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BRIEF REPORT

Brief Report: Prevalence and Severity of Auditory Sensory Over-Responsivity in Autism as Reported by Parents and Caregivers

Tana B. Carson[1] · Matthew J. Valente[2] · Bradley J. Wilkes[3] · Lynne Richard[1]

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Abstract

Auditory sensory over-responsivity (aSOR) is a frequently reported sensory feature of autism spectrum disorders (ASD); however, there is little consensus regarding its prevalence and severity. This cross-sectional study uses secondary data from the Autism Diagnostic Interview-Revised (ADI-R; Item 72: *undue sensitivity to noise*) housed in the US National Institute of Mental Health Data Archives to identify prevalence and severity of aSOR. Of the 4104 subjects with ASD ages 2–54 ($M = 9$, SD = 5.8) who responded to item 72, 60.1% ($n = 1876$) had aSOR currently (i.e., point prevalence) and 71.1% ($n = 2221$) reported having aSOR ever (i.e., lifetime prevalence). aSOR prevalence and severity were affected by age, but there were no associations with sex.

Keywords Autism · Auditory · Sensory · Prevalence · Severity

Introduction

Although individuals with autism spectrum disorders (ASD) have been shown to have normal hearing (Gravel et al., 2006; Tharpe et al., 2006), auditory sensory over-responsivity (aSOR) is a commonly reported sensory feature of ASD (Gomes et al., 2008; Tomchek & Dunn, 2007). However, there is no consensus on the prevalence and severity of this problem within the ASD population. aSOR in ASD ranges widely across studies from 15 to 100% (Green et al., 2015; Khalfa et al., 2004; Schoen, 2009; Stefanelli et al., 2020; Tomchek & Dunn, 2007) whereas the point prevalence in pediatric population ranges from 3.2 to 17.1% (Coelho et al., 2007; Pienkowski et al., 2014; Rosing et al., 2016).

aSOR (hyper-reactivity, hyper-responsivity, sensory over-responsivity, and hyperacusis) is characterized by discomfort and irritability in response to auditory stimuli (Rosenhall et al., 1999; Stefanelli et al., 2020) and involves aversive responses to everyday sounds that do not bother others. aSOR differs from hyper-sensitivity which involves having a decreased threshold for detecting auditory stimuli. Although aSOR has been shown to differ in children with ASD compared to typically developing controls, hyper-sensitivity does not (Gravel et al., 2006; Rosenhall et al., 1999).

Across the lifespan for those with ASD, anxiety and over-responsivity to different types of sensory stimuli have been closely linked (Gillott & Standen, 2007; Green & Ben-Sasson, 2010; Mazurek et al., 2012; Syu & Lin, 2018). In fact, sensory over-responsivity has been shown to be a positive predictor of anxiety later in development for those with ASD (Green et al., 2012). For children with ASD, sensory over-responsivity has been linked to both child stress (Rance et al., 2017) and increased family stress (Kirby et al., 2015; Reynolds & Lane, 2008; Schaaf et al., 2011). There are currently no standardized assessments focused solely on evaluating aSOR in children, nor are there evidence-based treatments available for aSOR in ASD.

The purpose of this brief report is to utilize a large dataset to identify the prevalence of aSOR in ASD including both: (a) point prevalence, the proportion of individuals with ASD who have aSOR at the time of assessment, and (b) lifetime prevalence, the proportion of individuals who have ever had aSOR, even if they no longer have it at the time of assessment. Additionally, this study aims to evaluate age and sex

✉ Tana B. Carson
tcarson@fiu.edu

[1] Department of Occupational Therapy, Florida International University, 11200 SW 8th St., AHC3, Miami, FL 33199, USA

[2] Department of Psychology, Center for Children and Families, Florida International University, Miami, FL, USA

