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

Supplemental Information

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Supplemental Methods

Search Strategy and Article Selection

PubMed was queried twice on using the following Boolean strings:

1. ((((autis*[Abstract] OR asperger*[Abstract] OR PDD*[Abstract] OR ASD[Abstract])) OR (autis*[Title] OR asperger*[Title] OR PDD*[Title] OR ASD[Title]))) AND ((hyperacusi* OR phonophobia OR misophonia))
2. (((((autis*[Title] OR asperger*[Title] OR PDD[Title] OR ASD[Title])) OR (autis*[Abstract] OR asperger*[Abstract] OR PDD[Abstract] OR ASD[Abstract]))) AND (sound* OR noise* OR auditory OR loud*)) AND (sensitiv* OR hypersensitiv* OR hyper-sensitiv* OR reactiv* OR hyper-reactiv* OR hyperreactiv* OR over-reactiv* OR overreactiv* OR overresponsiv* or over-responsiv* or hyperresponsiv* or hyper-responsiv* OR intolerance OR fear OR phobi* OR react* OR discomfort OR uncomfort*)

Filters were utilized such that results were published between 1/1/1993 and 09/29/2019, included human studies only, and were published in English. We conducted two analogous searches of the peer-reviewed literature using the ProQuest search engine. The Boolean strings used were:

1. noft((autis* OR asperger* OR PDD* OR ASD)) AND (hyperacusi* OR phonophobia OR misophonia) AND stype.exact("Scholarly Journals") AND la.exact("English") AND subt.exact("humans") AND PEER(yes) AND pd(>19930101))
2. noft(autis* OR asperger* OR PDD OR ASD) AND ((sound* OR noise* OR auditory OR loud*) AND (sensitiv* OR hypersensitiv* OR hyper-sensitiv* OR reactiv* OR hyper-reactiv* OR hyperreactiv* OR over-reactiv* OR overreactiv* OR overresponsiv* OR over-responsiv* OR hyperresponsiv* OR hyper-responsiv* OR intolerance OR fear OR phobi* OR react* OR discomfort OR uncomfort*)) AND stype.exact("Scholarly Journals") AND la.exact("English") AND subt.exact("humans") AND PEER(yes) AND pd(>19930101))

The ProQuest Dissertations and Theses database was queried separately using the following Boolean strings:

1. noft((autis* OR asperger* OR PDD* OR ASD)) AND (hyperacusi* OR phonophobia OR misophonia) AND ("Dissertations & Theses") AND la.exact("English") AND subt.exact("humans") AND PEER(yes) AND pd(>19930101))

2. noft(autis* OR asperger* OR PDD OR ASD) AND ((sound* OR noise* OR auditory OR loud*) AND noft(sensitiv* OR hypersensitiv* OR hyper-sensitiv* OR reactiv* OR hyper-reactiv* OR hyperreactiv* OR over-reactiv* OR overreactiv* OR overresponsiv* OR over-responsiv* OR hyperresponsiv* OR hyper-responsiv* OR intolerance OR fear OR phobi* OR react* OR discomfort OR uncomfort*)) AND ("Dissertations & Theses") AND la.exact("English") AND subt.exact("humans") AND PEER(yes) AND pd(>19930101))

Targeted Google Scholar searches were used to identify articles containing item-level data for one of several measures known to assess decreased sound tolerance. These measures included the Autism Diagnostic Interview–Revised (ADI-R; Lord et al., 1994), (Short) Sensory Profile ([S]SP; Dunn, 1999; McIntosh et al., 1999), Modified Checklist or Autism in Toddlers (M-CHAT; Robins et al., 2001), M-CHAT Revised with Follow-up (M-CHAT-R/F; Robins et al., 2014), and the Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT; Matson et al., 2011). Articles containing relevant item-level data from these instruments were found using the following search strings:

1. ("autism diagnostic interview" OR "ADI-R") AND ("sensitivity to noise" OR "item 72")
2. (autism OR asperger OR PDD) AND "sensory profile" AND ("loud noises" OR "unexpected or loud noises" OR "unexpected loud noises")
3. (autism OR asperger OR PDD OR ASD) AND ("M-CHAT" OR "modified checklist for autism in toddlers") AND ("oversensitive to noise" OR "item 11")
4. (autism OR asperger OR PDD OR ASD) AND ("M-CHAT R/F" OR "modified checklist for autism in toddlers revised") AND ("upset by everyday noises" OR "item 12")
5. "BISCUIT" AND "Reactions to normal, everyday sounds"

A targeted gray literature search was performed by querying the past five years of abstracts (2015–2019) for the International Society for Autism Research (INSAR) annual meeting using the search string "auditory OR loud OR noise OR sound OR hyperacusis OR phonophobia OR misophonia."

All database searches were performed on 09/29/2019, with the targeted Google Scholar searches repeated on 9/18/2020.

Abstracts of all articles were reviewed by author ZJW, an MD/PhD candidate with previous experience conducting and assisting with meta-analytic projects. Full-text review was conducted by the same author, with author ES (a graduate student in biomedical sciences with previous meta-analysis experience) independently reviewing 20% of these articles (including all studies flagged by ZJW for inclusion) to allow for the calculation of inter-rater agreement (quantified using the percentage of agreement and Cohen's (1960) Kappa). Any disagreements at this stage were discussed by the two authors until consensus was reached. All articles included in the meta-analysis were then subjected to forward and backward citation tracing using Google Scholar, and potentially relevant articles were selected by author ZJW based on full-text review. Author ES independently reviewed the full texts of these articles and determined their eligibility for inclusion. Again, all disagreements were discussed and settled once consensus was reached. This process concluded on 09/18/2020, and all studies published after this date were deemed ineligible for the meta-analysis.

Articles included in the meta-analysis were required to satisfy a number of selection criteria. First, each included article needed to report relevant outcome data on a sample of at least 20 individuals of any age with confirmed diagnoses of DSM-5 autism spectrum disorder (ASD) or any previous iteration of this diagnosis defined in the DSM-IV, DSM-IV-TR, or ICD-10 (e.g., autistic disorder, Asperger's disorder, pervasive developmental disorder–not otherwise specified, etc.), excluding Rett Syndrome. Diagnoses of ASD were not required to be confirmed by the research team and could be either self-reported, determined by clinical evaluation, based on a standardized instrument, or listed in a database/medical record. Notably, individuals screening

positive for ASD on the M-CHAT or similar measure was not sufficient for classification of an individual as having ASD. Studies including both individuals with and without ASD were excluded if outcome of interest was not reported for the ASD group alone.

Furthermore, all included articles were required to provide the information necessary to calculate the prevalence of hyperacusis in the ASD group using cross-sectional data. Notably, experimental studies that reported cross-sectional prevalence rates in ASD samples (e.g., Porges et al., 2014) were not excluded. No clinical consensus currently exists regarding the optimal diagnostic procedure for hyperacusis in ASD, and researchers have defined hyperacusis "caseness" in this population in myriad ways. Thus, in order to best capture the ways in which hyperacusis has been defined in the ASD literature, we utilized study authors' individual definitions of hyperacusis (or synonymous terms such as "sound sensitivity" or "auditory over-responsivity") to estimate prevalence rates in each study. However, we chose to exclude studies with definitions of hyperacusis based on (a) heightened sensory reactivity in multiple modalities (e.g., both auditory and tactile hyperreactivity; cf. S. A. Green et al., 2019), (b) measures of sensory reactivity that combine hyperreactivity with hyporeactivity and/or sensory seeking behaviors (e.g., the SP Auditory Processing subscale; cf. Matsuzaki et al., 2012), and (c) median splits (S. A. Green et al., 2019) and similar procedures based on specific percentiles of the ASD sample's score distribution. We also excluded articles where the methods used to determine the presence of hyperacusis were not clearly stated (e.g., Thabet & Zaghloul, 2013), as well as studies in which all participants were selected to have complaints of decreased sound tolerance (e.g., Amir et al., 2018; Lucker, 2013), as we believed the prevalence estimates derived from these studies were not valid. Some author definitions of hyperacusis were based on behavioral observations of sound tolerance (Gomes et al., 2004; Rosenhall et al., 1999) or psychoacoustic tests such as loudness discomfort levels

