & Hardan AY (2012). A retrospective review of the effectiveness of aripiprazole in the treatment of sensory abnormalities in autism. Journal of Child and Adolescent Psychopharmacology, 22(3), 245–248. 10.1089/cap.2010.0103 [PubMed: 22537360]

Gillum LA, Gouveia C, Dorsey ER, Pletcher M, Mathers CD, McCulloch CE, & Johnston SC (2011). NIH disease funding levels and burden of disease. PLoS ONE, 6(2), e16837. 10.1371/journal.pone.0016837 [PubMed: 21383981]

Gomes E, Pedroso FS, & Wagner MB (2008). Auditory hypersensitivity in the autistic spectrum disorder. Pró-Fono Revista de Atualização Científica, 20(4), 279–284. 10.1590/s0104-56872008000400013 [PubMed: 19142473]

Gomes E, Rotta NT, Pedroso FS, Sleifer P, & Danesi MC (2004). Auditory hypersensitivity in children and teenagers with autistic spectrum disorder. Arquivos De Neuro-Psiquiatria, 62(3B), 797–801. 10.1590/s0004-282x2004000500011 [PubMed: 15476072]

Grapel JN, Cicchetti DV, & Volkmar FR (2015). Sensory features as diagnostic criteria for autism: Sensory features in autism. The Yale Journal of Biology and Medicine, 88(1), 69–71. [PubMed: 25745375]

Green D, Chandler S, Charman T, Simonoff E, & Baird G (2016). Brief report: DSM-5 sensory behaviours in children with and without an autism spectrum disorder. Journal of Autism and Developmental Disorders, 46(11), 3597–3606. 10.1007/s10803-016-2881-7 [PubMed: 27475418]

Griffith GM, Totsika V, Nash S, & Hastings RP (2011). 'I just don't fit anywhere': Support experiences and future support needs of individuals with Asperger syndrome in middle adulthood. Autism, 16(5), 532–546. 10.1177/1362361311405223 [PubMed: 21610188]

Gronau QF, Sarafoglou A, Matzke D, Ly A, Boehm U, Marsman M, Leslie DS, Forster JJ, Wagenmakers E-J, & Steingroever H (2017). A tutorial on bridge sampling. Journal of Mathematical Psychology, 81, 80–97. 10.1016/j.jmp.2017.09.005 [PubMed: 29200501]

Gronau QF, Singmann H, & Wagenmakers E-J (2020). bridgesampling: An R package for estimating normalizing constants. Journal of Statistical Software, 92(10), 1–29. 10.18637/jss.v092.i10

Grzadzinski R, Dick C, Lord C, & Bishop S (2016). Parent-reported and clinician-observed autism spectrum disorder (ASD) symptoms in children with attention deficit/hyperactivity disorder (ADHD): Implications for practice under DSM-5. Molecular Autism, 7(1), 7. 10.1186/s13229-016-0072-1 [PubMed: 26788284]

Guo C, Luo M, Wang X, Huang S, Meng Z, Shao J, Zhang X, Shao Z, Wu J, Robins DL, & Jing J (2019). Reliability and validity of the Chinese version of Modified Checklist for Autism in Toddlers, Revised, with Follow-Up (M-CHAT-R/F). Journal of Autism and Developmental Disorders, 49(1), 185–196. 10.1007/s10803-018-3682-y [PubMed: 30047095]

Hall AJ, Humphriss R, Baguley DM, Parker M, & Steer CD (2016). Prevalence and risk factors for reduced sound tolerance (hyperacusis) in children. International Journal of Audiology, 55(3), 135–141. 10.3109/14992027.2015.1092055 [PubMed: 26642866]

Hamza TH, van Houwelingen HC, & Stijnen T (2008). The binomial distribution of meta-analysis was preferred to model within-study variability. Journal of Clinical Epidemiology, 61(1), 41–51. 10.1016/j.jclinepi.2007.03.016 [PubMed: 18083461]

Hattier MA, Matson JL, May AC, & Whiting SE (2012). Repetitive/restricted behaviours and interests in children with cerebral palsy and autism spectrum disorder. Developmental Neurorehabilitation, 15(3), 178–184. 10.3109/17518423.2012.657306 [PubMed: 22582848]

Hayward SM, McVilly KR, & Stokes MA (2019). Autism and employment: What works. Research in Autism Spectrum Disorders, 60, 48–58. 10.1016/j.rasd.2019.01.006

Hazen EP, Stornelli JL, O'Rourke JA, Koesterer K, & McDougle CJ (2014). Sensory symptoms in autism spectrum disorders. Harvard Review of Psychiatry, 22(2), 112–124. 10.1097/01.HRP.0000445143.08773.58 [PubMed: 24614766]

Hedley D, Cai R, Uljarević M, Wilmot M, Spoor JR, Richdale A, & Dissanayake C (2018). Transition to work: Perspectives from the autism spectrum. Autism, 22(5), 528–541. 10.1177/1362361316687697 [PubMed: 28387577]

Higgins JPT, & Thompson SG (2002). Quantifying heterogeneity in a meta-analysis. Statistics in Medicine, 21(11), 1539–1558. 10.1002/sim.1186 [PubMed: 12111919]

Hoijtink H, Gu X, Mulder J, & Rosseel Y (2019). Computing Bayes factors from data with missing values. Psychological Methods, 24(2), 253–268. 10.1037/met0000187 [PubMed: 29999379]

Hussein S, Bahmei B, Gustafson K, Fisher R, Iarocci G, Arzanpour S, & Birmingham E (2019, May). Family experiences of auditory hypersensitivity in ASD. International Society for Autism Research Annual Meeting, Montreal, QC. https://insar.confex.com/insar/2019/webprogram/Paper30947.html

Inada N, Koyama T, Inokuchi E, Kuroda M, & Kamio Y (2011). Reliability and validity of the Japanese version of the Modified Checklist for Autism in Toddlers (M-CHAT). Research in Autism Spectrum Disorders, 5(1), 330–336. 10.1016/j.rasd.2010.04.016

IntHout J, Ioannidis JPA, Rovers MM, & Goeman JJ (2016). Plea for routinely presenting prediction intervals in meta-analysis. BMJ Open, 6(7), e010247. 10.1136/bmjopen-2015-010247