[3] Department of Applied Physiology and Kinesiology, University of Florida, Gainesville, FL, USA

effects on point prevalence and severity of aSOR among people with any form of ASD. Based on previous studies of age-related differences in anxiety (Llanes et al., 2020) and aSOR (Kern et al., 2006) among children with ASD, we hypothesize that the point prevalence and severity ratings of aSOR (e.g., mild to severe) would be greater in younger age groups compared to older age groups. Sex differences in ASD have been reported in core diagnostic criteria (Van Wijngaarden-Cremers et al., 2014), as well as in physiological domains such as brain morphology and function (Irimia et al., 2017, 2018; Zhang et al., 2018). Importantly, sex differences in ASD have also been identified in sensory hyper-reactivity behaviors (Tavassoli et al., 2014) and in underlying neural networks related to sensory processing (Cummings et al., 2020). Therefore, this study also aims to identify whether aSOR differentially occurs in males and females with ASD. Based on previous studies, it is anticipated that if sex-differences exist, females would have higher rates of aSOR than males. This information can be useful for developing targeted assessments and interventions.

Methods

Secondary Data Analysis

Secondary data (i.e., data previously collected by other studies) were extracted from the US National Institute of Mental Health Data Archives (NDA) by entering "autism diagnostic interview-revised" (ADI-R) search terms in the Data Structure (i.e., questionnaire) search feature of the Data Dictionary. Subjects were included if they had the following data elements (i.e., questionnaire items) as follows: (1) ADI-R item number 72; (2) all four ADI-R items required for calculating cut-off scores for ASD vs. non-ASD [(i) total for Section A (dbaes_atotal), (ii) total for Section B—nonverbal (dbaes_bnvtotal), (iii) Total for Section B—verbal (dbaes_bvtotal) and (iv) Total for Section C (dbaes_ctotal)], and (3) "interview_age" of two years and up. All other subjects were excluded. See Fig. 1 for a flowchart detailing inclusion and exclusion decisions and resulting sub-samples.

Study Sample

The original data package included $n = 7001$ subjects from 98 studies. After applying exclusion criteria, the remaining $n = 4104$ subjects with ASD and $n = 1492$ without ASD ages two and up were included in the study (Fig. 1). Data Elements "interview_age" and "sex" were used to generate descriptive statistics on demographics for the study sample. Race and ethnicity data were only available for a small subset of subjects (~ 10) therefore no information is included on these variables. Four age-groups were created following guidelines of the US NICHD guidelines previously reported for pediatric research (Williams et al., 2012) as follows: early childhood (2 to < 6 years), middle childhood (6 to < 12 years), early adolescent (12 to < 19 years), and adulthood (19–54.3 years).

Instrument

Autism Diagnostic Interviewer—Revised (ADI-R)

The Autism Diagnostic Interviewer—R (ADI-R) (Lord et al., 1994) was utilized to confirm/deny ASD diagnoses and to identify aSOR prevalence and severity via instrument item 72: *undue sensitivity to noise*. This item of the ADI-R was selected over items from other sensory processing measures available in the NDA because it is a standardized clinical assessment of autism that includes a detailed question regarding aSOR and is widely used in ASD research. At the time of this study, the ADI-R provided the largest sample available that included both gold standard ASD diagnostic data as well as a detailed instrument item regarding aSOR. See supplemental material (Appendix A) for a summary table of all aSOR items housed in the NDA from sensory questionnaires.

Auditory Sensory Over-Responsivity (aSOR)

One item of the ADI-R (No. 72) asks about atypical, hyper-reactive responses to everyday sounds. Although the item is referred to as "undue general sensitivity to noise", the description and coding is focused on aSOR behaviors rather than decreased thresholds for hearing sounds (i.e., sensitivity). Responses to this item are coded on a 4-point Likert scale from 0 to 3 (0 representing no evidence of aSOR and 1–3 representing evidence of aSOR ranging from mild to severe; Table 1). In order to receive a code of 1–3 on this item, parents/caregivers must report more than one instance of increased reactivity to "everyday sounds, such as household appliances or traffic, rather than a reaction to sudden, hard, or unexpected noise such as thunder or a loudspeaker". Additionally, to qualify for a rating of 1–3, this type of increased reactivity must occur for 3 months or more. Information was gathered regarding the individual's response to sounds currently as well as ever. For the purpose of this study, the current ratings were used to identify point prevalence, whereas the ever ratings were used to estimate lifetime prevalence.

