

## Research Article

# Listening Difficulties in Children With Fetal Alcohol Spectrum Disorders: More Than a Problem of Audibility

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**Purpose:** Data from standardized caregiver questionnaires indicate that children with fetal alcohol spectrum disorders (FASDs) frequently exhibit atypical auditory behaviors, including reduced responsivity to spoken stimuli. Another body of evidence suggests that prenatal alcohol exposure may result in auditory dysfunction involving loss of audibility (i.e., hearing loss) and/or impaired processing of clearly audible, “suprathreshold” sounds necessary for sound-in-noise listening. Yet, the nexus between atypical auditory behavior and underlying auditory dysfunction in children with FASDs remains largely unexplored.

**Method:** To investigate atypical auditory behaviors in FASDs and explore their potential physiological bases, we examined clinical data from 325 children diagnosed with FASDs at the University of Washington using the FASD 4-Digit Diagnostic Code. Atypical behaviors reported on the “auditory filtering” domain of the Short Sensory Profile were assessed to document their prevalence across FASD diagnoses and explore their relationship to reported hearing loss and/or central nervous system measures of cognition, attention, and

language function that may indicate suprathreshold processing deficits.

**Results:** Atypical auditory behavior was reported among 80% of children with FASDs, a prevalence that did not vary by FASD diagnostic severity or hearing status but was positively correlated with attention-deficit/hyperactivity disorder. In contrast, hearing loss was documented in the clinical records of 40% of children with fetal alcohol syndrome (FAS; a diagnosis on the fetal alcohol spectrum characterized by central nervous system dysfunction, facial dysmorphism, and growth deficiency), 16-fold more prevalent than for those with less severe FASDs (2.4%). Reported hearing loss was significantly associated with physical features characteristic of FAS.

**Conclusion:** Children with FAS but not other FASDs may be at a particular risk for hearing loss. However, listening difficulties in the absence of hearing loss—presumably related to suprathreshold processing deficits—are prevalent across the entire fetal alcohol spectrum. The nature and impact of both listening difficulties and hearing loss in FASDs warrant further investigation.

**F**etal alcohol spectrum disorder (FASD) is an umbrella term used to describe the range of physical, functional, and neurological abnormalities caused by prenatal alcohol exposure. It has been estimated that

FASDs affect at least 1% of children and perhaps more (Sampson et al., 1997), with prevalence estimates varying by geographic location and diagnostic methodology (Roozen et al., 2016). Among all FASDs, fetal alcohol syndrome (FAS) is the most visibly evident diagnosis, characterized by growth deficiency and a specific set of facial features (i.e., thin upper lip, smooth philtrum, and small eyes), in the presence of significant central nervous system (CNS) abnormality (Astley, 2004, 2013; Bertrand et al., 2005). However, the majority of people with an FASD lack these observable physical markers yet still experience a wide and variable range of functional impairments. These typically include learning, attention, impulse control, communication, and social skill deficits (Astley, 2004; Kodituwakku & Kodituwakku, 2014; Streissguth et al., 2004; Thorne, 2017).

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A small body of classic literature suggests that the teratogenic effects of prenatal alcohol exposure may involve insults to the auditory system potentially resulting in hearing loss (i.e., decreased sensitivity to low-level sound) and/or “suprathreshold” deficits affecting the processing of clearly audible sound essential for picking out sound targets in noise. A complex hierarchy of suprathreshold processing operations originating in the CNS and the auditory periphery is involved in separating out and making sense of the myriad acoustic signals generated by competing sound sources. Key mechanisms include the extraction of spatial information and precise encoding of the spectrotemporal features unique to each sound source, perceptual separation of sound streams and formation of distinct “auditory objects,” and selective attention to the target auditory object (reviewed in Bizley & Cohen, 2013; Carlyon, 2004; Griffiths & Warren, 2004; B. C. J. Moore & Gockel, 2012; Shinn-Cunningham, 2008, 2017; Shinn-Cunningham, Best, & Lee, 2017). Suprathreshold processing deficits can arise, for example, as a consequence of inadequate sensory encoding (e.g., broad frequency tuning on the basilar membrane [Kortlang, Mauermann, & Ewert, 2016] or temporal coding infidelities [Bharadwaj, Masud, Mehraei, Verhulst, & Shinn-Cunningham, 2015]), binaural/spatial processing deficits (Ross, Fujioka, Tremblay, & Picton, 2007), or impaired cognitive control processes (e.g., attention deployment) in the absence of auditory coding deficits (Lee, Larson, Maddox, & Shinn-Cunningham, 2014). Damage to these or other suprathreshold processes can compromise sound-in-noise listening abilities and—like hearing loss—can impact daily function (Hillock-Dunn, Taylor, Buss, & Leibold, 2015; Shinn-Cunningham, 2017). Such auditory dysfunction may contribute significantly to some of the communication impairments observed in FASDs. In the complex acoustic environments common to daily life (e.g., classrooms, restaurants), both hearing loss and suprathreshold listening deficits in the absence of hearing loss have the capacity to negatively impact speech perception (reviewed in Bronkhorst, 2015). Such degradation of the speech signal may limit the auditory information children rely on to decipher and learn fundamental linguistic rules (White-Schwoch et al., 2015).

In terms of processes associated with audibility, impairments in mechanisms and structures involved in detecting low-level sound have been observed in individuals with FAS and in laboratory animals with extensive prenatal alcohol exposure. Prenatally exposed rats were found to have significantly prolonged and/or reduced-amplitude auditory brainstem responses and damaged hair cell receptors indicative of sensorineural (inner ear/auditory nerve) hearing loss (Church et al., 1987; Church & Kaltenbach, 1997). Consistent with this, hearing loss of both sensorineural and conductive (middle ear) origin has been shown to be more prevalent in children with FAS than their typically developing counterparts (Church, Eldis, Blakley, & Bawle, 1997; Church & Gerkin, 1988; Rössig, Wässer, & Oppermann, 1994). Conductive hearing loss related to recurrent otitis media has been commonly observed in children with craniofacial anomalies, including those with FAS, at rates ranging

from 38% to 93% (reviewed in Church & Kaltenbach, 1997). Importantly, however, there is little research exploring sensorineural or conductive hearing loss in individuals who have FASDs but do not have the sentinel physical findings (i.e., facial dysmorphism and/or growth deficiency) of FAS. Cohen-Kerem, Bar-Oz, Nulman, Papaioannou, and Koren (2007) observed hearing loss and otitis media to be no more prevalent in those with diagnoses across the fetal alcohol spectrum than is found in the general population, but confirmatory data are lacking. This potential divergence in prevalence of hearing loss in FAS versus other FASDs may be due to an embryonic neuroectodermal syndrome hypothesized to underlie the craniofacial and ocular anomalies seen primarily in FAS; inner ear dysfunction may be associated with this syndrome (Church & Kaltenbach, 1997).

Research investigating the suprathreshold processing sequelae of prenatal alcohol exposure is even more limited and again has been restricted primarily to human subjects with FAS or laboratory animals exposed prenatally to moderate-to-heavy levels of alcohol. Slowed transmission of neural impulses in the auditory brainstem (Church, Abel, Kaltenbach, & Overbeck, 1996), delayed auditory event-related potentials (Kaneko, Riley, & Ehlers, 1993), and a reduction in the size of key auditory brainstem structures (Church & Kaltenbach, 1997) have been seen in prenatally exposed rats. Similarly, attenuated auditory event-related potentials have been observed in children with FAS (Kaneko, Ehlers, Phillips, & Riley, 1996; reviewed in Church & Kaltenbach, 1997). Data regarding the functional consequences of suprathreshold deficits due to prenatal alcohol exposure are particularly sparse. In one of the sole behavioral studies to date, Church et al. (1997) observed listening deficits in 100% of children with FAS ( $n = 12$ ) tested on sound-in-noise dichotic listening tasks; however, most of these individuals had comorbid hearing loss, rendering it difficult to disentangle the relationship between functional impairment, hearing loss, and suprathreshold deficits. Moreover, as with hearing loss, little is known about the prevalence of listening difficulties (defined by D. R. Moore, 2018, as problems with hearing or listening despite normal audiometry) or the potential for suprathreshold processing deficits in individuals with FASDs but without FAS (cf. Stephen et al., 2012).