Jastreboff PJ, & Jastreboff MM (2015). Decreased sound tolerance: Hyperacusis, misophonia, diplacousis, and polyacousis. In Aminoff MJ, Boller F, & Swaab DF (Eds.), Handbook of clinical neurology (Vol. 129, pp. 375–387). Elsevier. 10.1016/B978-0-444-62630-1.00021-4 [PubMed: 25726280]

Johnson CP, Myers SM, & Council on Children With Disabilities. (2007). Identification and evaluation of children with autism spectrum disorders. Pediatrics, 120(5), 1183–1215. 10.1542/peds.2007-2361 [PubMed: 17967920]

Jones RSP, Quigney C, & Huws JC (2003). First-hand accounts of sensory perceptual experiences in autism: A qualitative analysis. Journal of Intellectual & Developmental Disability, 28(2), 112–121. 10.1080/1366825031000147058

Jussila K, Junttila M, Kielinen M, Ebeling H, Joskitt L, Moilanen I, & Mattila M-L (2020). Sensory abnormality and quantitative autism traits in children with and without autism spectrum disorder in an epidemiological population. Journal of Autism and Developmental Disorders, 50(1), 180–188. 10.1007/s10803-019-04237-0 [PubMed: 31583623]

Kamio Y, Haraguchi H, Stickley A, Ogino K, Ishitobi M, & Takahashi H (2015). Brief report: Best discriminators for identifying children with autism spectrum disorder at an 18-month health check-up in Japan. Journal of Autism and Developmental Disorders, 45(12), 4147–4153. 10.1007/s10803-015-2527-1 [PubMed: 26189180]

Kanner L (1943). Autistic disturbances of affective contact. Nervous Child, 2, 217–250.

Kara B, Mukaddes NM, Altınkaya I, Güntepe D, Gökçay G, & Özmen M (2014). Using the Modified Checklist for Autism in Toddlers in a well-child clinic in Turkey: Adapting the screening method based on culture and setting. Autism, 18(3), 331–338. 10.1177/1362361312467864 [PubMed: 23175752]

Khalfa S, Dubal S, Veuillet E, Perez-Diaz F, Jouvent R, & Collet L (2002). Psychometric normalization of a hyperacusis questionnaire. ORL, 64(6), 436–442. 10.1159/000067570 [PubMed: 12499770]

Kientz MA, & Dunn W (1997). A comparison of the performance of children with and without autism on the Sensory Profile. American Journal of Occupational Therapy, 51(7), 530–537. 10.5014/ajot.51.7.530

Kim SH, Joseph RM, Frazier JA, O'Shea TM, Chawarska K, Allred EN, Leviton A, & Kuban KK (2016). Predictive validity of the Modified Checklist for Autism in Toddlers (M-CHAT) born very preterm. The Journal of Pediatrics, 178, 101–107.e2. 10.1016/j.jpeds.2016.07.052 [PubMed: 27592094]

Klintwall L, Holm A, Eriksson M, Carlsson LH, Olsson MB, Hedvall Å, Gillberg C, & Fernell E (2011). Sensory abnormalities in autism: A brief report. Research in Developmental Disabilities, 32(2), 795–800. 10.1016/j.ridd.2010.10.021 [PubMed: 21111574]

Kopecky K, Broder-Fingert S, Iannuzzi D, & Connors S (2013). The needs of hospitalized patients with autism spectrum disorders: A parent survey. Clinical Pediatrics, 52(7), 652–660. 10.1177/0009922813485974 [PubMed: 23624619]

Kozlowski AM, Matson JL, Worley JA, Sipes M, & Horovitz M (2012). Defining characteristics for young children meeting cutoff on the Modified Checklist for Autism in Toddlers. Research in Autism Spectrum Disorders, 6(1), 472–479. 10.1016/j.rasd.2011.07.007

Kuiper MWM, Verhoeven EWM, & Geurts HM (2019). Stop making noise! Auditory sensitivity in adults with an autism spectrum disorder diagnosis: Physiological habituation and subjective detection thresholds. Journal of Autism and Developmental Disorders, 49(5), 2116–2128. 10.1007/s10803-019-03890-9 [PubMed: 30680585]

Lai M-C, Kassee C, Besney R, Bonato S, Hull L, Mandy W, Szatmari P, & Ameis SH (2019). Prevalence of co-occurring mental health diagnoses in the autism population: A systematic review and meta-analysis. The Lancet Psychiatry, 6(10), 819–829. 10.1016/s2215-0366(19)30289-5 [PubMed: 31447415]

Landon J, Shepherd D, & Lodhia V (2016). A qualitative study of noise sensitivity in adults with autism spectrum disorder. Research in Autism Spectrum Disorders, 32, 43–52. 10.1016/j.rasd.2016.08.005

Law JK, Rubenstein E, Marvin AR, Toroney J, & Lipkin PH (2016, May). Auditory sensitivity issues in children with autism spectrum disorders: Characteristics and burden. Pediatric Academic Societies Meeting, Baltimore, MD. https://iancommunity.org/sites/default/files/galleries/conference-presentations/Law_PAS_2016.pdf

Levitin DJ, Cole K, Lincoln A, & Bellugi U (2005). Aversion, awareness, and attraction: Investigating claims of hyperacusis in the Williams syndrome phenotype. Journal of Child Psychology and Psychiatry, 46(5), 514–523. 10.1111/j.1469-7610.2004.00376.x [PubMed: 15845131]

Lord C, Pickles A, McLennan J, Rutter M, Bregman J, Folstein S, Fombonne E, Leboyer M, & Minshew N (1997). Diagnosing autism: Analyses of data from the Autism Diagnostic Interview. Journal of Autism and Developmental Disorders, 27(5), 501–517. 10.1023/A:1025873925661 [PubMed: 9403369]

Lord C, Rutter M, & Le Couteur A (1994). Autism Diagnostic Interview-Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. Journal of Autism and Developmental Disorders, 24(5), 659–685. 10.1007/BF02172145 [PubMed: 7814313]