Logistic Regression Analyses

To identify the prevalence in our sample, *undue sensitivity to noise (current)* was treated as a binary variable (0 = no evidence of hyper-reactivity and 1 = evidence of

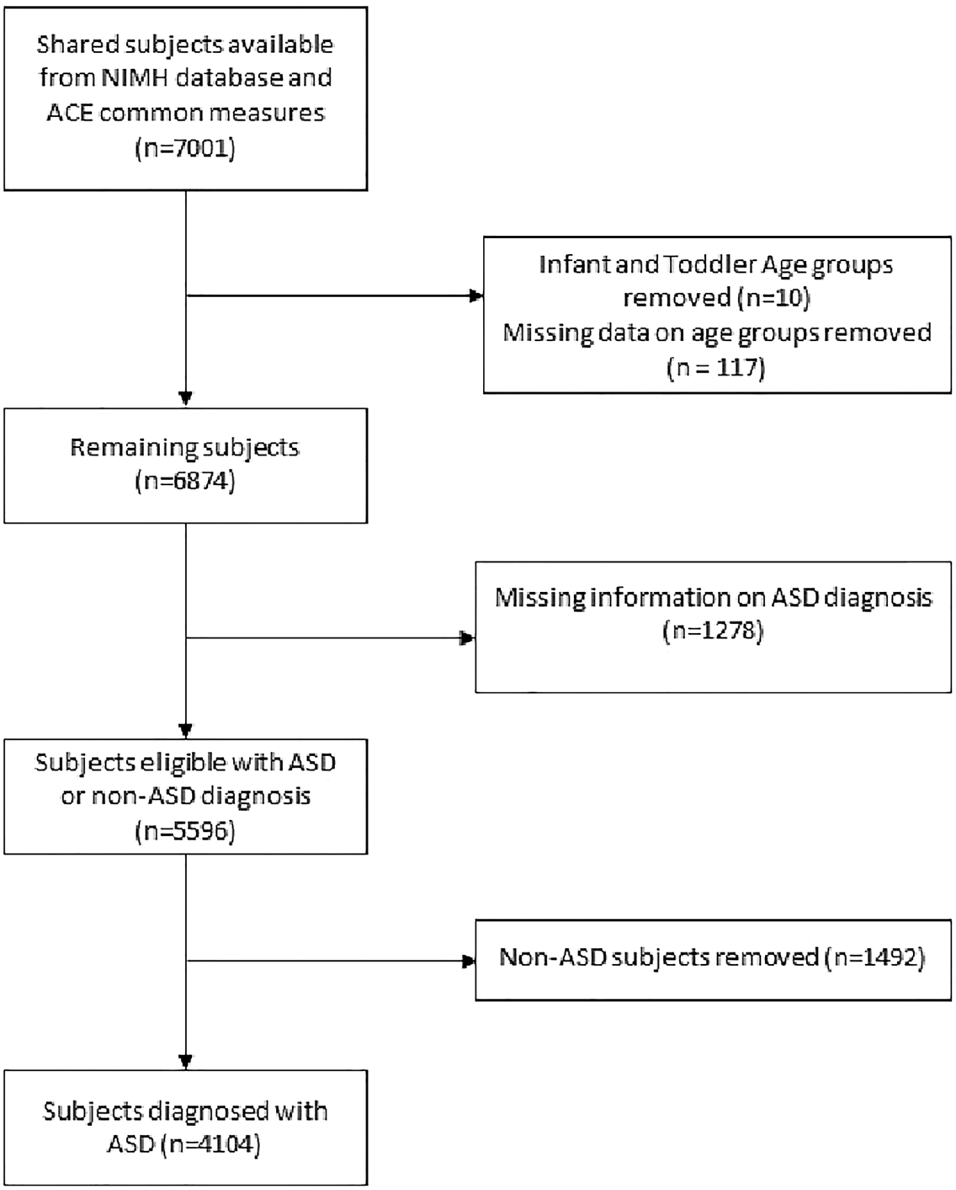

Fig. 1 Flowchart of subject inclusion and exclusion from the study sample. This table represents the decisions that were made to establish the ASD ($n = 4104$) and non-ASD ($n = 1492$) sub-samples