(Demopoulos & Lewine, 2016). These "objective" measures were included in our study but analyzed in a separate meta-analytic model from the other measures. In addition to studies reporting hyperacusis according to author definitions, we further included studies that utilized face-valid questionnaire items assessing hyperacusis in ASD. These questionnaire items included both the specific items targeted in the Google Scholar search (e.g., M-CHAT item 11: "Oversensitive to noise"; Robins et al., 2001) and items on other questionnaires deemed by both authors ZJW and ES to represent hyperacusis (see Supplemental Tables S2–S3 for a full list of included measures). Studies reporting item-level data were required to report item response frequencies or the proportion of individuals endorsing a non-zero response; reports of item means and standard deviations were not sufficient for inclusion.

Data Extraction and Moderators

Data was extracted from each article by author ZJW, with author ES independently extracting data from 20% of articles. Inter-rater reliability of data extraction was calculated using the fixed-rater consistency intraclass correlation coefficient *ICC*(3,1) (Shrout & Fleiss, 1979). Outcome data of interest included the total sample size of the ASD group (n_{ASD}) and the number of individuals in the ASD group classified as having hyperacusis ($n_{Hyperacusis}$). As both n_{ASD} and $n_{Hyperacusis}$ were required for the binomial-normal meta-analysis model, we back-calculated $n_{Hyperacusis}$ for studies that reported only n_{ASD} and a prevalence proportion (or equivalent measure such as the sensitivity of a hyperacusis item for the diagnosis of ASD). In cases where the reported proportion did not approximately correspond with an integer value of $n_{Hyperacusis}$, we assumed that some individuals were missing data on hyperacusis classification and adjusted the total n_{ASD} downwards until the reported proportion produced an integer value ± 0.2 (to account for rounding error). When possible, prevalence data was gathered from as many separate sub-samples or

conditions as possible to maximize moderator variance (e.g., if data were reported separately for males and females, these results were not pooled and instead constituted two separate samples in our model). If multiple measures of hyperacusis were used in a single study (e.g., both a current and lifetime prevalence estimate), we recorded both prevalence estimates and accounted for the within-study dependency of these values in our meta-analytic model.

In addition to effect size information, we extracted a number of potential study-level moderator variables. As we included both current and lifetime estimates of hyperacusis prevalence in our study, we extracted data indicating whether each estimate was based on a lifetime estimate or not. When not specified, we assumed that measures were assessing current prevalence. Furthermore, in accordance with a recent report showing temporal changes in the degree to which individuals with ASD differ from neurotypical controls on a range of measures (Rødgaard et al., 2019), we extracted and tested the effect of publication year on hyperacusis prevalence. We further tested the moderating effect of (logarithmically transformed) sample size as a method of detecting "small-study" effects on prevalence (Sterne et al., 2000). We further analyzed aggregate sample demographics using the same indicators proposed by Lai and colleagues (2019), including the mean age of the study sample, the proportion of females in the sample, and the proportion of individuals in the sample with cognitive abilities in the intellectually disabled range (e.g., IQ or DQ < 70). Not all studies reported the mean age of a given sample, and thus we imputed this value using (a) the median age or (b) the midpoint of the reported age range in such cases. Proportion of individuals with IQ/DQ < 70 was often not reported in studies, and thus, we frequently calculated this value using the provided mean and standard deviation of IQ/DQ scores in the ASD group, assuming normally distributed scores (Lai et al., 2019). If only a mean/median IQ/DQ score was provided without a standard deviation, we calculated the approximate proportion of individuals

with IQ/DQ < 70 using a normal distribution with standard deviation of 15. Full-scale IQ/DQ variables were used preferentially for these calculations, but when only subtest scores were reported, we used nonverbal IQ/DQ or verbal IQ/DQ to approximate this value, preferring the former if both were reported. If only mental age estimates were given, the mean and standard deviation of the mental age were divided by the mean chronological age, and these values were used to approximate the normal distribution of IQ scores. If a study reported multiple sub-samples but only provided demographics for the overall sample, those demographics were applied to all sub-samples as approximates for each sub-sample's unique demographics. Lastly, in order to quantify the effect of study context on prevalence rates, we extracted the 2019 United Nations Human Development Index (HDI) for each study's country of origin (available at http://hdr.undp.org/en/content/2019-human-development-index-ranking). When data in a sample came from multiple countries, we calculated the arithmetic mean of HDI values for all participating countries. This index was also used in the meta-analysis of Lai et al. (2019), and it has been found to be related to the prevalence of other psychosomatic conditions, including chronic widespread pain, in the general population (Andrews et al., 2018). Given the increased prevalence of chronic widespread pain in countries with lower HDI (Andrews et al., 2018), we hypothesized that hyperacusis may also show an increasing prevalence as HDI decreases.

Although the MOOSE guidelines (Stroup et al., 2000) suggest that study quality be evaluated in meta-analyses, there is a scant literature on appropriate quality indicators for cross-sectional prevalence studies (Zeng et al., 2015), and the reliability of tools used for this purpose is often poor (Shamliyan et al., 2011). We have thus devised our own set of quality criteria, based in part upon those used in a previous systematic review of pediatric hyperacusis prevalence (Nemholt et al., 2015; Rosing et al., 2016) and those used to grade ASD sample characterization in a prior

meta-analysis by our group (Z. J. Williams et al., 2020). Conceptually, we defined higher-quality studies as those including samples with better demographic and clinical characterization and hyperacusis measures that were both reliable and able to differentiate hyperacusis from other sound tolerance complaints such as misophonia. The full criteria, listed in Supplemental Table S1 and reported for each study in Supplemental Table S2, include five items concerning sample type and characterization and three items concerning the methods used to assess hyperacusis in a given sample. Author ZJW scored each study sample individually on all eight quality criteria. Scores on this measure are calculated by taking the mean of the eight items, and possible scores range from 0–1, with higher values indicating higher quality. This quality score was used as a potential moderator in our meta-regression analysis to determine whether study quality systematically biased the prevalence in one direction or the other.

Meta-analysis Model

As many studies on this topic report multiple estimates of hyperacusis prevalence (e.g., both current and lifetime prevalence, two questions measuring slightly different aspects of sound tolerance on a questionnaire, or multiple measures of sensory processing evaluated in the same sample), many of the extracted prevalence values were dependent on one another due to being estimated from the same study cohorts. Thus, in order to appropriately account for these dependencies, we utilized a three-level 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). Thus, the baseline random-effects model for a given analysis was a hierarchical binomial-normal generalized linear model with logit link function and random intercept terms for each study and effect size within a given study. In addition, for the two models in which current and lifetime prevalence were both estimated (i.e., subjective measures and the ADI-R sensitivity

analysis), we also included the fixed effect of measure timeframe (i.e., whether a measure assessed current or lifetime prevalence) in the baseline model, using this parameter to create model-based estimates of both current and lifetime prevalence simultaneously. Notably, as the models of current ADI-R prevalence (used in meta-regression) did not contain multiple effects per study, these models were fit as standard two-level meta-analysis models (i.e., only including a random effect of study) with all other model specifications identical to the previously described three-level models.