Lorenz T, Frischling C, Cuadros R, & Heinitz K (2016). Autism and overcoming job barriers: Comparing job-related barriers and possible solutions in and outside of autism-specific employment. PLoS One, 11(1), e0147040. 10.1371/journal.pone.0147040 [PubMed: 26766183]

Maenner MJ, Shaw KA, Baio J, Washington A, Vehorn A, DiRienzo M, Christensen DL, Wiggins LD, Pettygrove S, Andrews JG, Lopez M, Hudson A, Baroud T, Schwenk Y, White T, Rosenberg CR, Lee L-C, Harrington RA, Huston M, … Dietz PM (2020). Prevalence of autism spectrum disorder among children aged 8 Years—Autism and developmental disabilities monitoring network, 11 Sites, United States, 2016. MMWR Surveillance Summaries, 69(4), 1–12. 10.15585/mmwr.ss6904a1

Matheis M, Matson JL, Hong E, & Cervantes PE (2019). Gender differences and similarities: Autism symptomatology and developmental functioning in young children. Journal of Autism and Developmental Disorders, 49(3), 1219–1231. 10.1007/s10803-018-3819-z [PubMed: 30443700]

Matson JL, Kozlowski AM, Fitzgerald ME, & Sipes M (2013). True versus false positives and negatives on the Modified Checklist for Autism in Toddlers. Research in Autism Spectrum Disorders, 7(1), 17–22. 10.1016/j.rasd.2012.02.011

Matson JL, Matheis M, Burns CO, Esposito G, Venuti P, Pisula E, Misiak A, Kalyva E, Tsakiris V, Kamio Y, Ishitobi M, & Goldin RL (2017). Examining cross-cultural differences in autism spectrum disorder: A multinational comparison from Greece, Italy, Japan, Poland, and the United States. European Psychiatry, 42, 70–76. 10.1016/j.eurpsy.2016.10.007 [PubMed: 28212508]

McIntosh DN, Miller LJ, & Shyu V (1999). Development and validation of the Short Sensory Profile. In Dunn W (Ed.), Sensory Profile manual (pp. 59–73). Psychological Corporation.

Mercati O, Huguet G, Danckaert A, André-Leroux G, Maruani A, Bellinzoni M, Rolland T, Gouder L, Mathieu A, Buratti J, Amsellem F, Benabou M, Van-Gils J, Beggiato A, Konyukh M, Bourgeois J-P, Gazzellone MJ, Yuen RKC, Walker S, … Bourgeron T (2017). CNTN6 mutations are risk factors for abnormal auditory sensory perception in autism spectrum disorders. Molecular Psychiatry, 22(4), 625–633. 10.1038/mp.2016.61 [PubMed: 27166760]

Moher D, Liberati A, Tetzlaff J, Altman DG, & The PRISMA Group. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. PLoS Medicine, 6(7), e1000097. 10.1371/journal.pmed.1000097 [PubMed: 19621072]

Myne S, & Kennedy V (2018). Hyperacusis in children: A clinical profile. International Journal of Pediatric Otorhinolaryngology, 107, 80–85. 10.1016/j.ijporl.2018.01.004 [PubMed: 29501317]

Nemholt SS, Schmidt JH, Wedderkopp N, & Baguley DM (2015). Prevalence of tinnitus and/or hyperacusis in children and adolescents: Study protocol for a systematic review. BMJ Open, 5(1), e006649. 10.1136/bmjopen-2014-006649

Nupur AD, Miah MS, & Moniruzzaman S (2016). Status of sensory profile score among children with autism in selected centers of Dhaka city. Bangladesh Journal of Occupational Therapy and Rehabilitation, 1(1), 20–27.

O'Connor K (2012). Auditory processing in autism spectrum disorder: A review. Neuroscience and Biobehavioral Reviews, 36(2), 836–854. 10.1016/j.neubiorev.2011.11.008 [PubMed: 22155284]

Pahan S (2003). Symptom profile in children with pervasive development disorder [Master's Thesis, Central Institute of Psychiatry]. ProQuest Dissertations and Theses. https://search.proquest.com/docview/1825999164/16D5F92A2E924C2DPQ/1

Pastor DA, & Lazowski RA (2017). On the multilevel nature of meta-analysis: A tutorial, comparison of software programs, and discussion of analytic choices. Multivariate Behavioral Research, 53(1), 74–89. 10.1080/00273171.2017.1365684 [PubMed: 28952787]

Paulin J, Andersson L, & Nordin S (2016). Characteristics of hyperacusis in the general population. Noise & Health, 18(83), 178–184. 10.4103/1463-1741.189244 [PubMed: 27569405]

Pienkowski M, Tyler RS, Roncancio ER, Jun HJ, Brozoski T, Dauman N, Coelho CB, Andersson G, Keiner AJ, Cacace AT, Martin N, & Moore BCJ (2014). A review of hyperacusis and future directions: Part II. Measurement, mechanisms, and treatment. American Journal of Audiology, 23(4), 420–436. 10.1044/2014_AJA-13-0037 [PubMed: 25478787]

Porges SW, Bazhenova OV, Bal E, Carlson N, Sorokin Y, Heilman KJ, Cook EH, & Lewis GF (2014). Reducing auditory hypersensitivities in autistic spectrum disorder: Preliminary findings evaluating the listening project protocol. Frontiers in Pediatrics, 2, 80. 10.3389/fped.2014.00080 [PubMed: 25136545]

Potgieter I, Fackrell K, Kennedy V, Crunkhorn R, & Hoare DJ (2020). Hyperacusis in children: A scoping review. BMC Pediatrics, 20(1), 319. 10.1186/s12887-020-02223-5 [PubMed: 32600446]

R Core Team. (2020). R: A language and environment for statistical computing (4.0.2) [Computer software]. R Foundation for Statistical Computing. https://www.R-project.org/

Radziwon KE, Manohar S, Auerbach B, Liu X, Chen G-D, & Salvi R (2020). Preclinical animal behavioral models of hyperacusis and loudness recruitment. In Pucheu S, Radziwon KE, & Salvi R (Eds.), New therapies to prevent or cure auditory disorders (pp. 135–157). Springer International Publishing. 10.1007/978-3-030-40413-0_6