To date, there has been no systematic investigation of auditory dysfunction across the fetal alcohol spectrum. The limited data available suggest that individuals who have FASDs but not FAS may be less likely to experience hearing loss due to the absence of structural impairments associated with an embryologic neuroectodermal syndrome. We therefore predict that atypical auditory behaviors on the Short Sensory Profile (SSP; McIntosh, Miller, Shyu, & Dunn, 1999)—a norm-referenced caregiver report questionnaire used clinically to assess sensory processing dysfunction—will be present in children with FASDs and will be unrelated to hearing loss. It is not obvious, a priori, whether such listening difficulties—which presumably derive chiefly from suprathreshold deficits associated with particular CNS processes that may or may not be coupled to those measures

of CNS dysfunction used to inform FASD diagnoses—will vary according to diagnostic severity.

To investigate this hypothesis and to more generally explore the prevalence and nature of auditory dysfunction across FASDs, we leveraged existing information about auditory-related behaviors and reported hearing status, in addition to other diagnostic and neurobehavioral variables (including standardized measures of cognition/IQ, attention, and language function) present in a clinical database collected by the Washington State Fetal Alcohol Syndrome Diagnostic & Prevention Network (FAS DPN). Information about peripheral hearing status was obtained from caregiver-provided records and reports submitted to the database. Data regarding auditory behaviors were gleaned from caregiver ratings of child auditory function on the SSP, which assesses multiple “domains” of sensory processing. In this study, we focused on the “auditory filtering” domain, which consists primarily of five items on which caregivers are asked to rate their child’s behavior in situations that require either attending to or tuning out sound stimuli (see Jirikowic, Thorne, McLaughlin, Lee, & Astley, 2018, for full analysis and discussion of results in this sample population across all SSP domains). The SSP items were formulated based on neurobehavioral theories of sensory processing and integration (Dunn, 1997) and therefore lack reference to the highly specified auditory processing theories of auditory science. However, the auditory filtering items nonetheless serve to provide a standardized clinical measure of higher order listening behaviors, with a particular focus on attentional processes. The central role of attention in the behaviors probed by these items has been confirmed by principal component analyses showing that ratings on the auditory filtering items (plus one other item from an earlier long-version Sensory Profile) cluster together into a discrete “inattention/distractibility” factor (Dunn & Brown, 1997).

Despite prior reports that auditory filtering is consistently one of the SSP domains on which children with FASDs are most often rated as demonstrating a “definite difference” ( $> 2$  SDs below the normative mean; Abele-Webster, Magill-Evans, & Pei, 2012; Carr, Agnihotri, & Keightley, 2010; Franklin, Deitz, Jirikowic, & Astley, 2008; Hansen & Jirikowic, 2013; Jirikowic, Olson, & Kartin, 2008), there has been little discussion of the potential impairments that may underlie atypical auditory behaviors in children with FASDs (Abele-Webster et al., 2012). This study thus sought to investigate auditory dysfunction in FASDs by exploring auditory behavioral outcomes and their relationship to neurobehavioral measures and reported hearing loss as a means to generate preliminary hypotheses about the underlying mechanisms involved. The study had three specific aims:

- Specific Aim 1: to quantify the prevalence of atypical auditory behaviors—as measured by auditory filtering domain scores on the SSP—across the spectrum of fetal alcohol disorders. We hypothesized that the proportion of children rated with a definite difference in auditory filtering scores would be high across the fetal alcohol spectrum.
- Specific Aim 2: to explore whether atypical auditory filtering domain scores for children with FASDs are related to (a) reported hearing loss and/or (b) measures of cognition/IQ, attention, and language function indicative of CNS impairment. Using neurodevelopmental CNS impairment as a proxy for potential suprathreshold processing deficits, we hypothesized that atypical auditory filtering scores in children with FASDs would be related more strongly to these deficits than to reported hearing loss.
- Specific Aim 3: to quantify the prevalence of hearing loss across the spectrum of fetal alcohol disorders. We hypothesized that children with FAS would have a higher prevalence of reported hearing loss than children with other FASDs.

## Method

### Participants

Selected data from children seen at FAS DPN clinics between 2000 and 2016 were retrospectively examined. At the time of their diagnostic evaluation, all patients evaluated at these clinics were invited to have their FASD clinical data entered into the FAS DPN research database for use in future research studies. Patient/caregiver consent was obtained in accordance with University of Washington Human Subjects Division oversight and approval.

All patients in the FAS DPN database were evaluated for FASDs using the 4-Digit Diagnostic Code (updated and coded according to criteria from the 2004 version; Astley, 2004), an interdisciplinary approach to diagnosis guided by empirically validated criteria (Astley, 2004, 2013). The four digits of the FASD 4-Digit Diagnostic Code reflect the magnitude of expression of the four key diagnostic features of FAS, in the following order: (a) growth deficiency, (b) FAS facial phenotype, (c) CNS structural/functional abnormalities, and (d) prenatal alcohol exposure. The magnitude of expression of each feature is case defined and ranked independently on a 4-point Likert scale, with 1 reflecting complete absence of the FAS feature and 4 reflecting a strong and classic presentation of the feature. Each Likert rank is specifically case defined. There are 102 four-digit codes that fall broadly under the umbrella of FASDs. These codes cluster into four clinically meaningful diagnostic subcategories (Astley, 2004): FAS, partial FAS (PFAS), static encephalopathy/alcohol exposed (SE/AE), and neurobehavioral disorder/alcohol exposed (ND/AE).

Data used in this study were from children in the FAS DPN research database who met the following inclusionary criteria: (a) between 3.00 and 10.99 years old at the time of diagnostic clinic visit, (b) diagnosed with FASDs (FAS, PFAS, SE/AE, or ND/AE), and (c) had SSP results available in the research database. Any subjects with missing data on more than one third of the items in any SSP domain were excluded.

## Measures

The FAS DPN research database contains more than 2,000 fields of information on each patient evaluated for FASDs. The selected variables used in this analysis were measures related to auditory behavior (auditory filtering scores on the SSP), reported hearing status (hearing loss and history of otitis media), and measures of growth, facial morphology, and CNS structure and function used to derive each child's 4-Digit Diagnostic Code FASD diagnosis. These variables are described in more detail below.

### Auditory Behaviors

**SSP.** The SSP (McIntosh et al., 1999), standardized for ages 3–11 years, is a 38-item caregiver-report questionnaire designed to identify atypical behavioral responses to sensation. A short-form version of the longer Sensory Profile (Dunn, 1999), the SSP encompasses seven domains of sensory processing—tactile sensitivity, taste/smell sensitivity, movement sensitivity, underresponsive/seeking sensation, auditory filtering, low energy/weak, and visual/auditory sensitivity—with total scores in each domain norm-referenced, based on the performance of 1,037 children without known disabilities (Dunn & Brown, 1997). The SSP is considered to be a reliable and valid measurement tool, with high internal consistency for total score (Cronbach's  $\alpha = .96$ ) and section scores (Cronbach's  $\alpha = .82$ –.89; Cronbach's  $\alpha$  for the auditory filtering domain = .87).