Parameter estimation for the meta-analytic models was performed in a Bayesian framework, fit using *Stan* (Carpenter et al., 2017) via the R package *brms* (Bürkner, 2017, 2018). Weakly informative priors were chosen for the model, with a Normal(0,1) prior on logit-scale regression coefficients (including the intercept term) and a Half-Cauchy(0.3) prior on $\tau_{(2)}$ and $\tau_{(3)}$, the standard deviations of the random intercept terms. The former prior was chosen to reflect our expectation that the overwhelming majority of intercept values would lie in the range [-2, 2] (approximately 12–88% when transformed back to a proportion), whereas the latter prior was selected based on strong parameter recovery ability in a previous simulation study of Bayesian meta-analysis models (D. R. Williams et al., 2018). Model parameters were estimated via Markov chain Monte Carlo (MCMC) using the No U-turn Sampler (Homan & Gelman, 2014). Posterior distributions of the parameters were based on 40,000 post-warmup MCMC draws from five separate Markov chains. Convergence for each model was confirmed by examination of Markov chain trace plots, as well as values of the Gelman–Rubin (1992) convergence diagnostic $\hat{R} < 1.01$ (examined in each imputed model separately in the case of multiple imputations). Parameter summaries from their posterior distributions were operationalized as the maximum *a priori* value

(MAP, i.e., the mode) and the 95% highest-density credible interval (CrI; Kruschke & Liddell, 2018).

To assess the heterogeneity of studies in a given analysis, we calculated the unstandardized level 2 and 3 variance parameters, $\tau^2_{(2)}$ and $\tau^2_{(3)}$, respectively. The overall I^2 statistic (Higgins & Thompson, 2002), reflecting heterogeneity from both levels, was also reported to provide a comparison to other meta-analyses. To further aid in the interpretation of I^2, we also report the $I^2_{(2)}$ and $I^2_{(3)}$ statistics (Cheung, 2014), which reflecting the standardized proportion of heterogeneity attributable to between-study (level 2) and within-study (level 3) variance, respectively. In addition, we calculated a model-based 95% prediction interval (IntHout et al., 2016) based on the 95% highest density interval of the posterior predictive distribution of prevalence estimates. This interval, whose variance is the sum of $\tau^2_{(2)}$ and $\tau^2_{(3)}$, provides estimates of the range of prevalence estimates that can be expected if future studies on this topic are conducted. These indices are presented in Supplemental Table S4. Notably, given the extreme skew of many of these heterogeneity estimates, we chose to represent their point estimates using medians rather than MAP values.

In addition to the baseline models, we analyzed the moderators of current hyperacusis prevalence (in both the subjective model and the ADI-R model) by fitting a series of Bayesian meta-regression models, each including one putative moderator. Each meta-regression model was then compared to its respective baseline (intercept-only) model, allowing us to determine whether the moderator explained meaningful amounts of between-study heterogeneity. The marginal likelihood of each model was calculated using the *bridgesampling* R package (Gronau et al., 2017, 2020), and the ratio of these likelihoods was used to calculate a model comparison Bayes factor (i.e., Bayes factor of augmented model 1 relative to baseline model 0: BF_{10}). Although Bayes

factors quantify evidence along a continuum, qualitative descriptions of the degree of evidence have been proposed to simplify interpretation (Jeffreys, 1961; Wagenmakers et al., 2011). In line with these guidelines, we consider Bayes factors > 3 to indicate substantial evidence in favor of the augmented model (supporting a moderator effect) and Bayes factors < 1/3 to indicate substantial evidence in favor of the baseline model (supporting the absence of a moderator effect). All meta-regression models containing significant moderators of prevalence (i.e., $BF_{10} > 3$) were further explored, and the effects of the moderator were quantified using estimated marginal means. Models with inconclusive results (i.e., $1/3 < BF_{10} < 3$) or those where the moderator's effect was likely minimal (i.e., $BF_{10} < 1/3$) were not interrogated further.

In order to quantify the effect of each moderating variable in our meta-regressions, we calculated R^2_{Het}, the standardized proportion of heterogeneity explained by the moderator, using the following equation:

$$R^2_{Het} = \frac{\tau^2_{M0} - \tau^2_{M1}}{\tau^2_{M0}}$$

where τ^2_{M0} is the (median) total heterogeneity parameter (i.e., $\tau^2_{(2)} + \tau^2_{(3)}$) for the baseline model and τ^2_{M1} is the (median) total heterogeneity parameter for the augmented model. Notably, heterogeneity estimates in the augmented model can be larger than those in the baseline model, resulting in negative values of R^2_{Het}. BF_{01} and R^2_{Het} values for all meta-regression models can be found in Table 2 of the main text.

Supplemental Table S1

Study quality rating criteria

Scale for Assessing the Quality of Included Studies: Total score is an average of all applicable items (Range 0–1)
Diagnosis and Characterization of Sample (5 items)
1. Demographic characteristics of participants in both groups are reported (0.5 points for age, 0.5 points for sex/gender)
2. Cognitive ability (IQ, DQ, or proportion of sample with intellectual disability/developmental delay) is reported for participants in the ASD group (1 point)
3. Rigorous diagnostic characterization must be assessed with standardized measures (0.5 points if diagnostic procedure included ADOS or ADI-R; additional 0.5 points if diagnostic procedure included both ADOS/ADI-R or judgment of an experienced clinician supported by ADOS/ADI-R; 0.5 points if diagnostic procedures necessary for 1 point used but for only a portion of the sample)
4. Hearing level assessed clinically (1 point if hearing level determined, including pure tone screening, OAE testing, or ABR testing; 0.5 points if hearing level self-reported [including for exclusion criteria]). Exclusion for hearing loss not necessary to receive points.
5. Sample obtained from epidemiologic population or large-scale registry (1 point; 0 points if sample derived from convenience, community, or clinical sample)
Characterization of Hyperacusis (3 items)
6. Reports specific method used to define hyperacusis (1 point for the following: if standardized measure, reports specific item content for hyperacusis item; if ad-hoc interview or questionnaire measure, provides exact wording of question(s) or specific free-responses that would qualify; if observational or objective measure, reports on the specific criteria necessary to categorize an individual as having hyperacusis).
7. Reliability of method used to determine hyperacusis (1 point if structured parent/child interview or objective/observational measure; 0.5 points if composite score on self/parent-report questionnaire, unstructured interview, or single questionnaire item with follow-up [e.g., M-CHAT R/F]; 0 points for single questionnaire item or unspecified questionnaire).
8. Hyperacusis measure differentiates between hyperacusis and other types of decreased sound tolerance (e.g., misophonia). (1 point if question(s) specifically mentions the intensity of the sound as being the aversive characteristic or attempts to exclude intolerance of very specific sound stimuli, i.e., the ADI-R; 0 points if insufficient information to determine)

Note. ASD = autism spectrum disorder; ADOS = Autism Diagnostic Observation Schedule; ADI-R = Autism Diagnostic Interview–Revised; OAE = otoacoustic emission; ABR = auditory brainstem response; M-CHAT R/F = Modified Checklist for Autism in Toddlers Revised with Follow-up