Richler J, Bishop SL, Kleinke JR, & Lord C (2007). Restricted and repetitive behaviors in young children with autism spectrum disorders. Journal of Autism and Developmental Disorders, 37(1), 73–85. 10.1007/s10803-006-0332-6 [PubMed: 17195920]

Rimland B, & Edelson SM (1995). Brief report: A pilot study of auditory integration training in autism. Journal of Autism and Developmental Disorders, 25(1), 61–70. 10.1007/BF02178168 [PubMed: 7608035]

Roberts LE, & Salvi R (2019). Overview: Hearing loss, tinnitus, hyperacusis, and the role of central gain. Neuroscience, 407, 1–7. 10.1016/j.neuroscience.2019.03.021 [PubMed: 30885639]

Robertson AE, & Simmons DR (2015). The sensory experiences of adults with autism spectrum disorder: A qualitative analysis. Perception, 44(5), 569–586. 10.1068/p7833 [PubMed: 26422904]

Robins DL, Casagrande K, Barton M, Chen C-MA, Dumont-Mathieu T, & Fein D (2014). Validation of the Modified Checklist for Autism in Toddlers, Revised with Follow-up (M-CHAT-R/F). Pediatrics, 133(1), 37–45. 10.1542/peds.2013-1813 [PubMed: 24366990]

Robins DL, Fein D, Barton ML, & Green JA (2001). The Modified Checklist for Autism in Toddlers: An initial study investigating the early detection of autism and pervasive developmental disorders. Journal of Autism and Developmental Disorders, 31(2), 131–144. 10.1023/A:1010738829569 [PubMed: 11450812]

Rødgaard E-M, Jensen K, Vergnes J-N, Soulières I, & Mottron L (2019). Temporal changes in effect sizes of studies comparing individuals with and without autism: A meta-analysis. JAMA Psychiatry, 76(11), 1124. 10.1001/jamapsychiatry.2019.1956 [PubMed: 31433441]

Rosenhall U, Nordin V, Sandström M, Ahlsén G, & Gillberg C (1999). Autism and hearing loss. Journal of Autism and Developmental Disorders, 29(5), 349–357. 10.1023/A:1023022709710

Rosing SN, Kapandais A, Schmidt JH, & Baguley DM (2016). Demographic data, referral patterns and interventions used for children and adolescents with tinnitus and hyperacusis in Denmark. International Journal of Pediatric Otorhinolaryngology, 89, 112–120. 10.1016/j.ijporl.2016.07.036 [PubMed: 27619040]

Rosing SN, Schmidt JH, Wedderkopp N, & Baguley DM (2016). Prevalence of tinnitus and hyperacusis in children and adolescents: A systematic review. BMJ Open, 6(6), e010596. 10.1136/bmjopen-2015-010596

Rutter M, Le Couteur A, & Lord C (2013). ADI-R: Autism Diagnostic Interview–Revised: Manual. Western Psychological Services.

Sandbank M, Bottema-Beutel K, Crowley S, Cassidy M, Dunham K, Feldman JI, Crank J, Albarran SA, Raj S, Mahbub P, & Woynaroski TG (2020). Project AIM: Autism intervention meta-analysis for studies of young children. Psychological Bulletin, 146(1), 1–29. 10.1037/bul0000215 [PubMed: 31763860]

Schoen SA, Lane SJ, Mailloux Z, May-Benson T, Parham LD, Roley SS, & Schaaf RC (2019). A systematic review of Ayres Sensory Integration intervention for children with autism. Autism Research, 12(1), 6–19. 10.1002/aur.2046 [PubMed: 30548827]

Schwarzer G, Chemaitelly H, Raddad LJA, & Rücker G (2019). Seriously misleading results using inverse of Freeman-Tukey double arcsine transformation in meta-analysis of single proportions. Research Synthesis Methods, 67(11), 974. 10.1002/jrsm.1348

Shah AD, Bartlett JW, Carpenter J, Nicholas O, & Hemingway H (2014). Comparison of random forest and parametric imputation models for imputing missing data using MICE: A CALIBER study. American Journal of Epidemiology, 179(6), 764–774. 10.1093/aje/kwt312 [PubMed: 24589914]

Shardell AR (2013). Parent experiences with early symptoms and diagnosis of children with mild autism and Asperger's syndrome [PhD Thesis, Capella University]. https://search.proquest.com/docview/1491162681/24492D9E74F43D8PQ/1

Sheldrake J, Diehl PU, & Schaette R (2015). Audiometric characteristics of hyperacusis patients. Frontiers in Neurology, 6(1), 105. 10.3389/fneur.2015.00105 [PubMed: 26029161]

Shrout PE, & Fleiss JL (1979). Intraclass correlations: Uses in assessing rater reliability. Psychological Bulletin, 86(2), 420–428. 10.1037/0033-2909.86.2.420 [PubMed: 18839484]

Silva LMT, & Schalock M (2012). Sense and Self-Regulation Checklist, a measure of comorbid autism symptoms: Initial psychometric evidence. American Journal of Occupational Therapy, 66(2), 177–186. 10.5014/ajot.2012.001578

Simonoff E (2020, June 3). Mental health in autistic people: Setting a research agenda for the coming decade [INSAR 2020 Virtual - Keynote Webinar]. International Society for Autism Research Annual Meeting. https://insar.confex.com/insar/2020/meetingapp.cgi/Paper/36311

Sinha Y, Silove N, Wheeler D, & Williams K (2006). Auditory integration training and other sound therapies for autism spectrum disorders: A systematic review. Archives of Disease in Childhood, 91(12), 1018–1022. 10.1136/adc.2006.094649 [PubMed: 16887860]

Sipes M, Matson JL, Worley JA, & Kozlowski AM (2011). Gender differences in symptoms of autism spectrum disorders in toddlers. Research in Autism Spectrum Disorders, 5(4), 1465–1470. 10.1016/j.rasd.2011.02.007

Smith RS, & Sharp J (2013). Fascination and isolation: A grounded theory exploration of unusual sensory experiences in adults with Asperger syndrome. Journal of Autism and Developmental Disorders, 43(4), 891–910. 10.1007/s10803-012-1633-6 [PubMed: 22923038]

Srisinghasongkram P, Pruksananonda C, & Chonchaiya W (2016). Two-step screening of the Modified Checklist for Autism in Toddlers in Thai children with language delay and typically developing children. Journal of Autism and Developmental Disorders, 46(10), 3317–3329. 10.1007/s10803-016-2876-4 [PubMed: 27460003]

Stein LI, Polido JC, & Cermak SA (2013). Oral care and sensory over-responsivity in children with autism spectrum disorders. Pediatric Dentistry, 35(3), 230–235. [PubMed: 23756306]

Stekhoven DJ (2013). missForest: Nonparametric Missing Value Imputation using Random Forest.