Scores from the SSP's auditory filtering domain are the primary focus of this study. The auditory filtering domain comprises six items focused on either attending to or tuning out sound stimuli:

- Item 22: is distracted or has trouble functioning if there is a lot of noise around
- Item 23: appears to not hear what you say (e.g., does not “tune in” to what you say, appears to ignore you)
- Item 24: can't work with background noise (e.g., fan, refrigerator)
- Item 25: has trouble completing tasks when the radio is on
- Item 26: doesn't respond when name is called but you know the child's hearing is OK
- Item 27: has difficulty paying attention

Caregivers rate their child's frequency of atypical sensory behaviors on a Likert scale ranging from 1 (*always exhibits this behavior*) to 5 (*never exhibits this behavior*). Lower scores indicate more atypical behaviors. Total scores across items in each domain are compared to a normative sample in order to derive “cut scores” (Dunn, 1999) that categorize domain scores into classification category groups reflecting either “typical performance” for a domain score  $\leq 1$  SD below the normative mean (i.e., auditory filtering cut scores: 23–30), “probable difference” for a domain score  $> 1$  but  $< 2$  SDs below the normative mean (i.e., auditory filtering cut scores: 20–22), or “definite difference” for a domain

score  $\geq 2$  SDs below the normative mean (i.e., auditory filtering cut scores: 6–19).

We conducted our primary analyses on auditory filtering domain total (summed total of scores on the six auditory filtering items) classification categories. We also examined score profiles across the six individual items. Of particular interest were Items 23 and 26, which are the only auditory filtering items explicitly involving auditory-related tasks (hearing what is said/tuning in and responding when name is called, respectively) and therefore most closely approximate measurable sound-in-noise listening behaviors. In contrast, the other auditory filtering items, particularly Items 24 and 25, appear to be more related to sound sensitivity/distractibility than to listening behaviors.

### Hearing Status

**Hearing loss and history of otitis media.** Data on hearing status are obtained by the FAS DPN team based on a careful review of the records available for a given patient, as hearing function is typically not prospectively evaluated as part of the FAS DPN diagnostic assessment. These clinical records yield heterogeneous information about hearing function (see Table 1), occasionally including thresholds measured during pure-tone audiograms but more typically consisting of formal reports (e.g., medical office hearing screens, school hearing screens, play-based audiometric procedures) indicating whether or not the individual had “passed” a hearing screen (sometimes at a given sound level[s], sometimes not)—in addition to newborn hearing screens based on auditory brainstem response and sometimes otoacoustic emission testing. Caregiver report on functional hearing levels and/or caregiver report regarding history of otitis media was also included. Due to the varying nature and quality of these data, they were used to assess only reported hearing loss and history of otitis media; reliable information related to presumed etiology of hearing loss was not available in this data set. Reported hearing loss was coded dichotomously, defined as audiometric thresholds  $> 25$  dB HL in one or both ears at any frequency tested and/or formal report documenting hearing loss. Normal/functional hearing was defined as (a) audiometric thresholds of 25 dB HL or better across all frequencies tested and/or formal document with report of hearing screen at a given sound successfully passed or (b) caregiver report of no hearing concerns. Hearing data that were inconclusive or unclear—for example, report of failed infant hearing screen but no indication of any subsequent auditory assessment or audiometric thresholds  $> 25$  dB HL but note of concurrent nasal congestion—were classified as missing in order to keep normal/functional hearing and hearing loss categories distinct and operationally valid. Caregiver report of history of otitis media/middle ear infection was also captured based on the number of reported cases: 0, 1, 2, or 3 or more.

### FASD Diagnosis and Measures of Physical Dysmorphia and Alcohol Exposure

**4-Digit Diagnostic Code FASD diagnosis (FAS, PFAS, SE/AE, and ND/AE).** See full description above, along with Astley (2004) for additional details.

**Table 1.** Demographic and clinical profiles of the study sample.

Characteristic	N (valid %)
Gender	
Female	124 (38.2)
Male	201 (61.8)
Age at diagnosis (years)	
3–5.9	117 (36.0)
6–10.9	208 (64.0)
M (SD), range	6.9 (2.1), 3.0–10.9
Race/ethnicity	
Caucasian	157 (48.3)
African American	33 (10.2)
Native American/Canadian	23 (7.1)
Hispanic	14 (4.3)
Other (including mixed race)	98 (30.2)
FASD diagnosis	
FAS	13 (4.0)
PFAS	19 (5.5)
SE/AE	96 (29.5)
ND/AE	197 (60.6)
Type of auditory report	
Threshold info from audiogram	83 (25.5)
Formal report, no thresholds	167 (51.4)
Newborn screening	15 (4.6)
Caregiver report regarding hearing	53 (16.3)
Not reported	7 (2.5)
Short Sensory Profile respondent	
Parent unspecified	170 (52.3)
Foster parent	56 (17.2)
Adoptive parent	27 (8.3)
Legal guardian	15 (4.6)
Other family	47 (14.5)
Not reported	10 (3.1)

*Note.* Demographic and clinical profiles of the 325 children with fetal alcohol spectrum disorder (FASD) in the clinical study sample. FASD diagnoses were made by an interdisciplinary team using the 4-Digit Diagnostic Code (Astley, 2004), based on four clinically meaningful diagnostic subcategories: FAS = fetal alcohol syndrome; PFAS = partial FAS; SE/AE = static encephalopathy/alcohol exposed; and ND/AE = neurobehavioral disorder/alcohol exposed.

**Growth deficiency.** This measure (“growth rank”: 1 = *none*, 2 = *mild*, 3 = *moderate*, and 4 = *severe*), which yields the first digit in the FASD 4-Digit Diagnostic Code, documents the magnitude of prenatal and/or postnatal growth deficiency (see Astley, 2004, for additional details).

**FAS facial phenotype.** This measure (“face rank”: 1 = *none*, 2 = *mild*, 3 = *moderate*, and 4 = *severe*), which yields the second digit in the FASD 4-Digit Diagnostic Code, documents the magnitude of expression of the FAS facial phenotype, as defined by short palpebral fissure lengths, a smooth philtrum, and a thin upper lip (see Astley, 2004, for additional details).

**Likelihood of structural/neurodevelopmental CNS abnormality.** This measure (“CNS rank”: 1 = *unlikely*, 2 = *possible*, 3 = *probable*, and 4 = *definite*) yields the third digit in the FASD 4-Digit Diagnostic Code. The first three levels (1 = *unlikely*, 2 = *possible*, and 3 = *probable*) document presumed likelihood of CNS structural abnormality as assessed by the interdisciplinary FAS DPN team on the basis of a variety of standardized assessments of function including executive function, memory, cognition, social/

adaptive skills, academic achievement, language, motor, attention, and activity level. The fourth level (4 = *definite*) documents potential CNS abnormality of presumed prenatal origin on the basis of structural (e.g., microcephaly or observable brain abnormalities) and/or neurological (e.g., seizures or other hard neurological signs) evidence. To the degree possible, clinical assessment rules out traumatic brain injury or postnatal disease processes unrelated to prenatal alcohol exposure in assigning CNS rank (see Astley, 2004, for additional details).

**Microcephaly.** Microcephaly (0 = *no*, 1 = *yes*) is defined by the FASD 4-Digit Diagnostic Code as an occipital frontal circumference 2 or more *SDs* below the mean ( $\leq$  third percentile; see Astley, 2004, for additional details).

**Prenatal alcohol exposure.** This measure (“alcohol rank”: 1 = *confirmed absence of exposure*; 2 = *unknown exposure*; 3 = *confirmed exposure, level unknown or moderate*; and 4 = *confirmed exposure, level high*), which yields the fourth digit in the FASD 4-Digit Diagnostic Code, documents the magnitude of alcohol exposure. Alcohol exposure is ranked according to the quantity, timing, frequency, and certainty of exposure during pregnancy and is based on best available records and/or direct report from the biological mother/witnesses to the exposure. A diagnosis under the umbrella of FASDs requires a confirmed prenatal alcohol exposure (Rank 3 or 4) with one exception: FAS. A diagnosis of FAS requires the Rank 4 FAS facial phenotype, which is so highly specific to (caused only by) prenatal alcohol exposure that presence of the Rank 4 FAS facial phenotype offsets the need for a confirmed history of alcohol exposure (see Astley, 2004, for additional details).