Supplemental Table S2

Samples included in meta-analysis of "subjective" measures

	Reference	Hyperacusis Measure	n_{ASD}	n_{HYP}	P_{HYP}	Prop. Female	Prop. ID/DD	Age (Yrs)	Country	Quality (0-1)	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
1	Lord et al. (1997) [Sample 1]	ADI-R (Item 72-Lifetime > 0)	142	85	0.599	0.245	0.563	14.50	Multiple	0.750	1	1	1	0	0	1	1	1
2	Lord et al. (1997) [Sample 2]	ADI-R (Item 72-Lifetime > 0)	59	36	0.610	0.245	0.000	21.40	Multiple	0.750	1	1	1	0	0	1	1	1
3	Lord et al. (1997) [Sample 3]	ADI-R (Item 72-Lifetime > 0)	91	61	0.670	0.245	1.000	11.90	Multiple	0.750	1	1	1	0	0	1	1	1
4	Kientz & Dunn (1997)	SP (Item 1 < 3)	32	8	0.250	0.188	NA	8.00	USA	0.375	1	0	0	0	0	1	0	1
5	VerMaas-Lee (1999) [Item 2]	ESP (Item 2 Always/Often)	40	10	0.250	0.171	NA	4.58	USA	0.375	1	0	0	0	0	1	0	1
5	VerMaas-Lee (1999) [Item 11]	ESP (Item 11 Always/Often)	41	9	0.220	0.171	NA	4.58	USA	0.375	1	0	0	0	0	1	0	1
6	Robins et al. (2001)	M-CHAT (Item 11 Endorsed)	39	16	0.410	NA	0.899	2.30	USA	0.313	0.5	1	0	0	0	1	0	0
7	Pahan (2003) [ADI-R]	ADI-R (Item 72-Lifetime > 0)	26	18	0.692	0.192	0.846	8.67	India	0.875	1	1	1	1	0	1	1	1
8	Gomes et al. (2004) [Interview]	Parent Interview (Current)	46	11	0.239	0.152	NA	10.85	Brazil	0.313	1	0	0	1	0	0	0.5	0
9	Tomchek (2005)	SSP (Item 34 < 3)	400	182	0.455	0.130	NA	4.13	USA	0.438	1	0	0.5	0	0	1	0	1
9	Tomchek & Dunn (2007)	SSP (Item 34 < 3)	281	143	0.509	0.164	NA	4.30	USA	0.438	1	0	0.5	0	0	1	0	1
10	Levitin et al. (2005) ["Odynacusis"]	Parent Survey (Lifetime)	30	10	0.333	0.200	0.436	18.20	USA	0.625	1	1	1	0	0	1	0	1
10	Levitin et al. (2005) ["Auditory Allodynia"]	Parent Survey (Lifetime)	30	8	0.267	0.200	0.436	18.20	USA	0.625	1	1	1	0	0	1	0	1
11	Downs et al. (2005) [Sample 1]	Parent Interview (Current)	59	20	0.339	0.203	0.274	6.08	USA	0.438	1	1	0	1	0	0	0.5	0
12	Downs et al. (2005) [Sample 2]	Parent Interview (Current)	15	9	0.600	0.000	0.000	12.33	USA	0.438	1	1	0	1	0	0	0.5	0
13	Downs et al. (2005) [Sample 3]	Parent Interview (Current)	13	6	0.462	0.231	0.447	6.25	USA	0.438	1	1	0	1	0	0	0.5	0
14	Tharpe et al. (2006)	Parent Survey (Current)	22	17	0.773	0.136	0.864	5.58	USA	0.500	1	1	0	1	0	1	0	0
15	Bishop et al. (2006) [Sample 1]	ADI-R (Item 72-Current > 0)	165	58	0.352	0.156	0.871	2.45	USA	0.750	1	1	1	0	0	1	1	1
16	Bishop et al. (2006) [Sample 2]	ADI-R (Item 72-Current > 0)	39	14	0.359	0.190	1.000	2.08	USA	0.750	1	1	1	0	0	1	1	1
17	Bishop et al. (2006) [Sample 3]	ADI-R (Item 72-Current > 0)	65	18	0.277	0.190	1.000	2.08	USA	0.750	1	1	1	0	0	1	1	1
18	Bishop et al. (2006) [Sample 4]	ADI-R (Item 72-Current > 0)	116	29	0.250	0.190	0.000	2.08	USA	0.750	1	1	1	0	0	1	1	1
19	Bishop et al. (2006) [Sample 5]	ADI-R (Item 72-Current > 0)	120	57	0.475	0.190	1.000	4.46	USA	0.750	1	1	1	0	0	1	1	1
20	Bishop et al. (2006) [Sample 6]	ADI-R (Item 72-Current > 0)	125	56	0.448	0.190	1.000	4.46	USA	0.750	1	1	1	0	0	1	1	1
21	Bishop et al. (2006) [Sample 7]	ADI-R (Item 72-Current > 0)	73	27	0.370	0.190	0.000	4.46	USA	0.750	1	1	1	0	0	1	1	1
22	Bishop et al. (2006) [Sample 8]	ADI-R (Item 72-Current > 0)	64	37	0.578	0.190	0.000	4.46	USA	0.750	1	1	1	0	0	1	1	1
23	Bishop et al. (2006) [Sample 9]	ADI-R (Item 72-Current > 0)	79	41	0.519	0.190	1.000	8.96	USA	0.750	1	1	1	0	0	1	1	1
24	Bishop et al. (2006) [Sample 10]	ADI-R (Item 72-Current > 0)	58	26	0.448	0.190	1.000	8.96	USA	0.750	1	1	1	0	0	1	1	1
25	Bishop et al. (2006) [Sample 11]	ADI-R (Item 72-Current > 0)	59	40	0.678	0.190	0.000	8.96	USA	0.750	1	1	1	0	0	1	1	1
26	Richler et al. (2007)	ADI-R (Item 72-Current > 0)	102	61	0.598	0.190	0.000	8.96	USA	0.750	1	1	1	0	0	1	1	1

(Table Continues)