Stekhoven DJ, & Bühlmann P (2012). MissForest—Non-parametric missing value imputation for mixed-type data. Bioinformatics, 28(1), 112–118. 10.1093/bioinformatics/btr597 [PubMed: 22039212]

Stenberg N, Bresnahan M, Gunnes N, Hirtz D, Hornig M, Lie KK, Lipkin WI, Lord C, Magnus P, Reichborn-Kjennerud T, Schjølberg S, Surén P, Susser E, Svendsen BK, von Tetzchner S, Øyen A-S, & Stoltenberg C (2014). Identifying children with autism spectrum disorder at 18 months in a general population sample. Paediatric and Perinatal Epidemiology, 28(3), 255–262. 10.1111/ppe.12114 [PubMed: 24547686]

Sterne JAC, Gavaghan D, & Egger M (2000). Publication and related bias in meta-analysis: Power of statistical tests and prevalence in the literature. Journal of Clinical Epidemiology, 53(11), 1119–1129. 10.1016/S0895-4356(00)00242-0 [PubMed: 11106885]

Stiegler LN, & Davis R (2010). Understanding sound sensitivity in individuals with autism spectrum disorders. Focus on Autism and Other Developmental Disabilities, 25(2), 67–75. 10.1177/1088357610364530

Stijnen T, Hamza TH, & Özdemir P (2010). Random effects meta-analysis of event outcome in the framework of the generalized linear mixed model with applications in sparse data. Statistics in Medicine, 29(29), 3046–3067. 10.1002/sim.4040 [PubMed: 20827667]

Stroup DF, Berlin JA, Morton SC, Olkin I, Williamson GD, Rennie D, Moher D, Becker BJ, Sipe TA, Thacker SB, & Group, for the M. O. O. S. in E. M. (2000). Meta-analysis of observational studies in epidemiology: A proposal for reporting. JAMA, 283(15), 2008–2012. 10.1001/jama.283.15.2008 [PubMed: 10789670]

Tan Y-H, Xi C-Y, Jiang S-P, Shi B-X, Wang L-B, & Wang L (2012). Auditory abnormalities in children with autism. Open Journal of Psychiatry, 2(1), 33–37. 10.4236/ojpsych.2012.21005

Tavassoli T, Bellesheim K, Siper PM, Wang AT, Halpern D, Gorenstein M, Grodberg D, Kolevzon A, & Buxbaum JD (2016). Measuring sensory reactivity in autism spectrum disorder: Application and simplification of a clinician-administered sensory observation scale. Journal of Autism and Developmental Disorders, 46(1), 287–293. 10.1007/s10803-015-2578-3 [PubMed: 26340959]

Tavassoli T, Brandes-Aitken A, Chu R, Porter L, Schoen S, Miller LJ, Gerdes MR, Owen J, Mukherjee P, & Marco EJ (2019). Sensory over-responsivity: Parent report, direct assessment measures, and neural architecture. Molecular Autism, 10(1), 4. 10.1186/s13229-019-0255-7 [PubMed: 30740199]

Tavassoli T, Miller LJ, Schoen SA, Nielsen DM, & Baron-Cohen S (2014). Sensory over-responsivity in adults with autism spectrum conditions. Autism, 18(4), 428–432. 10.1177/1362361313477246 [PubMed: 24085741]

Tharpe AM, Bess FH, Sladen DP, Schissel H, Couch S, & Schery T (2006). Auditory characteristics of children with autism. Ear and Hearing, 27(4), 430–441. 10.1097/01.aud.0000224981.60575.d8 [PubMed: 16825892]

Tomchek SD (2005). Characterizing sensory processing in autism spectrum disorders [PhD Thesis, University of Kentucky]. https://uknowledge.uky.edu/gradschool_diss/455/

Tomchek SD, & Dunn W (2007). Sensory processing in children with and without autism: A comparative study using the Short Sensory Profile. American Journal of Occupational Therapy, 61(2), 190–200. 10.5014/ajot.61.2.190

Troyb E, Orinstein A, Tyson K, Eigsti I-M, Naigles L, & Fein D (2014). Restricted and repetitive behaviors in individuals with a history of ASDs who have achieved optimal outcomes. Journal of Autism and Developmental Disorders, 44(12), 3168–3184. 10.1007/s10803-014-2182-y [PubMed: 25030967]

Tyler RS, Pienkowski M, Roncancio ER, Jun HJ, Brozoski T, Dauman N, Coelho CB, Andersson G, Keiner AJ, Cacace AT, Martin N, & Moore BCJ (2014). A review of hyperacusis and future directions: Part I. Definitions and manifestations. American Journal of Audiology, 23(4), 402–419. 10.1044/2014_AJA-14-0010 [PubMed: 25104073]

Van den Noortgate W, López-López JA, Marín-Martínez F, & Sánchez-Meca J (2012). Three-level meta-analysis of dependent effect sizes. Behavior Research Methods, 45(2), 576–594. 10.3758/s13428-012-0261-6

Ventola P, Kleinman J, Pandey J, Wilson L, Esser E, Boorstein H, Dumont-Mathieu T, Marshia G, Barton M, Hodgson S, Green J, Volkmar F, Chawarska K, Babitz T, Robins D, & Fein D (2007). Differentiating between autism spectrum disorders and other developmental disabilities in children who failed a screening instrument for ASD. Journal of Autism and Developmental Disorders, 37(3), 425–436. 10.1007/s10803-006-0177-z [PubMed: 16897377]