### Measures of CNS Function

In the context of a careful medical review to document any potential postnatal sources of underlying CNS abnormality (e.g., traumatic brain injury, seizure disorder), the FAS DPN clinical assessment of CNS function includes a review of available neurodevelopmental assessments in the medical record, as well as documentation of current CNS function using common behavioral measures. For the current study, the following clinical measures available in the record were used to gauge the presence and severity of neurodevelopmental CNS impairment.

**Attention-deficit/hyperactivity disorder diagnosis.** This variable (0 = *no*, 1 = *yes*) documents whether the subject has a confirmed previous diagnosis of attention-deficit/hyperactivity disorder (ADHD) from a qualified provider or as a result of the FAS DPN clinical assessment.

**Cognition/full-scale IQ and language function.** The following domains of function were analyzed using a 3-point severity score: 1 = *within normal limits*, that is, performance no lower than 0.9 *SD* below the mean; 2 = *mildly to moderately impaired*, that is, performance 1.0–1.9 *SDs* below the mean on a standardized measure; and 3 = *severely impaired*, that is, performance 2 or more *SDs* below the mean on a standardized measure. The severity score was derived by the FAS DPN clinical team based on aggregate data in each respective domain, including standardized test

scores administered as part of the diagnostic clinic and/or scores available in the clinical records.

Cognitive data included standardized IQ scores from a variety of norm-referenced tests for intellectual and cognitive abilities, including the Wechsler Intelligence Scale for Children (Wechsler, 2003), the Stanford–Binet Intelligence Scales (Bain & Allin, 2005), and the Differential Ability Scales (Elliott, 2007). Normal cognition was defined as full-scale IQ (FSIQ) > 85, mildly impaired as 70 < FSIQ ≤ 85, and severely impaired as FSIQ ≤ 70.

Language function was quantified by a range of both standardized and structured clinical assessments, including but not limited to the various editions of the Clinical Evaluation of Language Fundamentals (Semel, Wiig, & Secord, 2013), the Preschool Language Scale (Zimmerman, Steiner, & Pond, 2002), and the Comprehensive Assessment of Spoken Language (Carrow-Woolfolk, 1999), and assessment of narrative production. Both direct testing and information from a detailed review of records were used to establish a clinical ranking based on the 3-point severity scale described above.

## Analyses

All analyses were conducted using SPSS Version 19.0. Descriptive statistics (means, standard deviations, proportions) were used to profile the study population. Relationships between auditory filtering domain score categories (typical performance, probable difference, and definite difference) and selected clinical variables (Aims 1 and 2) were assessed using chi-squared ( $\chi^2$ ) tests when outcomes were assessed on nominal scales and Mantel–Haenszel linear-by-linear association chi-squared tests of trend ( $\chi^2_{MH}$ ) when outcomes were assessed on ordinal scales. Similarly, relationships between variables related to peripheral hearing status and selected clinical variables (Aim 3) were evaluated using chi-squared, linear-by-linear, and Fisher’s exact tests, as appropriate. Results were considered significant at two-sided  $p$  values of < .05. The effect sizes of significant results were estimated using Cramer’s  $V$  ( $\phi_C$ ) for chi-squared analyses, Spearman correlation coefficient ( $\rho$ ) for linear-by-linear trend tests (Agresti, 2007), and phi coefficients ( $\phi$ ) for Fisher’s exact results. Post hoc tests were performed for significant omnibus chi-squared statistics by estimating the  $p$  value of the adjusted residuals (i.e., the  $z$  scores). Because of the exploratory nature of the study,  $p$  values for post hoc analyses should be interpreted with caution.

## Results

The data set analyzed ( $N = 325$ ) consisted of 124 girls and 201 boys, ranging in age from 3.03 to 10.97 years. Table 1 provides demographic and clinical characteristics and data type/origin. In this sample, the largest proportion of children were diagnosed with ND/AE, followed by SE/AE. As expected, about one of 10 children was diagnosed with FAS or PFAS. This study sample is a good representation of the larger FAS DPN population (see Astley, 2010; see Table 2).

Of the 325 participants who met the inclusionary criteria (which included data available for at least two thirds of the SSP items in any given domain), 43 individuals were still missing some SSP data, nine of whom were missing data in the auditory filtering domain. To handle these missing data, the average of the subjects’ remaining scores in the incomplete domain was calculated, and this value replaced the missing score(s) in that domain, for that subject. This approach was selected to control for bias while still fairly representing the child’s auditory filtering profile. To maintain the ordinal format of the data, those newly calculated item scores that were not whole numbers were rounded to the closest whole number.

### *Aim 1: Prevalence of Atypical Auditory Behaviors Across FASD Diagnoses*

A majority (80.0%) of children with FASDs in our clinical sample were rated by their caregivers as exhibiting a definite difference (2  $SD$ s below the normative mean) on the auditory filtering domain of the SSP (see Figure 1). No significant linear relationship was observed between outcomes on auditory filtering domain score categories and severity of FASD diagnosis; however, the raw cross-tabulation numbers unexpectedly show that the percentage of children diagnosed with FAS who were reported with a definite difference on the auditory filtering items (61.5%) was lower than the equivalent figure for children with either PFAS (78.9%), SD/AE (87.5%), or ND/AE (77.7%). Correspondingly, the percentage of children with FAS rated with typical performance was higher than those with less severe diagnoses (see Table 2, top). When the cross-tabulation was collapsed to assess the prevalence of definite difference reported among FAS (61.5%) versus all other FASD diagnoses combined (80.8%), the contrast remained nonsignificant (Fisher’s exact test,  $p = .19$ ).

Due to this unexpected (albeit nonsignificant) finding in which children with less severe FASD diagnoses were more frequently rated with atypical auditory behaviors than were children with FAS, post hoc analyses were conducted to more closely examine the relationships between auditory filtering outcomes and various physical features of FASDs (i.e., growth deficiency, FAS facial features, likelihood of CNS structural abnormalities and microcephaly), in addition to prenatal alcohol exposure (see Table 2, bottom). The post hoc analyses revealed a similar pattern of association. Atypical auditory behaviors were prevalent across the full spectrum of physical outcomes and tended to be more prevalent among those with less severe physical outcomes, especially those with less severe facial features.

### *Aim 2: Relationship Between Atypical Auditory Behaviors and Measures Indicative of Reported Hearing Dysfunction or CNS Impairment*

No observable relationship was found between outcomes on auditory filtering domain score categories and measures of peripheral hearing status, either reported hearing loss or

**Table 2.** Prevalence of Short Sensory Profile auditory filtering domain scores across fetal alcohol spectrum disorder (FASD) diagnoses.