Supplemental Table S2, Continued

	Reference	Hyperacusis Measure	n_{ASD}	n_{HYP}	P_{HYP}	Prop. Female	Prop. ID/DD	Age (Yrs)	Country	Quality (0-1)	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
27	Billstedt et al. (2007)	DISCO ("Acuteness of Hearing"-Current)	105	56	0.533	0.286	0.952	25.50	Sweden	0.500	1	1	0	0	0	1	1	0
28	Ventola et al. (2007)	ADI-R (Item 72-Current > 0)	150	58	0.387	0.180	0.789	2.23	USA	0.750	1	1	1	0	0	1	1	1
29	Allison et al. (2008)	Q-CHAT (Item 24 > 2)	41	16	0.390	0.244	NA	2.58	UK	0.250	1	0	0	0	0	1	0	0
30	Canal-Bedia et al. (2011)	M-CHAT (Item 11 Endorsed)	23	4	0.174	NA	NA	2.42	Spain	0.625	0.5	0	1	0	1	1	0.5	1
31	Inada et al. (2011)	M-CHAT (Item 11 Endorsed)	20	1	0.050	0.200	NA	1.50	Japan	0.250	1	0	0	0	0	1	0	0
32	Egelhoff (2011) [Item 10a]	ABQ (Item 10a > 3)	169	95	0.562	NA	NA	11.60	USA	0.250	0.5	0	0	0.5	0	1	0	0
32	Egelhoff (2011) [Item 11a]	ABQ (Item 11a > 3)	169	19	0.112	NA	NA	11.60	USA	0.250	0.5	0	0	0.5	0	1	0	0
32	Egelhoff (2011) [Item 16c]	ABQ (Item 16c > 3)	169	45	0.266	NA	NA	11.60	USA	0.250	0.5	0	0	0.5	0	1	0	0
32	Egelhoff (2011) [Item 19a]	ABQ (Item 19a > 3)	169	56	0.331	NA	NA	11.60	USA	0.375	0.5	0	0	0.5	0	1	0	1
33	Klintwall et al. (2011)	Parent Interview (Current)	208	91	0.438	0.154	0.361	2.25	Sweden	0.375	1	1	0	0	0	0	1	0
34	Sipes et al. (2011) [Sample 1]	BISCUIT (Item 11 Endorsed)	221	88	0.398	0.000	0.562	2.16	USA	0.500	1	1	0	0	1	1	0	0
35	Sipes et al. (2011) [Sample 2]	BISCUIT (Item 11 Endorsed)	73	22	0.301	0.000	0.000	2.19	USA	0.500	1	1	0	0	1	1	0	0
36	Sipes et al. (2011) [Sample 3]	BISCUIT (Item 11 Endorsed)	65	20	0.308	1.000	0.545	2.22	USA	0.500	1	1	0	0	1	1	0	0
37	Sipes et al. (2011) [Sample 4]	BISCUIT (Item 11 Endorsed)	26	4	0.154	1.000	0.000	2.16	USA	0.500	1	1	0	0	1	1	0	0
35	Matheis et al. (2019) [Sample 1]	BISCUIT (Item 11 Endorsed)	462	192	0.416	0.000	0.000	2.16	USA	0.500	1	1	0	0	1	1	0	0
37	Matheis et al. (2019) [Sample 2]	BISCUIT (Item 11 Endorsed)	149	79	0.530	1.000	0.000	2.14	USA	0.500	1	1	0	0	1	1	0	0
34	Matheis et al. (2019) [Sample 3]	BISCUIT (Item 11 Endorsed)	542	265	0.489	0.000	1.000	2.19	USA	0.500	1	1	0	0	1	1	0	0
36	Matheis et al. (2019) [Sample 4]	BISCUIT (Item 11 Endorsed)	164	84	0.512	1.000	1.000	2.13	USA	0.500	1	1	0	0	1	1	0	0
38	Albores-Gallo et al. (2012)	M-CHAT (Item 11 Endorsed)	117	53	0.453	0.248	NA	4.40	Mexico	0.438	1	0	1	0.5	0	1	0	0
39	Kozlowski et al. (2012)	M-CHAT (Item 11 Endorsed)	141	52	0.369	0.241	0.385	2.05	USA	0.500	1	1	0	0	1	1	0	0
40	Silva & Schalock (2012)	SSRC (Item "Reacts poorly to certain everyday noises." > 1)	99	68	0.687	0.182	NA	3.90	USA	0.250	1	0	0	0	0	1	0	0
41	Hattier et al. (2012)	BISCUIT (Item 11 Endorsed)	21	7	0.333	0.333	NA	2.24	USA	0.500	1	1	0	0	1	1	0	0
42	Bhatara et al. (2013)	Parent Interview (Lifetime)	28	18	0.643	0.179	0.071	13.40	Canada	0.250	1	1	0	0	0	0	0	0
43	Bishop et al. (2013) [SSC Data]	ADI-R (Item 72-Current > 0)	1825	1181	0.647	0.140	0.314	8.90	USA	0.938	1	1	1	0.5	1	1	1	1
43	Chaste et al. (2015) [SSC Data]	ADI-R (Item 72-Lifetime > **1**)	2576	1676	0.651	0.140	0.314	8.90	USA	0.938	1	1	1	0.5	1	1	1	1
44	Matson et al. (2013) [Sample 1]	M-CHAT (Item 11 Endorsed)	150	54	0.360	0.273	NA	1.71	USA	0.500	1	1	0	0	1	1	0	0
45	Matson et al. (2013) [Sample 2]	M-CHAT (Item 11 Endorsed)	101	16	0.158	0.248	NA	2.10	USA	0.500	1	1	0	0	1	1	0	0
46	Kopecky et al. (2013)	Parent Survey (Current)	80	31	0.388	0.300	NA	14.60	USA	0.375	1	0	0	0	0	1	0	1
47	Shardell (2013)	FYI (Item 2 Sometimes/Often)	96	70	0.729	0.135	0.000	1.00	USA	0.563	1	1	0	0.5	1	1	0	0
48	Stein et al. (2013)	Parent Survey (Current)	182	150	0.824	0.187	NA	9.24	USA	0.125	1	0	0	0	0	0	0	0
49	Azouz et al. (2014a)	Parent Survey (Current)	30	17	0.567	0.233	1.000	5.45	Egypt	0.500	1	1	1	1	0	0	0	0

(Table Continues)

Supplemental Table S2, Continued

	Reference	Hyperacusis Measure	n_{ASD}	n_{HYP}	P_{HYP}	Prop. Female	Prop. ID/DD	Age (Yrs)	Country	Quality (0-1)	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
50	Kara et al. (2014)	ADI-R (Item 72-Current > 0)	45	27	0.600	NA	NA	2.00	Turkey	0.188	0.5	0	0	0	0	1	0	0
51	Stenberg et al. (2014)	ADI-R (Item 72-Lifetime > 0)	173	20	0.116	0.133	NA	1.50	Norway	0.250	1	0	0	0	1	0	0	0
52	Azouz et al. (2014b)	Parent Interview (Current)	30	4	0.133	0.133	NA	5.77	Egypt	0.188	1	0	0	0	0	0	0.5	0
53	Troyb et al. (2014) [Current]	ADI-R (Item 72-Current > 0)	43	28	0.651	0.089	0.000	13.76	USA	0.813	1	1	1	0.5	0	1	1	1
53	Troyb et al. (2014) [Lifetime]	ADI-R (Item 72-Lifetime > 0)	43	34	0.791	0.089	0.000	13.76	USA	0.813	1	1	1	0.5	0	1	1	1
54	Porges et al. (2014) [Sample 1]	Parent Survey (Current)	36	18	0.500	0.306	NA	4.61	USA	0.188	1	0	0.5	0	0	0	0	0
55	Porges et al. (2014) [Sample 2]	Parent Survey (Current)	28	12	0.429	0.179	NA	4.39	USA	0.188	1	0	0.5	0	0	0	0	0
56	Porges et al. (2014) [Sample 3]	Parent Survey (Current)	50	23	0.460	0.120	NA	4.44	USA	0.188	1	0	0.5	0	0	0	0	0
57	Porges et al. (2014) [Sample 4]	Parent Survey (Current)	32	16	0.500	0.156	NA	4.73	USA	0.188	1	0	0.5	0	0	0	0	0
58	Carrington et al. (2014)	DISCO ("Distress caused by sounds"-Lifetime)	36	22	0.611	0.111	0.500	7.13	UK	0.500	1	1	0	0	0	1	1	0
59	Grapel et al. (2015)	Endorsed "Upset by loud sounds"	529	365	0.690	NA	NA	19.06	USA	0.313	0.5	0	0	0	0	1	0	1
60	Kamio et al. (2015)	M-CHAT (Item 11 Endorsed)	51	7	0.137	0.314	0.510	1.55	Japan	0.500	1	1	1	0	0	1	0	0
61	Danesh et al. (2015)	HQ Total Score > 28	55	38	0.691	0.164	NA	17.80	USA	0.500	1	0	0	0.5	0	1	0.5	1
62	Grzadzinski et al. (2016) [Lifetime]	ADI-R (Item 72-Lifetime > 0)	164	113	0.689	0.159	0.000	9.00	USA	0.750	1	1	1	0	0	1	1	1
62	Grzadzinski et al. (2016) [Current]	ADI-R (Item 72-Current > 0)	164	103	0.628	0.159	0.000	9.00	USA	0.750	1	1	1	0	0	1	1	1
63	Nupur et al. (2016)	SSP (Item 34 < 3)	80	14	0.175	0.150	NA	5.89	Bangladesh	0.375	1	0	0	0	0	1	0	1
64	Hall et al. (2016)	Child Interview (Current)	29	12	0.414	NA	NA	11.00	UK	0.563	0.5	0	0	1	1	1	1	0
65	Srisinghasongkram et al. (2016)	M-CHAT (Item 11 Endorsed)	54	9	0.167	0.185	NA	2.50	Thailand	0.250	1	0	0	0	0	1	0	0
66	Green et al. (2016)[ADI-R]	ADI-R (Item 72-Current > 0)	116	59	0.509	0.130	0.433	11.60	UK	0.875	1	1	1	0	1	1	1	1
66	Green et al. (2016) [SSP]	SSP (Item 34 < 3)	116	43	0.371	0.130	0.433	11.60	UK	0.750	1	1	1	0	1	1	0	1
67	Kim et al. (2016)	M-CHAT (Item 11 Endorsed)	58	13	0.224	0.345	0.677	2.00	USA	0.688	1	1	1	0.5	1	1	0	0
68	Law et al. (2016) [Current]	Parent Survey (Current)	814	631	0.775	0.242	NA	10.30	USA	0.250	1	0	0	0	1	0	0	0
68	Law et al. (2016) [Lifetime]	Parent Survey (Lifetime)	814	704	0.865	0.242	NA	10.30	USA	0.250	1	0	0	0	1	0	0	0
69	Cervantes et al. (2017)	BISCUIT (Item 11 Endorsed)	370	171	0.462	0.243	NA	2.22	USA	0.500	1	1	0	0	1	1	0	0
70	Mercati et al. (2017)	ADI-R (Item 72-Lifetime > 0)	596	399	0.669	NA	NA	NA	Multiple	0.750	0	0	1	1	1	1	1	1
71	Matson et al. (2017) [Sample 1]	BISCUIT (Item 11 Endorsed)	39	15	0.385	0.179	NA	2.39	Greece	0.375	1	0	1	0	0	1	0	0
72	Matson et al. (2017) [Sample 2]	BISCUIT (Item 11 Endorsed)	50	12	0.240	0.280	NA	2.48	Italy	0.375	1	0	1	0	0	1	0	0
73	Matson et al. (2017) [Sample 3]	BISCUIT (Item 11 Endorsed)	49	19	0.388	0.245	NA	2.47	Japan	0.250	1	0	0	0	0	1	0	0
74	Matson et al. (2017) [Sample 4]	BISCUIT (Item 11 Endorsed)	58	26	0.448	0.259	NA	2.42	Poland	0.250	1	0	0	0	0	1	0	0
75	Matson et al. (2017) [Sample 5]	BISCUIT (Item 11 Endorsed)	54	26	0.481	0.130	NA	2.38	USA	0.250	1	0	0	0	0	1	0	0