VerMaas-Lee JR (1999). Parent ratings of children with autism on the Evaluation of Sensory Processing (ESP). [Master's Thesis, University of Southern California]. ProQuest Dissertations and Theses. http://digitallibrary.usc.edu/cdm/ref/collection/p15799coll16/id/33167

Volkmar FR, & McPartland JC (2014). From Kanner to DSM-5: Autism as an evolving diagnostic concept. Annual Review of Clinical Psychology, 10(1), 193–212. 10.1146/annurev-clinpsy-032813-153710

Wagenmakers E-J, Wetzels R, Borsboom D, & van der Maas HLJ (2011). Why psychologists must change the way they analyze their data: The case of psi: Comment on Bem (2011). Journal of Personality and Social Psychology, 100(3), 426–432. 10.1037/a0022790 [PubMed: 21280965]

Ware JJ, & Munafò MR (2015). Significance chasing in research practice: Causes, consequences and possible solutions. Addiction, 110(1), 4–8. 10.1111/add.12673

Weitlauf AS, Sathe N, McPheeters ML, & Warren ZE (2017). Interventions targeting sensory challenges in autism spectrum disorder: A systematic review. Pediatrics, 139(6), e20170347. 10.1542/peds.2017-0347 [PubMed: 28562287]

Williams DR, Rast P, & Bürkner PC (2018). Bayesian meta-analysis with weakly informative prior distributions. PsyArXiv. 10.31234/osf.io/7tbrm

Williams ZJ (2020). Short Sensory Profile in autism. In Volkmar FR (Ed.), Encyclopedia of Autism Spectrum Disorders (pp. 1–7). Springer. 10.1007/978-1-4614-6435-8_102311-1

Williams ZJ, Abdelmessih PG, Key AP, & Woynaroski TG (2020). Cortical auditory processing of simple stimuli is altered in autism: A meta-analysis of auditory evoked responses. Biological Psychiatry: Cognitive Neuroscience and Neuroimaging. 10.1016/j.bpsc.2020.09.011

Williams ZJ, Failla MD, Gotham KO, Woynaroski TG, & Cascio C (2018). Psychometric evaluation of the Short Sensory Profile in youth with autism spectrum disorder. Journal of Autism and Developmental Disorders, 48(12), 4231–4249. 10.1007/s10803-018-3678-7 [PubMed: 30019274]

Wong YS, Yang C-C, Stewart L, Chiang C-H, Wu C-C, & Iao L-S (2018). Use of the Chinese version Modified Checklist for Autism in Toddlers in a high-risk sample in Taiwan. Research in Autism Spectrum Disorders, 49, 56–64. 10.1016/j.rasd.2018.01.010

Yuen T, Penner M, Carter MT, Szatmari P, & Ungar WJ (2018). Assessing the accuracy of the Modified Checklist for Autism in Toddlers: A systematic review and meta-analysis. Developmental Medicine & Child Neurology, 60(11), 1093–1100. 10.1111/dmcn.13964 [PubMed: 29992541]

Zelaya CE, Lucas JW, & Hoffman HJ (2015). Percentage of adults with selected hearing problems, by type of problem and age group—National Health Interview Survey, United States, 2014. Morbidity and Mortality Weekly Report, 64(37), 1058.

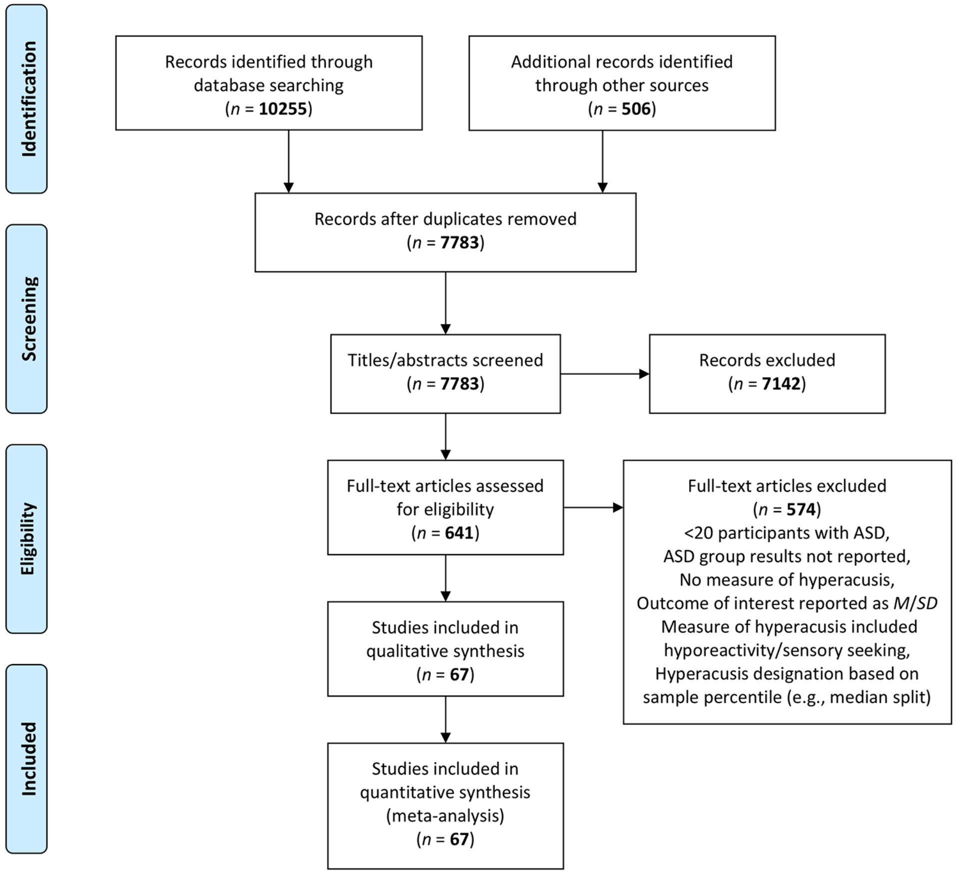

Figure 1. PRISMA flow diagram detailing the identification, screening and selection of studies for inclusion in the analysis

Note. “Database searching” includes searches of PubMed, ProQuest, ProQuest Dissertations and Theses, and targeted Google Scholar searches. Studies identified through other sources include 470 abstracts from the 2015–2019 International Society for Autism Research (INSAR) annual meetings and 46 potentially eligible studies identified by forward and backward citation tracing of included articles. Specific keywords and full details of the search procedure are available in the Supplemental Methods.