FASD diagnostic features	Auditory filtering domain score category			Statistic ( <i>p</i> ) [effect size]
	Definite difference	Probable difference	Typical performance	
	<i>n</i> (valid %)	<i>n</i> (valid %)	<i>n</i> (valid %)	
Prevalence				
Overall	260 (80.0)	33 (10.2)	32 (9.8)	—
By FASD diagnosis				Linear $\chi^2_{MH}(1, N = 325) = 0.02 (.88)$
ND/AE	153 (77.7)	21 (10.7)	23 (11.7)	
SE/AE	84 (87.5)	8 (8.3)	4 (4.2)	
PFAS	15 (78.9)	3 (15.8)	1 (5.3)	
FAS	8 (61.5)	1 (7.7)	4 (30.8)	
Post hoc analyses: association between SSP auditory filtering domain score categories and physical features of FASD and prenatal alcohol exposure				
Growth deficiency (growth rank)				Linear $\chi^2_{MH}(1, N = 325) = 1.59 (.21)$
1: Normal	199 (80.6)	26 (10.5)	22 (8.9)	
2: Mild	31 (83.8)	2 (5.4)	4 (10.8)	
3: Moderate	16 (76.2)	3 (14.3)	2 (9.5)	
4: Severe	14 (70.0)	2 (10.0)	4 (20.0)	
FAS facial phenotype (face rank)				Linear $\chi^2_{MH}(1, N = 325) = \mathbf{18.48 (<.001)}$ [ <i>p</i> = <b>-.24</b> ]
1: Normal	147 (89.1)	11 (6.7)	7 (4.2)	
2: Mild	82 (72.6)	16 (14.2)	15 (13.3)	
3: Moderate	21 (67.7)	4 (12.9.7)	6 (19.4)	
4: Severe	10 (62.5)	2 (12.5)	4 (25.0)	
Likelihood of CNS structural abnormality (CNS rank)				Linear $\chi^2_{MH}(1, N = 325) = 0.52 (.47)$
1: Unlikely	0 (0.0)	0 (0.0)	0 (0.0)	
2: Possible	153 (77.7)	21 (10.7)	23 (11.7)	
3: Probable	65 (87.8)	6 (8.1)	3 (4.1)	
4: Definite	42 (77.8)	6 (11.1)	6 (11.1)	
Microcephaly				Linear $\chi^2_{MH}(1, N = 321) = 2.24 (.13)$
No	227 (81.1)	28 (10.0)	25 (8.9)	
Yes (< 3 percentile)	30 (73.2)	4 (9.8)	7 (17.1)	
Prenatal alcohol exposure (alcohol rank)				Linear $\chi^2_{MH}(1, N = 323) = 2.47 (.12)$
3: Moderate	124 (77.5)	16 (10.0)	20 (12.5)	
4: High	135 (82.8)	17 (10.4)	11 (6.7)	

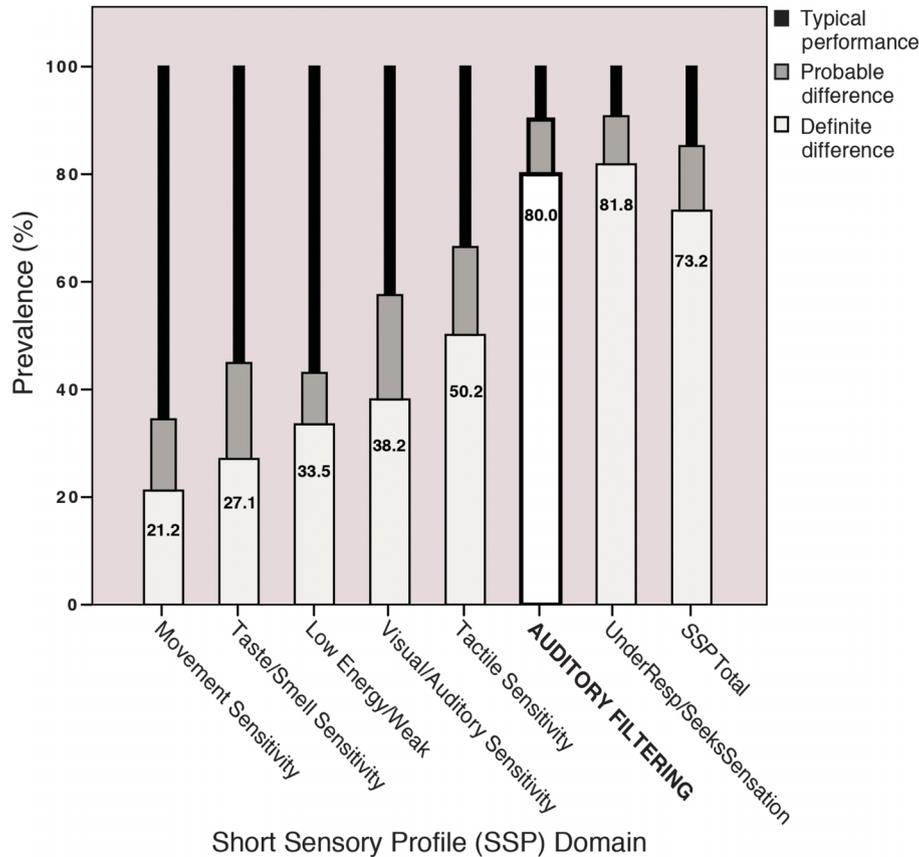
*Note.* Prevalence of Short Sensory Profile auditory filtering domain scores across FASD diagnoses. No linear relationship between auditory filtering scores (“definite difference”: domain score  $\geq 2$  SDs below the normative mean; “probable difference”: domain score  $> 1$  but  $< 2$  SDs below the normative mean; or “typical performance”: domain score  $\leq 1$  SD below the normative mean) and FASD diagnosis was observed (FASD diagnostic abbreviations [Astley, 2004]: fetal alcohol syndrome [FAS], partial FAS [PFAS], static encephalopathy/alcohol exposed [SE/AE], and neurobehavioral disorder/alcohol exposed [ND/AE]). Post hoc analyses of the relationship between auditory filtering scores and physical features of FASD show that atypical auditory behaviors tended to be more prevalent among those with less severe physical outcomes, especially less severe facial features. The four key diagnostic features of FASD (growth deficiency, FAS facial phenotype, CNS abnormalities, and prenatal alcohol exposure) are case defined on 4-point Likert scales (ranks) and comprise the four digits of the FASD 4-Digit Diagnostic Code (Astley, 2004). Alcohol Ranks 1 and 2 are not included here because individuals diagnosed with FASD cannot by definition have Alcohol Rank 1 (confirmed lack of prenatal alcohol exposure) and Alcohol Rank 2 (unknown, indicates unknown prenatal alcohol exposure). SSP = Short Sensory Profile; CNS = central nervous system. Bold values are significant at  $p < .05$ .

history of otitis media (see Table 3, top). With respect to measures associated with CNS impairment (see Table 3, bottom), there was no observable relationship between auditory filtering outcomes and FSIQ scores or language function; however, a relationship between auditory filtering scores and ADHD diagnosis was observed. Post hoc analyses documented a high prevalence of children with a definite difference in auditory filtering scores among those with (88.3%) and without (72.6%) ADHD, but the prevalence was significantly higher among those with ADHD.

We additionally assessed the effect of age on auditory filtering outcomes in order to ensure that none of the observed relationships was confounded by age. Based on data reported by Dunn (1999) indicating that younger and older children may score differently on some sections of the long-

form Sensory Profile (with scores indicating that younger children may perform better on items related to inattention/distractibility), we split the sample into two groups. They were (a) preschool children (3.00–5.99 years old at the time of FAS DPN clinic visit) and (b) school-aged children (6.00–10.99 years old at clinic visit). This split was also motivated by the knowledge that school-aged children may have a wider range of cognitive assessments available in the clinical record to examine the relationships between auditory behaviors and CNS function. A chi-squared test comparing outcomes on auditory filtering domain score categories across groups did show an effect of age,  $\chi^2(2, N = 325) = 9.40, p = .009, \phi_C = .17$ , with post hoc comparisons indicating that definite differences in auditory filtering were more likely to be reported ( $p = .005$ ) in the older age group

**Figure 1.** Profile of Short Sensory Profile domain scores in the clinical study sample of 325 children diagnosed with fetal alcohol spectrum disorders. Represented here is the percentage of children whose total score in each domain (and total score across all domains: “SSP Total”) falls into the categories of “definite difference” (light gray bars, except in the case of the auditory filtering domain, which is white for emphasis); domain score  $\geq 2$  SDs below the normative mean; “probable difference” (dark gray bars); domain score  $> 1$  but  $< 2$  SDs below the normative mean; or “typical performance” (black bars); domain score  $\leq 1$  SD below the normative mean. Auditory filtering domain scores showed among the highest ratings of children with definite and probable differences.