(Table Continues)

Supplemental Table S2, Continued

	Reference	Hyperacusis Measure	n_{ASD}	n_{HYP}	P_{HYP}	Prop. Female	Prop. ID/DD	Age (Yrs)	Country	Quality (0-1)	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
76	Wong et al. (2018)	M-CHAT (Item 11 Endorsed)	113	49	0.434	0.106	0.616	2.60	China	0.375	1	1	0	0	0	1	0	0
77	Williams et al. (2018)	SSP (Item 34 < 3)	388	108	0.278	0.201	NA	7.34	USA	0.688	1	0	0.5	0	1	1	1	1
78	Guo et al. (2019)	M-CHAT-R/F (Item 12 Endorsed)	82	32	0.390	NA	NA	1.89	China	0.375	0.5	0	0	0	0	1	0.5	1
79	Tavassoli et al. (2019) [SSP]	SSP > 1 SD above control mean	76	58	0.763	0.132	NA	9.60	USA	0.563	1	1	0.5	0	0	1	0	1
80	Carrington et al. (2019) [Lifetime]	DISCO ("Distress caused by sounds"-Lifetime)	71	26	0.366	0.352	0.000	34.89	UK	0.500	1	1	0	0	0	1	1	0
80	Carrington et al. (2019) [Current]	DISCO ("Distress caused by sounds"-Current)	71	24	0.338	0.352	0.000	34.89	UK	0.500	1	1	0	0	0	1	1	0
81	Hussein et al. (2019)	Parent Survey (Current)	90	77	0.856	0.233	NA	10.50	Canada	0.250	1	0	0	0	0	1	0	0
82	Bennet et al. (2019)	Parent Survey (Current)- "Moderate" or "Severe"	604	252	0.417	0.810	NA	12.10	USA	0.375	1	0	0	0	1	1	0	0
83	de Giambattista et al. (2019) [Sample 1]	ASAS ("Unusual fear/distress due to unexpected noises")	80	45	0.563	0.825	0.000	11.50	Italy	0.375	1	1	0	0	0	1	0	0
84	de Giambattista et al. (2019) [Sample 2]	ASAS ("Unusual fear/distress due to unexpected noises")	70	32	0.457	0.871	0.000	8.90	Italy	0.375	1	1	0	0	0	1	0	0
85	Jussila et al. (2020)	Parent Survey (Current)	28	12	0.429	0.393	NA	8.00	Finland	0.500	1	0	1	0	1	1	0	0
86	Dai et al. (2020) [Sample 1]	M-CHAT-R/F (Item 12 Endorsed)	41	2	0.049	0.402	NA	1.59	USA	0.563	1	0	1	0	0	1	0.5	1
87	Dai et al. (2020) [Sample 2]	M-CHAT-R/F (Item 12 Endorsed)	77	12	0.156	0.361	NA	1.51	USA	0.563	1	0	1	0	0	1	0.5	1

Note. Multiple independent samples from within the same study are given separate sample numbers. Overlapping samples have the same sample number. n_{ASD} = number of participants with ASD on whom hyperacusis data is available (may differ from total *N* in study); n_{HYP} = number of individuals with ASD classified as having hyperacusis in the sample; P_{HYP} = estimate of hyperacusis prevalence calculated from n_{ASD} and n_{HYP}; Prop. Female = proportion of females in the sample; Prop. ID/DD = proportion of individuals in sample with cognitive ability (IQ or DQ in the intellectual/developmental disability range [< 70]); Quality = mean of study quality items Q1–Q8 (listed in Supplemental Table S1). Scores range from 0–1, with higher scores indicating higher study quality overall; SSC = Simons Simplex Collection(Fischbach & Lord, 2010); (S)SP = (Short) Sensory Profile; ESP = Evaluation of Sensory Processing; M-CHAT (R/F) = Modified Checklist for Autism in Toddlers (Revised with Follow-up); DISCO = Diagnostic Interview for Social Communication Disorders; Q-CHAT = Quantitative Checklist for Autism in Toddlers; ABQ = Auditory Behavior Questionnaire; BISCUIT = Baby and Infant Screen for Children with aUtIsm Traits; SSRC = Sense and Self-Regulation Checklist; FYI = First Year Inventory; ASAS = Australian Scale for Asperger's Syndrome.

Supplemental Table S3

Samples included in the meta-analysis of "objective" measures

	Reference	Hyperacusis Measure	n_{ASD}	n_{HYP}	P_{HYP}	Prop. Female	Prop. ID/DD	Age (Yrs)	Country	Quality (0-1)	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
88	Rosenhall et al. (1999)	Observed intolerance of 80 dB click	111	20	0.18	0.231	0.724	7.32	Sweden	0.750	1	1	0	1	0	1	1	1
7	Pahan (2003) [Observation]	Ritvo-Freeman Real Life Rating Scale	26	15	0.577	0.192	0.846	8.67	India	0.750	1	1	1	1	0	1	1	0
8	Gomes et al. (2004) [Observation]	Observed intolerance of 90 dB warble	46	2	0.043	0.152	—	10.85	Brazil	0.500	1	0	0	1	0	0	1	1
89	Tan et al. (2012) [Hypersensitivity]	Direct observation and parent report	156	77	0.494	0.09	—	3.5	China	0.500	1	0	0	1	0	1	1	0
89	Tan et al. (2012) [Phonophobia]	Direct observation and parent report	156	55	0.353	0.09	—	3.5	China	0.500	1	0	0	1	0	1	1	0
90	Demopoulos & Lewine (2016)	Speech LDL < 3 SD below typical mean	41	15	0.366	0.2	0.297	10.78	USA	0.875	1	1	1	1	0	1	1	1
91	Tavassoli et al. (2016)	SP:3D-A	32	5	0.156	0.2	0	8.7	USA	0.625	1	1	1	0	0	1	1	0
79	Tavassoli et al. (2019) [SP:3D-A]	SP:3D-A	76	16	0.211	0.132	—	9.6	USA	0.563	1	1	0.5	0	0	1	1	0

Note. Studies that reported a subjective measure are given the same sample number as previously. n_{ASD} = number of participants with ASD on whom hyperacusis data is available (may differ from total *N* in study); n_{HYP} = number of individuals with ASD classified as having hyperacusis in the sample; P_{HYP} = estimate of hyperacusis prevalence calculated from n_{ASD} and n_{HYP}; Prop. Female = proportion of females in the sample; Prop. ID/DD = proportion of individuals in sample with cognitive ability (IQ or DQ in the intellectual/developmental disability range [< 70]); Quality = mean of study quality items Q1–Q8 (listed in Supplemental Table S1). Scores range from 0–1, with higher scores indicating higher study quality overall; LDL = loudness discomfort level; SP:3D-A = Sensory Processing Three-Dimensions Assessment (formerly the Sensory Processing Scales Assessment).