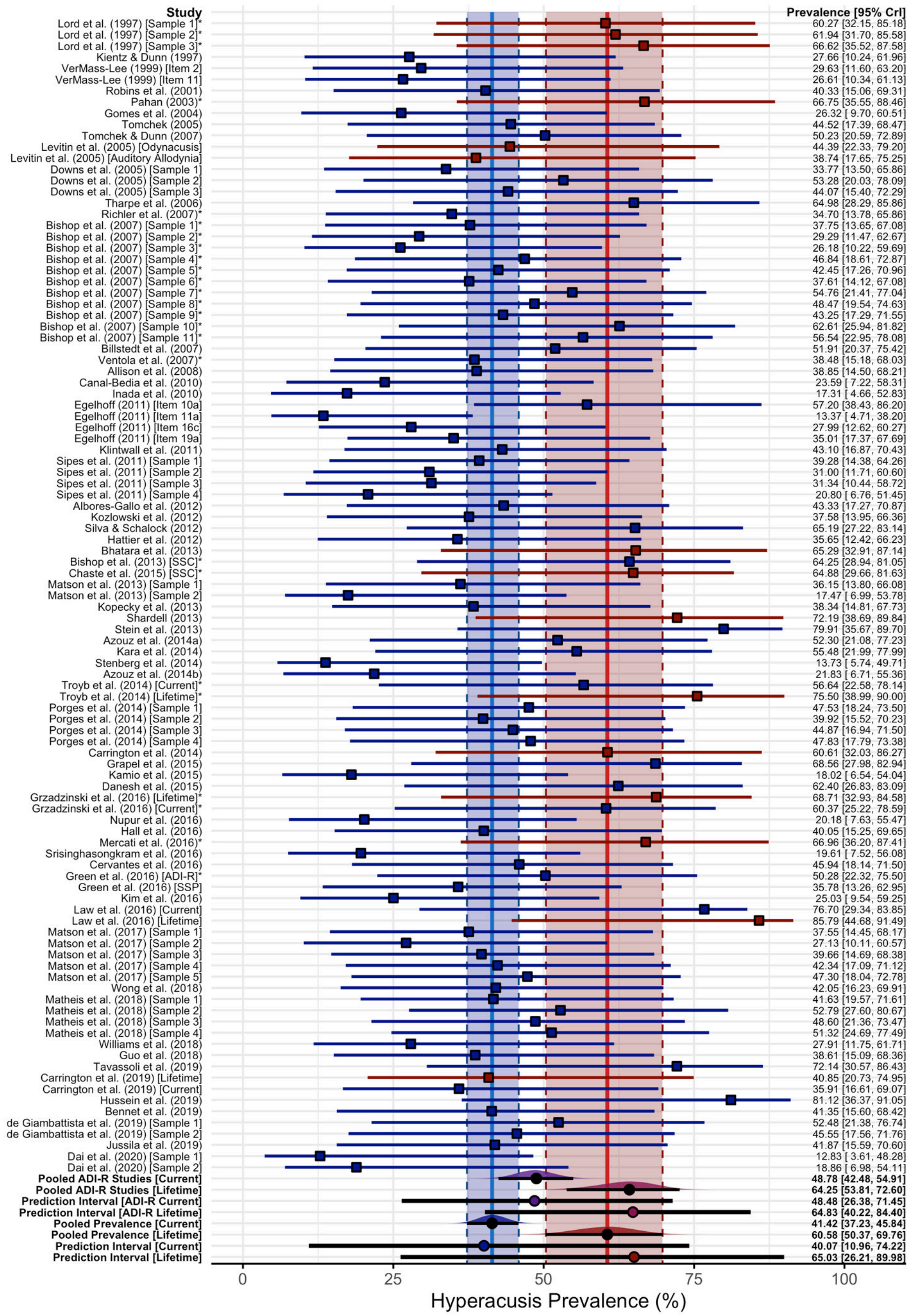

Figure 2. Posterior density forest plot of current and lifetime hyperacusis prevalence in individuals with autism spectrum disorder using subjective measures

Note. Current prevalence estimates are colored blue, while lifetime prevalence estimates are colored red. Studies utilizing the Autism Diagnostic Interview–Revised (ADI-R) are marked with an asterisk (*). The estimated prevalence and 95% credible interval (CrI) for each study represent the posterior distribution of the prevalence in that sample, conditional on prior beliefs and the observed data. Raw prevalence values from each study can be found in Supplemental Table S1.