Table 1 ADI-R code, description, and study code for undue general sensitivity to noise

ADI-R code and description	Study code
0 = No general sensitivity to noise	Normal
1 = Slight sensitivity to noise: somewhat sensitive to loud sounds such as the vacuum cleaner, motorbikes or other appliances	Mild
2 = Definite sensitivity to noises that are not distressing to most other people, the sensitivity being accompanied by a clear behavioral change (such as avoidance, hands over ears, or crying)	Moderate
3 = Definite sensitivity to noises to the extent that subject's distress/disturbance in relation to certain noises interferes with family or household routines	Severe
9 = N/K (not known) or not asked	Missing

hyper-reactivity). To investigate point prevalence and severity across age groups, logistic and multinomial logistic regression analyses were conducted using only the undue sensitivity to noise (current) item (Long, 1997). To investigate questions of severity between age groups, ordinal response categories: 0 = normal, 1 = mild, 2 = moderate, and

3 = severe were used. The reference category was zero for all logistic regression analyses. All analyses were completed using PROC LOGISTIC in SAS 9.4.

Results

Demographics

Of the 7001 shared subjects available with ADI-R data, 5596 had complete data on ADI-R sections A–C which were used to diagnosis ASD and, of those, 73.3% ($n = 4104$) met the cut-off for an ASD diagnosis (Fig. 1). Of the 98 collections, 85 collections contained data on individuals that were diagnosed with ASD and that were in age groups > 2. Of these 85 collections, the average completion rate for #72 was 73.89% and the median completion rate was 100%. The ASD sample was relatively young with an average interview age of 9.03, years (SD = 5.80). Most of the sample fell within two youngest age categories: early (2 to < 6 years; 37.6%) and middle childhood (6 to < 12 years; 38.6%; Table 2).

Point Prevalence of aSOR in ASD

Of the 4,104 subjects with ASD, 76% ($n = 3118$) had data on the *undue general sensitivity to noise (current)* item. In the ASD sample, 39.8% ($n = 1242$) received a "normal" caregiver-reported rating (i.e., 0), while 60.2% ($n = 1876$) were rated as having aSOR (ranging from mild to severe (i.e., ratings of 1–3; Table 3; Fig. 2). As expected, higher point prevalence rates were observed in those with ASD compared to those without ASD.

Lifetime Prevalence of aSOR in ASD

Of the 4,104 subjects with ASD, 76% ($n = 3125$) had data on the *undue general sensitivity to noise (ever)* item. In the ASD sample, 28.9% ($n = 904$) received a "normal" caregiver-reported rating (i.e., 0), while 71.1% ($n = 2221$) were rated as having aSOR (ranging from mild to severe (i.e., ratings of 1–3; Table 3; Fig. 2). As expected, higher lifetime prevalence rates were also observed in those with ASD compared to those without ASD.