Supplemental Table S4.

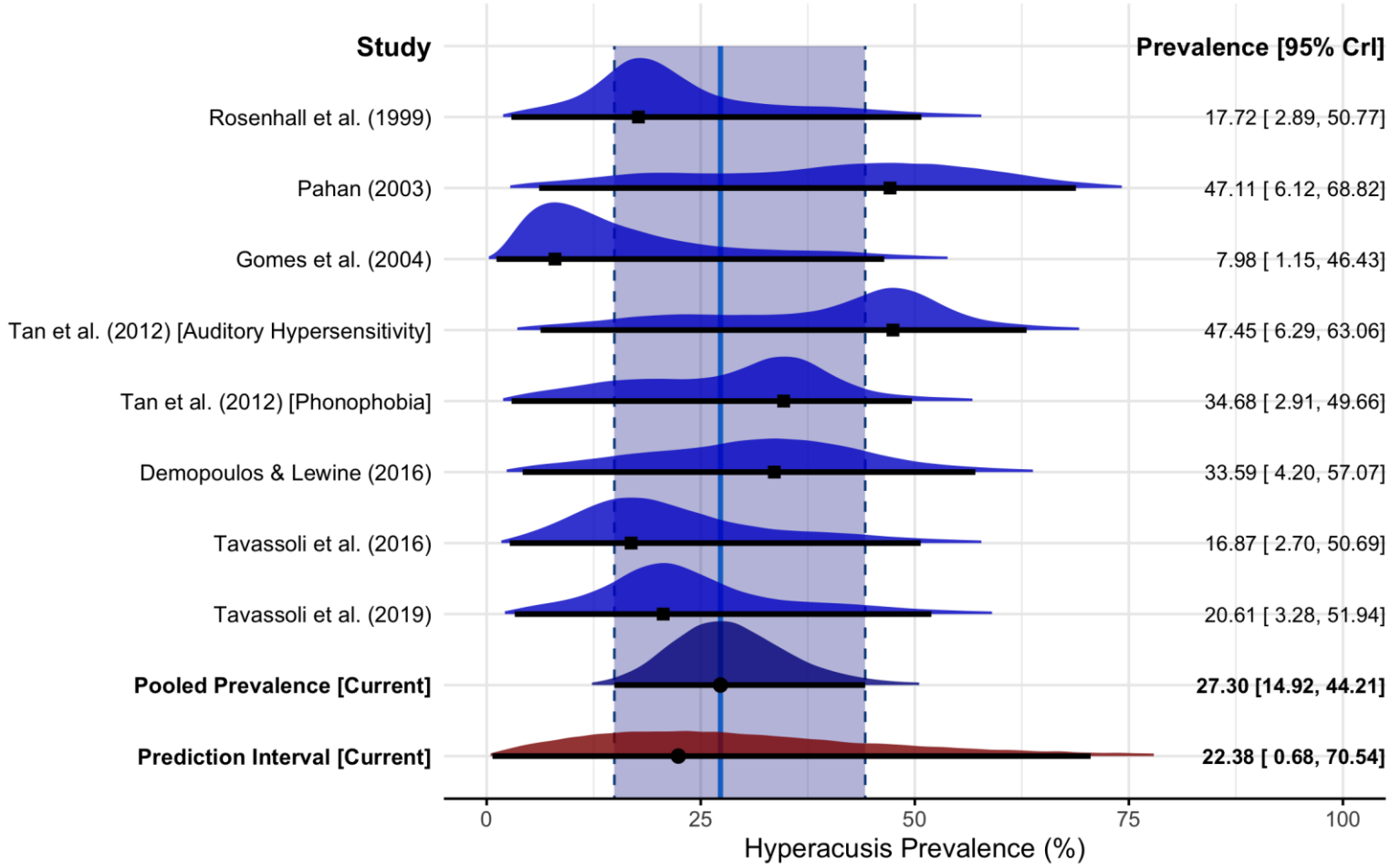
Heterogeneity metrics for each meta-analysis model

Outcome	$\tau^2_{(2)}$ [95% CrI]	$\tau^2_{(3)}$ [95% CrI]	I^2 [95% CrI]	$I^2_{(2)}$ [95% CrI]	$I^2_{(3)}$ [95% CrI]	95% PI (Current)	95% PI (Lifetime)
Subjective Measures	0.231 [0, 0.501]	0.361 [0.137, 0.683]	95.5% [94.0, 96.8]	37.6% [0.0, 69.0]	57.8% [26.6, 96.5]	[10.96, 74.22]	[26.21, 89.98]
ADI-R Only	0.070 [0, 0.289]	0.117 [0, 0.290]	91.8% [85.7, 95.6]	33.0% [0.0, 87.2]	57.8% [4.8, 94.3]	[26.38, 71.45]	[40.22, 84.40]
Objective Measures	0.304 [0, 2.683]	0.309 [0.004, 2.296]	92.1% [80.8, 99.1]	45.3% [0.1, 95.3]	44.3% [0.4, 95.6]	[0.68, 70.54]	—

Note. Point estimates presented are medians rather than modes. As a rule of thumb, I^2 values of 25%, 50% and 75% are typically considered the cutoffs for small, medium, and large amounts of heterogeneity. CrI = highest density credible interval; $\tau^2_{(2)}$ = level 2 heterogeneity (variance of the study intercept term); $\tau^2_{(3)}$ = level 3 heterogeneity (variance of the effect intercept term); I^2 = standardized heterogeneity metric (i.e., % of total variance accounted for by $\tau^2_{(2)}$ and $\tau^2_{(3)}$). $I^2_{(2)} = I^2$ for level 2 (between-study) heterogeneity only; $I^2_{(3)} = I^2$ for level 3 (within-study) heterogeneity only; PI = posterior predictive interval of effects from future studies (i.e., prediction interval); ADI-R = Autism Diagnostic Interview–Revised.

Supplemental Figure S1.