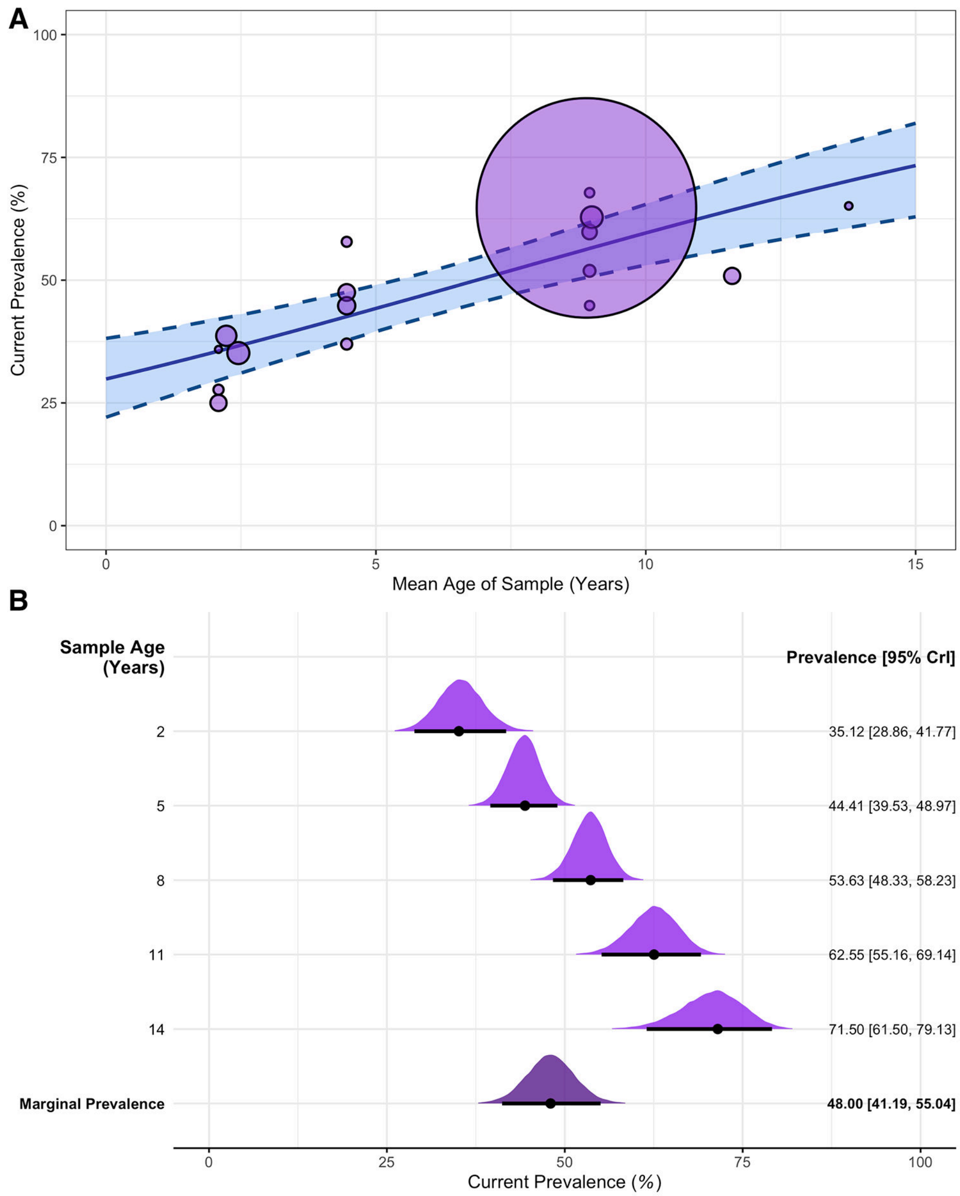

Figure 3. Moderation of current hyperacusis prevalence (ADI-R estimates) by sample age

(A) Meta-analytic scatter plot depicting the relationship between mean age and current hyperacusis prevalence estimates in studies using the Autism Diagnostic Interview–Revised (ADI-R). Plotted points are proportional to the sample size of each study. Fitted values from the Bayesian meta-regression model indicate that the prevalence of hyperacusis in autism spectrum disorder (ASD) increases substantially between early childhood and adolescence. **(B)** Posterior density plot of model-estimated prevalence values at ages 2, 5, 8, 11, and 14 years, compared to the marginal point prevalence of hyperacusis as measured by ADI-R item 72.

Table 1

Meta-analytic Prevalence Estimates, Prediction Intervals, and Heterogeneity Estimates

Prevalence Model	Effects	Samples	n_{ASD}	$n_{Hyperacusis}$	Current Prevalence [95% CrI]	95% PI (Current)	Lifetime Prevalence [95% CrI]	95% PI (Lifetime)	I^2 [95% CrI]	*ICC*(2)
Subjective Measures	103	87	13093	6989	41.42% [37.23, 45.84]	[10.96, 74.22]	60.58% [50.37, 69.76]	[26.21, 89.98]	95.5% [94.0, 96.8]	0.394
ADI-R Only	25	22	5028	3003	48.78% [42.48, 54.91]	[26.38, 71.45]	64.25% [53.81, 72.60]	[40.22, 84.40]	91.8% [85.7, 95.6]	0.369
Objective Measures	8	7	488	150	27.30% [14.92, 44.21]	[0.68, 70.54]	—	—	92.1% [80.8, 99.1]	0.504

Note. "Subjective Measures" refers to all self- or proxy-report questionnaire or interview measures, whereas "Objective Measures" refers to all structured or unstructured behavioral observations and psychoacoustic tasks such as loudness discomfort level testing. Effects = number of prevalence estimates in the model ($k_{(3)}$); Samples = number of independent samples in the model ($k_{(2)}$; not necessarily equal to the number of studies, as a number of studies provided data on multiple subgroups treated as independent samples); CrI = highest-density credible interval; PI = posterior predictive interval (i.e., prediction interval); I^2 = multi-level standardized heterogeneity estimate (includes both level 2 and 3 heterogeneity); *ICC*(2) = proportion of total heterogeneity attributable to level 2 (between-study) variance; ADI-R = Autism Diagnostic Interview–Revised.

Table 2

Bayes Factors and Heterogeneity Explained for Each Meta-Regression Model

Moderator Variable	Subjective Model		ADI-R Model	
	BF_{10}	R^2_{Het}	BF_{01}	R^2_{Het}
Publication Year	*0.018*	*0.001*	0.476	0.260
Sample Size (log-transformed)	1.286	0.059	*0.251*	*0.006*
Mean Age	1.528	0.101	**93.100**	**0.692**
Proportion Female	0.404	−0.005	1.057	<0.001
Proportion IQ/DQ < 70	0.466	0.010	1.521	0.152
Study Quality Score	0.462	−0.008	1.175	0.039
HDI	0.856	0.012	1.004	−0.017

Note. Values in bold indicate that a variable was a significant moderator in that model (i.e., $BF_{10} > 3$), whereas values in italics indicate that the moderating relationship of that variable was rejected (i.e., $BF_{10} < 1/3$). All other values are inconclusive. BF_{01} = Model comparison Bayes factor; R^2_{Het} = proportion of total heterogeneity explained by the moderator variable; ADI-R = Autism Diagnostic Interview–Revised; IQ/DQ = Intelligence Quotient/Developmental Quotient; ASD = Autism Spectrum Disorder; HDI = United Nations 2019 Human Development Index.