Table 2 Demographics

	n	%	M (SD)	Min–max	"Current" responses	"Ever" responses
ASD						
Sex						
Female	754	18.4%			605	606
Male	3350	81.6%			2513	2519
Interview age	4104		9 (5.8)	2–54.3		
Age groups with interview data						
Early childhood: 2 to < 6 years	1544	37.6%	4.1 (1.0)	2–5.9	1421	1419
Middle childhood: 6 to < 12 years	1584	38.6%	8.8 (1.7)	6–11.9	1133	1136
Early adolescent: 12 to < 19 years	743	18.1%	14.5 (1.8)	12–18.9	426	431
Adult: 19 and up	233	5.7%	25.3 (6.0)	19–54.3	138	139
Non-verbal subjects	1069	26%			958	958
Verbal subjects	3035	74%			2160	2167
Non-ASD						
Sex						
Female	413	27.7%			348	339
Male	1033	69.24%			806	782
Interview age	1492		8.1 (6.4)	2–53.5		
Age groups with interview data	1492					
Early childhood: 2 to < 6 years	751	50.3%	3.9 (0.9)	2–5.9	680	678
Middle childhood: 6 to < 12 years	470	31.5%	8.7 (1.8)	6–11.9	352	319
Early adolescent: 12 to < 19 years	180	12.1%	14.4 (1.9)	12–18.9	106	108
Adult: 19 and up	91	6.1%	26.9 (7.3)	19.2–53.5	54	54
Non-verbal subjects	181	12.1%			141	135
Verbal subjects	1174	78.7%			929	902

n is the number of subjects with data on the respective demographic variable. Percentages of demographic variables are computed using the total number of subjects in the ASD group (4104) and the non-ASD group (1492), respectively. In the case the percentages do not add up to 100%, the remaining percentage is due to missing data on the demographic variable

Table 3 Number and percent of subjects that endorsed each severity category of undue general sensitivity to noise–current (point prevalence) and –ever (lifetime prevalence) in both groups (ASD and non-ASD)

	ASD	Non-ASD
Undue general sensitivity to noise–current		
Normal	1242 (39.8)	679 (57.0)
aSOR (at any level)	1876 (60.2)	513 (43.0)
Mild	587 (18.8)	197 (16.5)
Moderate	1090 (35.0)	271 (22.7)
Severe	199 (6.4)	45 (3.8)
Total	3118 (100.0)	1192 (100.0)
Undue general sensitivity to noise–ever		
Normal	904 (28.9)	592 (39.7)
aSOR (at any level)	2221 (71.1)	567 (48.9)
Mild	397 (12.7)	156 (13.5)
Moderate	1374 (44.0)	329 (28.3)
Severe	450 (14.4)	82 (7.1)
Total	3125 (100.0)	1159 (100.0)

There were 986 observations missing for the ASD group on undue general sensitivity to noise–current, and 889 observations for the ASD group on undue general sensitivity to noise–ever. There were 300 observations missing for the non-ASD group on undue general sensitivity to noise–current, and 333 observations for the ASD group on undue general sensitivity to noise–ever. The prevalence of any level of aSOR—current was almost twice as prevalent in the ASD group compared to the non-ASD group in an analysis conditioned on sex and age group (OR = 1.99, 95% CI [1.73, 2.29]. The prevalence of any level of aSOR—ever was over twice as prevalent in the ASD group compared to the non-ASD group in an analysis conditioned on sex and age group (OR = 2.48, 95% CI [2.15, 2.86]

Prevalence and Severity of aSOR Between Age Groups

The predictors of interest were age, sex, and the interaction between sex and age group. Three dummy-codes were created to compare the odds of aSOR prevalence and aSOR severity across the four age groups. Only the subjects that were diagnosed with ASD were included in these analyses. After listwise deletion there were 3118 observations. There were no significant interactions of sex with age groups on the point prevalence or severity of aSOR, therefore these interaction terms were dropped from the model. Effects that were significant at alpha = 0.05 were reported below but odds ratios (OR), 95% confidence intervals, and *p*-values were reported for all predictors for prevalence and severity of aSOR in Table 4.

Predictors of aSOR Prevalence

There was no difference in the prevalence of aSOR across males and females. Subjects in middle childhood had higher odds of caregivers reporting aSOR (OR = 1.676, 95% CI [1.426, 1.970]) than those in early childhood. Subjects in early adolescence had higher odds of caregivers reporting aSOR (OR = 1.323, 95% CI [1.060, 1.650]) than those in early childhood. Subjects in early adolescence had lower odds of caregivers reporting aSOR than those in middle childhood (OR = 0.789, 95% CI [0.626, 0.994]) (see Table 4 for all age groups, 95% confidence intervals, and *p*-values).