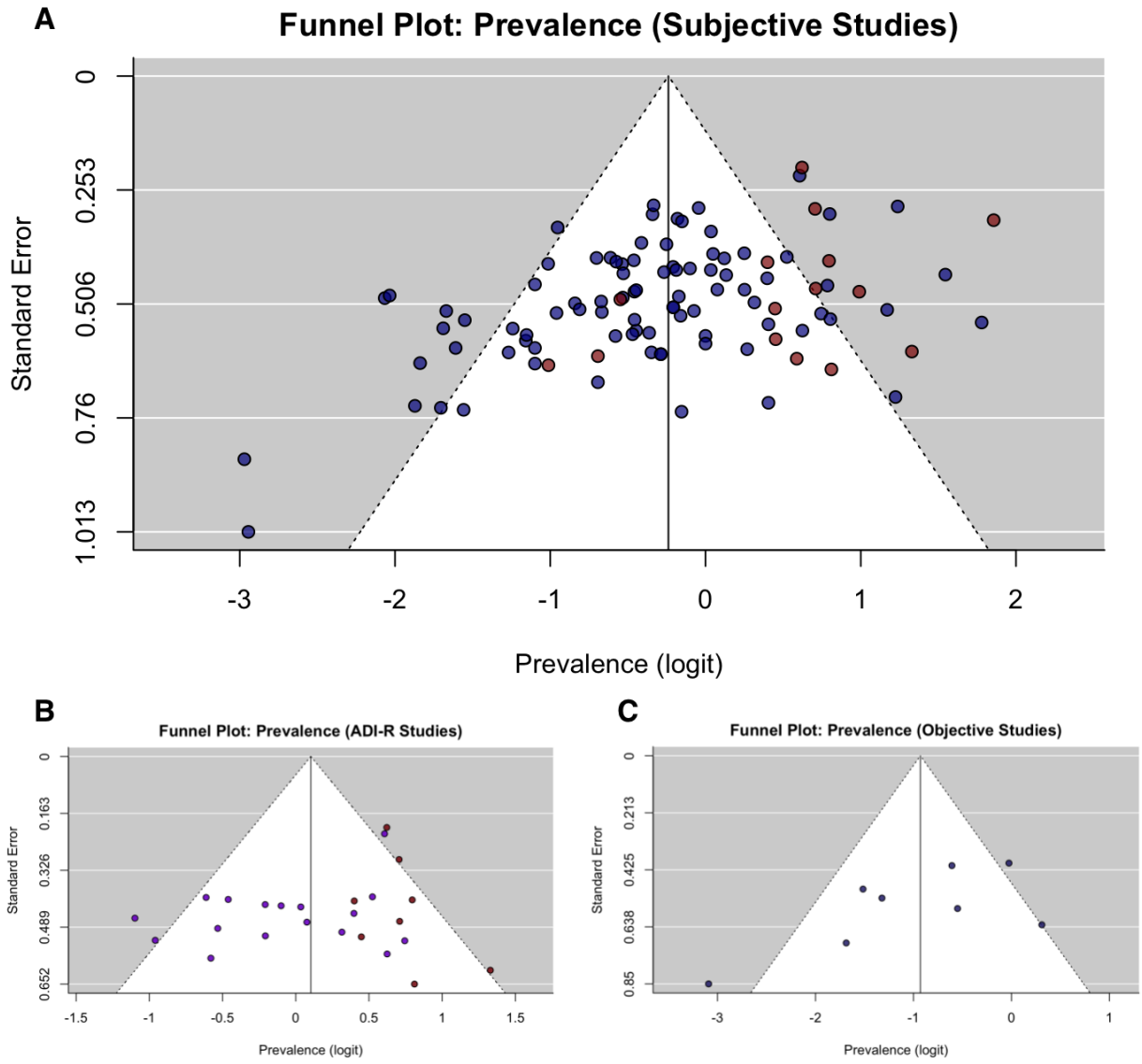
Posterior density forest plot of (current) hyperacusis prevalence measured using observational or objective measures



Note. The point estimate and 95% highest density credible interval (CrI) for each study represent the posterior distribution of that study's prevalence estimate, conditional on prior beliefs and the observed data. Raw prevalence estimates from each study can be found in Supplemental Table S3.

Supplemental Figure S2.

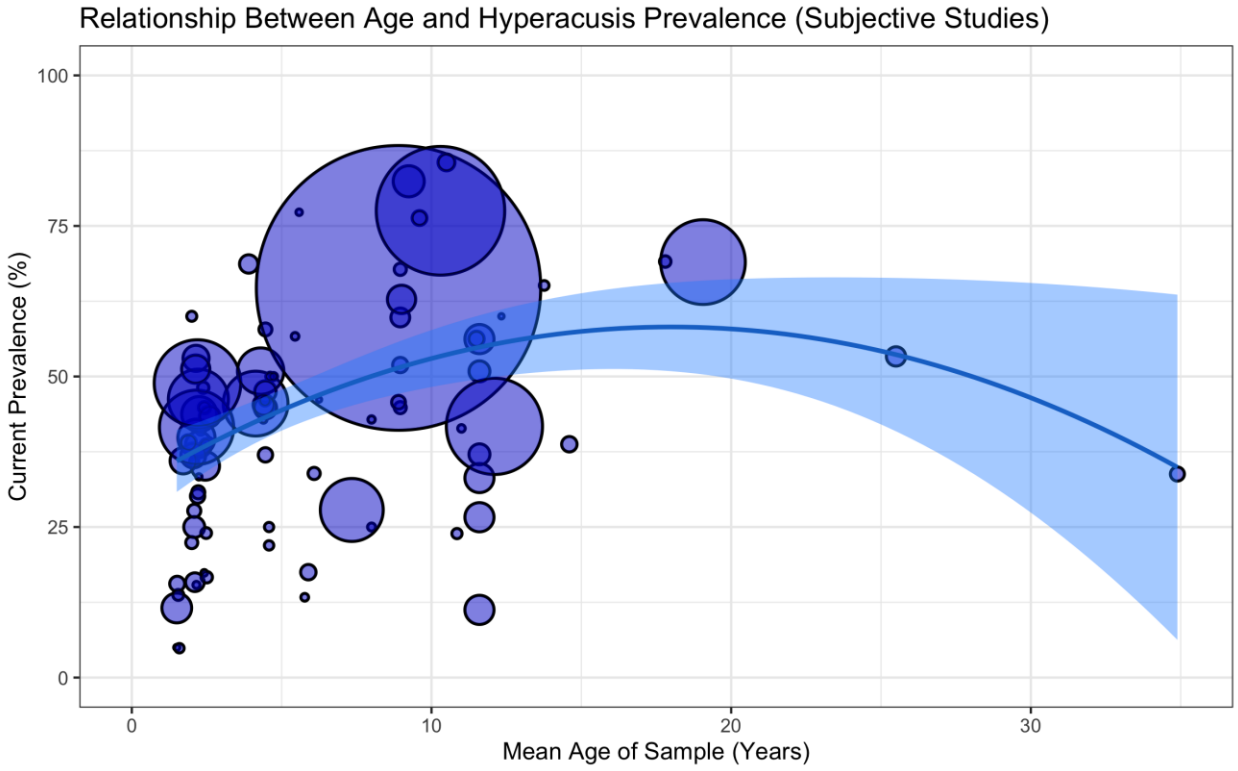
Graphical examination of publication bias in prevalence estimates using funnel plots



Note. **(A)** Funnel plot of subjective studies. Current prevalence estimates are colored blue, and lifetime estimates are colored red. **(B)** Funnel plot of ADI-R studies. Current prevalence estimates are colored purple, and lifetime estimates are colored deep pink. **(C)** Funnel plot of objective studies. Relative symmetry around the summary effect in all cases demonstrates a general lack of publication bias. This is expected, as the majority of studies did not report hyperacusis prevalence as a primary outcome.

Supplemental Figure S3.

Meta-analytic scatter plot depicting the curvilinear relationship between mean age and current hyperacusis prevalence estimates in the full sample of subjective studies.



Note. Plotted points are proportional to the sample size of each study. Although there are few studies with mean ages greater than 15, examination of the relationship between age and prevalence across the full sample allows us to extrapolate findings from the ADI-R studies (age range 2–14 years; see Figure 3 in main text). The fitted values of a weighted quadratic meta-regression indicate that the prevalence of hyperacusis in ASD may decrease with age as individuals enter adulthood. However, this conclusion is preliminary, and additional studies are required to further investigate the developmental trajectory of hyperacusis in adolescents and adults with ASD.

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