(85.1% with a definite difference among children 6 years of age and older) than in the younger (70.9% with a definite difference among those less than 6 years of age), consistent with Dunn’s findings. Based on this information, we separately assessed the influence of age on the relationship between auditory filtering outcomes and each of the diagnostic, neurobehavioral, and hearing-related variables probed in the study. We found that the presence (or lack thereof) of a linear relationship to auditory filtering outcomes did not differ between the two age groups for any of the variables examined with the exception of language function, where a linear relationship between auditory filtering scores and language function was observed in the older group,  $\chi^2_{MH}(1, N = 199) = 6.11, p = .013$ , but not the younger,  $\chi^2_{MH}(1, N = 110) = 1.67, p = .194$ . The prevalence of older children with a definite difference in auditory filtering scores was highest among those rated as having “severely impaired” language function (92.0%). This is approximately 10 percentage points higher than among those with “mildly to moderately impaired” language function (82.9%) and those without language impairment (79.6%).

### ***Aim 3: Assessing the Relationship Between Hearing Status and FASD Diagnostic Measures***

Relationships between measures of peripheral hearing status and FASD diagnosis—including component assessments of the presence of the physical features of FASDs (FAS facial phenotype, growth deficiency, likelihood of CNS structural abnormality, and microcephaly), magnitude of prenatal alcohol exposure, and FSIQ—were assessed (see Table 4). Reported hearing loss (defined as reported audiometric thresholds  $> 25$  dB HL in one or both ears at any frequency tested or formal report documenting hearing loss) was 16-fold more prevalent among the children diagnosed with FAS (40%: four individuals with reported hearing loss out of the 10 individuals with FAS with sufficient hearing/audiometric information in the data set) than among the children diagnosed with other FASD diagnoses (2.4%: seven with reported hearing loss out of 286 individuals [PFAS = 17, SE/AE = 88, ND/AE = 181]; see Figure 2, left). The prevalence of hearing loss increased linearly with increasing severity of FASD diagnosis. In contrast, no significant

**Table 3.** Short Sensory Profile auditory filtering domain scores: association with reported hearing status and central nervous system (CNS) function.

Hearing and CNS status	Auditory filtering domain categories			Statistic ( <i>p</i> ) [effect size]
	Definite difference <i>n</i> (valid %)	Probable difference <i>n</i> (valid %)	Typical performance <i>n</i> (valid %)	
Reported hearing loss (HL)				Linear $\chi^2_{MH}(1, N = 296) = 0.24 (.62)$
Normal/functional	233 (81.8)	27 (9.5)	25 (8.8)	
HL (one or both ears)	9 (81.8)	0 (0.0)	2 (18.2)	
Reported otitis media				Linear $\chi^2_{MH}(1, N = 319) = 1.89 (.17)$
No cases reported	132 (77.6)	16 (9.4)	22 (12.9)	
1–2 cases reported	36 (85.7)	4 (9.5)	2 (4.8)	
3+ cases reported	87 (81.3)	13 (12.1)	7 (6.5)	
Full-scale IQ (standard score)				Linear $\chi^2_{MH}(1, N = 287) = 2.11 (.15)$
Normal: > 85	98 (78.4)	11 (8.8)	16 (12.8)	
Mildly impaired: > 70 and ≤ 85	93 (80.2)	15 (12.9)	8 (6.9)	
Severely impaired: ≤ 70	40 (87.0)	3 (6.5)	3 (6.5)	
ADHD diagnosis				Linear $\chi^2_{MH}(1, N = 309) = \mathbf{15.79 (< .001)} [\rho = .21]$
No	119 (72.6)	19 (11.6)	26 (15.9)	
Yes	128 (88.3)	13 (9.0)	4 (2.8)	
Language function				Linear $\chi^2_{MH}(1, N = 309) = 1.04 (.31)$
Normal	76 (77.6)	9 (9.2)	13 (13.3)	
Mildly impaired	82 (83.7)	9 (9.2)	7 (7.1)	
Severely impaired	92 (81.4)	12 (10.6)	9 (8.0)	

*Note.* Association between Short Sensory Profile auditory filtering domain scores (“definite difference”: domain score  $\geq 2$  SDs below the normative mean; “probable difference”: domain score  $> 1$  but  $< 2$  SDs below the normative mean; or “typical performance”: domain score  $\leq 1$  SD below the normative mean) and reported hearing status and CNS function. Hearing loss defined as thresholds  $> 25$  dB HL at any frequency tested and/or formal report documenting hearing loss. Whereas atypical auditory behaviors were more prevalent among children with attention-deficit/hyperactivity disorder (ADHD), no linear relationships between auditory filtering scores and measures of hearing status were observed. See text for operational definitions of the three language function categories. Bold values are significant at  $p < .052$ .

relationship between history of otitis media and FASD diagnosis was observed (see Table 4 and Figure 2 [right]).

The prevalence of reported hearing loss increased significantly and linearly with increasing severity of growth deficiency, FAS facial phenotype, likelihood of CNS structural abnormality, and FSIQ deficit. Reported hearing loss was also significantly more prevalent among children with microcephaly. Although the prevalence of reported hearing loss among the children with high alcohol exposure was twofold greater than among children with moderate exposures, this contrast was not statistically significant. With respect to reported history of otitis media, no significant associations were observed with any of the FASD diagnostic variables examined.

## Discussion

The present results were obtained from a retrospective examination of data collected from children who were diagnosed with FASDs at Washington State FAS DPN clinics. They indicate that a large proportion of children with FASDs exhibit atypical, higher order auditory behaviors involving attending to or tuning out sound stimuli, as measured by caregiver ratings on the auditory filtering domain of the SSP. The prevalence of atypical auditory behaviors in this population was not observed to be significantly related to severity of FASD diagnosis; that is, a comparably high

proportion of children were reported with a definite difference in auditory filtering across fetal alcohol diagnoses. No relationship was observed between atypical auditory behaviors and reported estimates of peripheral hearing function—including hearing loss and history of otitis media potentially indicative of conductive hearing issues—based on data obtained from caregiver-provided records and reports. There was a relationship, however, between atypical auditory behaviors and ADHD diagnosis, presumably reflective of underlying CNS impairment. Children with ADHD were more likely to be reported with atypical behaviors in the auditory filtering domain. The prevalence of atypical auditory behaviors was not observed to be correlated across the sample with other measures of CNS function, including measures of cognition and language function.

Reported hearing loss was found to be related to FASD diagnosis; that is, hearing loss was 16-fold more prevalent among children with FAS (40%) than among children with other FASDs (2.4%), for whom the prevalence of hearing loss was similar to that estimated for the general U.S. adolescent population (2.3%; Lin, Niparko, & Ferrucci, 2011). An increased prevalence of reported hearing loss was also correlated with the presence of physical features associated with FAS: growth deficiency, facial dysmorphism, likelihood of CNS damage, and microcephaly. Reported hearing loss was also related to lower FSIQ scores but was not observed to be significantly related to the reported

**Table 4.** Association between reported hearing loss, otitis media, fetal alcohol spectrum disorder (FASD) diagnosis, and FASD features.