Predictors of aSOR Severity

There was no difference in the severity of aSOR across males and females. Subjects in the middle childhood group had higher odds of caregivers reporting mild aSOR (OR = 1.487, 95% CI [1.185, 1.867]), moderate aSOR (OR = 1.815, 95% CI [1.515, 2.175]), and severe aSOR (OR = 1.468, 95% CI [1.056, 2.041]), compared to subjects in early childhood. Subjects in early adolescence and in adulthood had significantly higher odds of caregivers reporting mild aSOR (OR = 1.955, 95% CI [1.476, 2.589]; OR = 1.885, 95% CI [1.203, 2.953], respectively) compared to subjects in early childhood. Subjects in early adolescence and in adulthood had lower odds of caregivers reporting moderate aSOR (OR = 0.570, 95% CI [0.434, 0.748]; OR = 0.641, 95% CI [0.420, 0.980], respectively) compared to subjects in middle childhood (see Table 4 for all age groups, 95% confidence intervals, and *p*-values).

Discussion

In this cross-sectional study, for individuals with ASD whose caregivers answered item 72 of the ADI-R, 60.2% ($n = 1876$) of individuals with ASD had some level of aSOR (mild to severe) at the time of assessment (i.e., point prevalence) and 71.1% ($n = 2221$) were reported to have had aSOR at some point in life (i.e., lifetime prevalence). For both point prevalence and lifetime prevalence, higher frequencies were observed in moderate cases than mild or severe cases. According to these data, the frequency of caregiver-reported aSOR peaks in middle childhood. These findings narrow the wide range of occurrence rates (15–100%) previously reported in the literature (Green et al., 2015; Khalfa et al., 2004; Schoen, 2009; Stefanelli et al., 2020; Tomchek & Dunn, 2007).

There were no effects of sex on either prevalence or severity of aSOR in ASD. This finding differs from neurophysiology indicating sex-differences in salience networks associated with SOR (Cummings et al., 2020) and other prominent behavioral features of ASD such as restricted repetitive behaviors that have been shown to differ in quality and quantity for females with ASD (McFayden et al., 2019). Both prevalence and severity of aSOR (*current* item ratings) did vary significantly as a function of age, however. Most

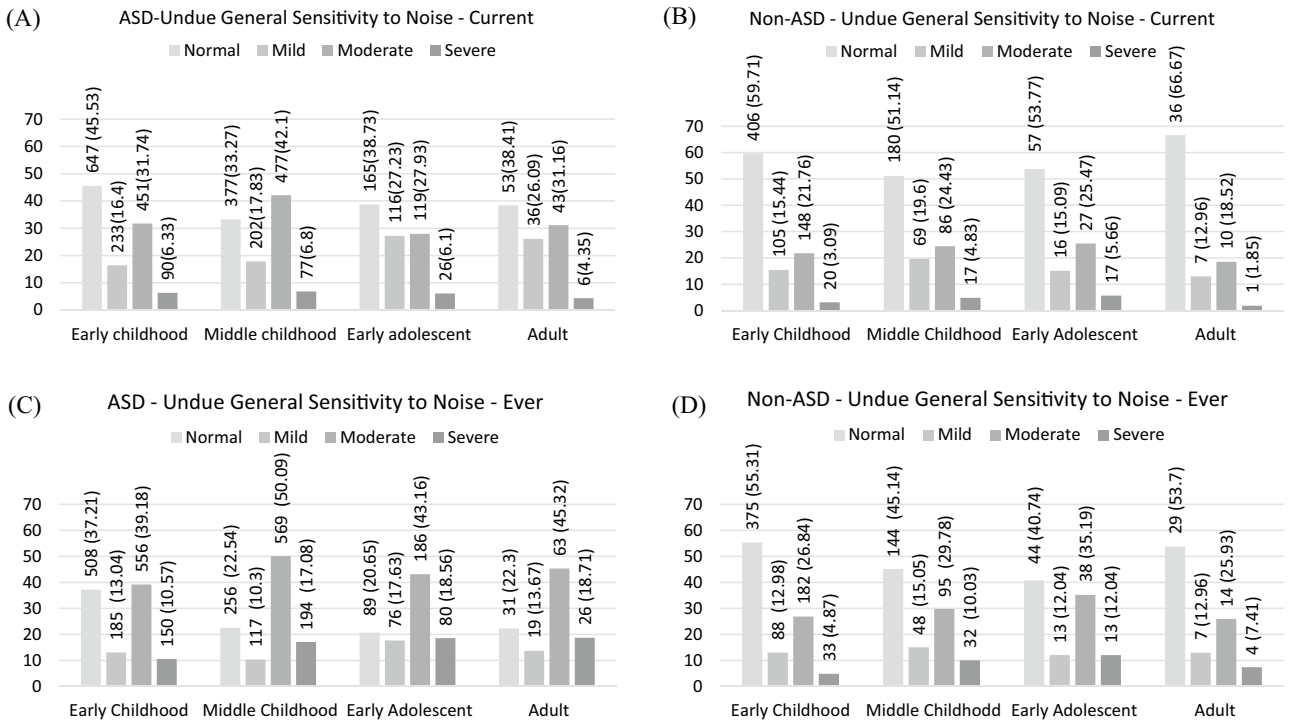

Fig. 2 Undue general sensitivity to noise–current and ever frequencey and percentages within each age group for ASD and non-ASD subjects. **a** Frequency and percentage of undue general sensitivity to noise–current for ASD. **b** Frequency and percentage of undue general sensitivity to noise–current for non-ASD. **c** Frequency and percentage of undue general sensitivity to noise–ever for ASD. **d** Frequency and percentage of undue general sensitivity to noise–ever for non-ASD. Percentages in parentheses represent percentage of the corresponding aSOR category within age group for those with complete data on the undue general sensitivity to noise–current item (ASD; $n=3118$, non-ASD; 1192) and undue general sensitivty to noise–ever item (ASD; $n=3125$, non-ASD; 1159)

Table 4 Age grouping as a predictors of aSOR prevalence and severity

	Odds endorsing aSOR versus normal	Odds endorsing mild versus normal	Odds endorsing moderate versus normal	Odds endorsing severe versus normal
Sex (male = 0; female = 1)	OR = 1.016, 95% CI [0.846, 1.219], $p < 0.8668$	OR = 1.063, 95% CI [0.827, 1.367], $p < 0.6341$	OR = 0.989, 95% CI [0.804, 1.216], $p < 0.9255$	OR = 1.029, 95% CI [0.703, 1.506], $p < 0.8831$
Comparison group = early childhood				
Middle childhood	OR = 1.676*, 95% CI [1.426, 1.970], $p < 0.001$	OR = 1.487*, 95% CI [1.185, 1.867], $p = 0.0006$	OR = 1.815*, 95% CI [1.515, 2.175], $p < 0.0001$	OR = 1.468*, 95% CI [1.056, 2.041], $p = 0.0224$
Early adolescent	OR = 1.323*, 95% CI [1.060, 1.650], $p = 0.0132$	OR = 1.955*, 95% CI [1.476, 2.589], $p < 0.0001$	OR = 1.034, 95% CI [0.794, 1.348], $p = 0.8024$	OR = 1.134, 95% CI [0.709, 1.811], $p = 0.6001$
Adulthood	OR = 1.340, 95% CI [0.936, 1.918], $p = 0.1093$	OR = 1.885*, 95% CI [1.203, 2.953], $p = 0.0057$	OR = 1.164, 95% CI [0.765, 1.771], $p = 0.4782$	OR = 0.814, 95% CI [0.340, 1.947], $p = 0.6431$
Comparison group = middle childhood				
Early adolescent	OR = 0.789*, 95% CI [0.626, 0.994], $p = 0.0444$	OR = 1.314, 95% CI [0.981, 1.761], $p = 0.0672$	OR = 0.570*, 95% CI [0.434, 0.748], $p < 0.0001$	OR = 0.772, 95% CI [0.477, 1.249], $p = 0.2919$
Adulthood	OR = 0.800, 95% CI [0.555, 1.152], $p = 0.2295$	OR = 1.267, 95% CI [0.803, 2.000], $p = 0.3093$	OR = 0.641*, 95% CI [0.420, 0.980], $p = 0.0040$	OR = 0.554, 95% CI [0.230, 1.335], $p = 0.1882$
Comparison group = early adolescent				
Adulthood	OR = 1.013, 95% CI [0.683, 1.504], $p = 0.9476$	OR = 0.964, 95% CI [0.593, 1.567], $p = 0.8827$	OR = 1.125, 95% CI [0.706, 1.794], $p = 0.6195$	OR = 0.718, 95% CI [0.280, 1.838], $p = 0.4893$