FASD diagnostic features	Reported hearing loss (one or two ears)		Reported cases: otitis media (OM)	
	Hearing loss, <i>n</i> (valid %)	Statistic ( <i>p</i> ) [effect size]	OM: 3+ cases, <i>n</i> (valid %)	Statistic ( <i>p</i> ) [effect size]
FASD diagnosis	Linear $\chi^2_{MH}(1, N = 296) = 17.77 (< .001) [\rho = -.15]$		Linear $\chi^2_{MH}(1, N = 319) = 0.23 (.63)$	
ND/AE	4 (2.2)		71 (36.4)	
SE/AE	2 (2.3)		27 (29.0)	
PFAS	1 (5.9)		4 (21.1)	
FAS	4 (40.0)		5 (41.7)	
Growth deficiency (growth rank)	Linear $\chi^2_{MH}(1, N = 296) = 7.32 (.007) [\rho = .15]$		Linear $\chi^2_{MH}(1, N = 319) = 1.15 (.28)$	
1: Normal	5 (2.2)		83 (34.3)	
2: Mild	3 (8.3)		13 (37.1)	
3: Moderate	0 (0.0)		6 (28.6)	
4: Severe	3 (17.6)		5 (23.8)	
FAS facial phenotype (face rank)	Linear $\chi^2_{MH}(1, N = 296) = 16.74 (< .001) [\rho = .17]$		Linear $\chi^2_{MH}(1, N = 319) = 0.52 (.47)$	
1: Normal	3 (1.9)		59 (36.6)	
2: Mild	2 (2.0)		33 (29.5)	
3: Moderate	2 (7.7)		9 (29.0)	
4: Severe	4 (30.8)		6 (40.0)	
Likelihood of CNS structural abnormality (CNS rank)	Linear $\chi^2_{MH}(1, N = 296) = 4.47 (.04) [\rho = .12]$		Linear $\chi^2_{MH}(1, N = 319) = 0.45 (.50)$	
1: Unlikely	0 (0.0)		0 (0.0)	
2: Possible	4 (2.2)		71 (36.4)	
3: Probable	3 (4.2)		20 (27.8)	
4: Definite	4 (9.1)		16 (30.8)	
Microcephaly	Fisher's exact ( <i>N</i> = 292) (.02) [ $\phi = .15$ ]		Linear $\chi^2_{MH}(1, N = 314) = 0.94 (.33)$	
No	7 (2.7)		95 (34.5)	
Yes ( $\leq$ 3rd percentile)	4 (11.8)		10 (25.6)	
Prenatal alcohol exposure (alcohol rank)	Fisher's exact ( <i>N</i> = 294) (.22)		Linear $\chi^2_{MH}(1, N = 317) = 0.81 (.37)$	
3: Moderate	3 (2.0)		47 (29.9)	
4: High	8 (5.4)		59 (36.9)	
Full-scale IQ (standard score)	Linear $\chi^2_{MH}(1, N = 259) = 4.00 (.045) [\rho = .12]$		Linear $\chi^2_{MH}(1, N = 282) = 3.71 (.05)$	
Normal ( $> 85$ )	1 (0.9)		48 (39.0)	
Mildly impaired ( $> 70$ and $\leq 85$ )	4 (3.8)		35 (30.7)	
Severely impaired: ( $\leq 70$ )	3 (6.8)		11 (24.4)	

*Note.* Association between hearing status (reported hearing loss and otitis media) and FASD diagnosis (abbreviations: ND/AE = neurobehavioral disorder/alcohol exposed; SE/AE = static encephalopathy/alcohol exposed; PFAS = partial FAS; and FAS = fetal alcohol syndrome [Astley, 2004]) and diagnostic physical features of FASDs. Reported hearing loss (see text for operational definition) was more prevalent among children diagnosed with FAS than with other FASD diagnoses, whereas no significant relationship between history of otitis media and FASD diagnosis was observed. (Values for *n* and valid % are shown here only for 3+ cases of otitis media, but analyses related to otitis media are conducted across three categories: 0 cases, 1–2 documented cases, and 3+ cases.) The prevalence of reported hearing loss increased with increasing severity of growth deficiency, FAS facial phenotype, likelihood of central nervous system (CNS) structural abnormality, microcephaly, and full-scale IQ deficit. No such relationships were observed with respect to reported history of otitis media; no significant associations were observed with any of the FASD diagnostic variables examined. The four key diagnostic features of FASD (growth deficiency, FAS facial phenotype, CNS abnormalities, and prenatal alcohol exposure) are case defined on 4-point Likert scales (ranks) and comprise the four digits of the FASD 4-Digit Diagnostic Code (Astley, 2004). Alcohol Ranks 1 and 2 are not included here because individuals diagnosed with FASD cannot by definition have Alcohol Rank 1 (confirmed lack of prenatal alcohol exposure) and Alcohol Rank 2 (unknown, indicates unknown prenatal alcohol exposure). Bold values are significant at  $p < .05$ .

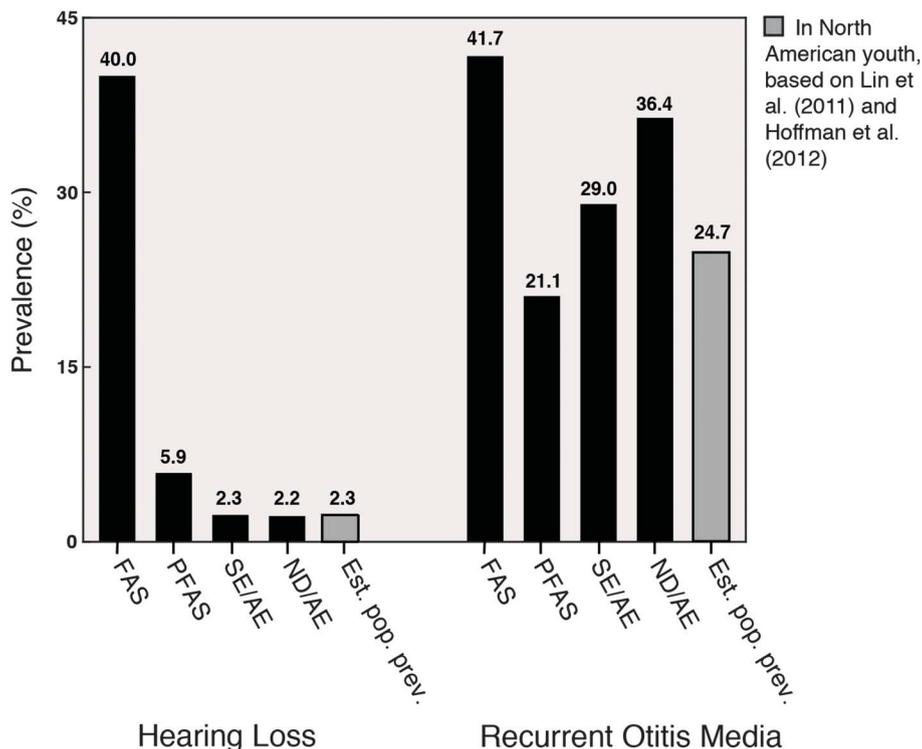
magnitude of prenatal alcohol exposure. In contrast, reported history of otitis media did not differ according to any of the above measures, including FASD diagnosis or the physical features of FAS. It is important to note that reported hearing loss was based on retrospective record review, not prospective audiologic assessment. Moreover, a relatively small number of individuals in the study sample were reported with hearing loss. Based on these two limitations, the present findings related to hearing loss should be interpreted with caution.