*Indicates statistically significant effect with $p < 0.05$

notably an increase was observed in both prevalence and severity during middle childhood when compared to early childhood. In fact, greater severity of aSOR was reported at every level (mild, moderate, and severe) in middle childhood. This shift in prevalence at middle childhood is not surprising considering similar shifts noted in anxiety and ADHD symptoms in ASD also observed during this developmental time period (Llanes et al., 2020). Lower aSOR severity in older age groups supports previous cross-sectional findings using the Sensory Profile (Dunn, 1999) with individuals with ASD ages 3–56 years (Kern et al., 2006). Although differences between age groups were observed, these data are not measured on the same individuals over the lifespan. Therefore, additional longitudinal observations are warranted to confirm whether a development process contributes to these differences in aSOR across development. The current study provides a cross-sectional observation of differences in the severity of aSOR across age groups.

Clinical Implications and Future Directions

Further research is needed to identify a critical time period for developing aSOR. Although specific aSOR assessments are lacking, the present study provides preliminary support for using the "undue sensitivity to noise" and "age of onset" items of the ADI-R toddler instrument to screen for aSOR in early childhood. It is evident from our results that aSOR may begin early in development, but that atypical hyperreactivity behaviors are more apparent at the transition from early childhood to school-age. The data presented in the current study at a minimum, support the importance of early screening and identification of aSOR at the start of school age in Kindergarten (age 6).

At present, there are no standardized, valid clinical measures of aSOR specifically. The findings of the present study reflect that aSOR is a significant problem affecting over half of those with ASD in the NDA whose caregivers answered item 72 of ADI-R. It should be noted that 24% of available subjects were missing data on this item and it is unclear how those subjects' data would have impacted prevalence rates. For example, in the ASD group, if we assume all missing data on item 72 (current) were normal, then the point prevalence of aSOR would be lower (45.7% aSOR; 54.2% normal); whereas, if all missing data had aSOR, then point prevalence would be higher (69.7% aSOR; 30.3% normal). Similarly, if we assume all missing data on item 72 (ever) were normal, then the lifetime prevalence of aSOR would be lower (54.1% aSOR; 45.9% normal); whereas, if we assume all missing data indicated aSOR, then lifetime prevalence would be higher (76.1% aSOR; 23.9% normal). Although these findings cannot be generalized beyond the NDA data available, they do support the need for developing a specific assessment of aSOR. Further research should be conducted to evaluate the presentation of aSOR across development, paying special attention to the transition between early and middle childhood.

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Declarations

Conflict of interest The authors have no relevant financial or non-financial interest to disclose.

Ethical Approval This is a secondary data analysis study and determined non-human subjects' research. The FIU Institutional Review Board has confirmed that no ethical approval is necessary.

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