### ***Auditory Dysfunction in Children With FASDs***

The observed prominence of auditory filtering as one of the SSP domains in which caregivers rated their children

with the highest frequencies of problem behaviors is consistent with previous findings in children diagnosed with FASDs (Abele-Webster et al., 2012; Carr et al., 2010; Franklin et al., 2008; Hansen & Jirikovic, 2013). Although there is evidence to suggest that one of the teratogenic effects of alcohol is insult to the developing auditory periphery that may impact audiometric hearing thresholds (Church & Kaltenbach, 1997), less is known about damage to other mechanisms—potentially centrally mediated—that may be implicated in the sound-in-noise listening and sound sensitivity/distractibility behaviors probed by the SSP. The present finding that the prevalence of atypical auditory behavior reported on the SSP is comparably high across the fetal alcohol spectrum is consistent with data from Carr et al. (2010) showing that 88% of children

**Figure 2.** Reported hearing loss (see text for operational definition) and recurrent otitis media (3+ occurrences) across fetal alcohol spectrum disorder (FASD) diagnoses (FASD diagnostic abbreviations [Astley, 2004]: fetal alcohol syndrome [FAS], partial FAS [PFAS], static encephalopathy/alcohol exposed [SE/AE], and neurobehavioral disorder/alcohol exposed [ND/AE]) and diagnostic/physical features of FASDs. Reported hearing loss was 16-fold more prevalent among children diagnosed with FAS (four out of 10 individuals with FAS were reported with hearing loss) than with other FASD diagnoses (seven out of 286 individuals reported with hearing loss; a prevalence similar to that observed in the general U.S. adolescent population [Lin et al., 2011]). History of recurrent otitis media was not observed to vary significantly across FASD diagnoses.



with alcohol-related neurodevelopmental disorder (analogous to ND/AE and SE/AE) and 80% of those with PFAS were rated with definite auditory filtering differences. Together, these findings suggest that these atypical auditory behaviors are widespread in individuals with FASDs, even those without the observable physical markers of FAS or PFAS. Although the prevalence of FAS itself is estimated to be one to three per 1,000 live births in the general population (Stratton, Howe, & Battaglia, 1996), individuals with fetal alcohol-related disorders across the entire spectrum are far more numerous (see Astley, 2010; Sampson et al., 1997), underscoring the clinically driven imperative to better characterize and understand auditory dysfunction across FASDs.

It is notable, however, that the atypical auditory behaviors seen on the SSP are not specific to FASDs. Whereas low auditory filtering scores do not appear to be common in typically developing children (for whom Tomchek and Dunn [2007] observed the lowest ratings of definite differences [3.1%] across domains to be in auditory filtering), children with a variety of neurodevelopmental disorders are frequently rated with atypical auditory filtering scores. In particular, 78% of toddlers (Tomchek & Dunn, 2007) and 70% of older children (Green, Chandler, Charman, Simonoff, & Baird, 2016) with autism spectrum disorder (ASD) were rated with a definite difference in the auditory

filtering domain (see also Al-Heizan, AlAbdulwahab, & Kachanathu, 2015; Ashburner, Ziviani, & Rodger, 2008; Baker, Lane, Angley, & Young, 2008; Chen, Rodgers, & McConachie, 2009; O'Donnell, Deitz, Kartin, Nalty, & Dawson, 2012; Tomchek, Huebner, & Dunn, 2014). For children with Down syndrome (Bruni, Cameron, Dua, & Noy, 2010), Williams syndrome (John & Mervis, 2010), and moderate intellectual developmental disabilities (Engel-Yeger, Hardal-Nasser, & Gal, 2011), SSP profiles in which definite differences in auditory filtering scores deviate from typical (43%, 59%, and 50%, respectively), but not notably more so than other domains, have also been observed.

### ***Hearing Loss Versus Listening Difficulties in Children With FASDs***

The lack of any observed relationship between low auditory filtering scores in children with FASDs and reported hearing status (either hearing loss or general conductive hearing concerns as represented by episodes of otitis media) preliminarily suggests that audibility was not a factor in the atypical auditory behaviors reported on the SSP. The finding that audibility issues (i.e., reported hearing loss) were no more prevalent in children diagnosed with an FASD

other than FAS compared to the general U.S. adolescent population (Lin et al., 2011), but were elevated in children with FAS, is consistent with previous data (Church & Gerkin, 1988; Church et al., 1997; Cohen-Kerem et al., 2007; Rössig et al., 1994). Church and Kaltenbach (1997) hypothesized that there may be a relationship between the craniofacial abnormalities seen in FAS and inner ear dysfunction, based on shared embryological origins in ectodermal tissue potentially susceptible to damage from prenatal alcohol exposure. The present finding that reported hearing loss is related to the severity of facial dysmorphism supports this hypothesis. However, hearing loss was additionally related to all of the other physical markers of FAS—including growth deficiency, likelihood of CNS structural abnormality, and microcephaly—which may or may not be associated with neuroectodermal impairments. It is possible that the gross structural sequelae of prenatal alcohol exposure—including structural deficits involving the inner ear—are somehow related. Notably, no significant relationship was seen between hearing loss and magnitude of prenatal alcohol exposure, although the lack of relationship observed may be due to potential unreliability of the data.

In contrast, reported episodes of otitis media typically associated with middle ear function were not observed to vary across FASD diagnoses. Rates of recurrent otitis media did not appear to differ greatly than would be expected in typically developing American children (Hoffman et al., 2013), contrary to past observations of increased otitis media in FAS (Church et al., 1997; Rössig et al., 1994). This difference may be due to the widespread adoption of the pneumococcal conjugate vaccine beginning in the early 2000s (Qureishi, Lee, Belfield, Birchall, & Daniel, 2014), which is reported to have precipitated a 28% drop in the annual prevalence of otitis media diagnoses in the United States between 1997 and 2007 (Hoffman et al., 2013).

Due to the reliance on retrospective record review rather than direct audiologic assessment at the time of the FAS DPN clinic visit, the above findings are preliminary. However, they do suggest that audibility is not a factor in the atypical auditory behaviors reported in children with FASDs. These listening difficulties—namely, problems with hearing or listening despite normal audiometry—are prevalent across FASDs. Listening difficulties, which have been linked to the controversial clinical diagnosis of (central) auditory processing disorder ([C]APD; American Academy of Audiology, 2010; American Speech-Language-Hearing Association, 2005), are frequently reported in children with a variety of developmental disorders, including specific language impairment, dyslexia, and ADHD (reviewed in de Wit et al., 2018), in addition to children without any known neurodevelopmental disorders. Depending on the diagnostic criteria they used, Wilson and Arnott (2013) found that anywhere from 7% to 96% of a sample of typically developing children could be said to have listening difficulties potentially indicative of CAPD.

There is a vast array of distinct but interrelated, centrally mediated suprathreshold auditory processes that support sound-in-noise listening, including accurate lower

order encoding of temporal and spatial attributes of the sound signal (reviewed in Bharadwaj et al., 2015; Stecker & Gallun, 2012) and intact spatial hearing processes (Cameron, Dillon, Glyde, Kanthan, & Kania, 2014). Each of these processes is critical to higher order operations by which the brain segregates the multitude of sound signals arriving at the ear into meaningful “auditory objects” (Griffiths & Warren, 2004; Shinn-Cunningham, 2008). Moreover, as detailed by D. R. Moore (2018), complex listening tasks such as those probed by some of the SSP auditory filtering items and other more auditory-specific tests depend not only on this interrelated network of central auditory processes but also on mechanisms more traditionally associated with the auditory periphery (e.g., high-threshold auditory nerve fiber synapses; see Kujawa & Liberman, 2015) and on central processes typically considered cognitive (e.g., attention, memory, and emotion). These cognitive processes interact with but may or may not involve auditory-specific components. Attention and other cognitive control processes are necessary to focus cognitive resources on the sound target (Anderson & Kraus, 2010; Eckert, Teubner-Rhodes, & Vaden, 2016; Fritz, Elhilali, David, & Shamma, 2007). In addition, they facilitate the quick switching of attention between spatiotemporal acoustic cues in order to enhance focus on relevant sound sources/speakers (an operation termed *active listening*, which is essential to following conversations in noisy, multitalker settings; see Larson & Lee, 2013, 2014; Shinn-Cunningham et al., 2017). All of these suprathreshold mechanisms are susceptible to breakdown and may be implicated in the listening difficulties observed in children with FASDs.

Although it is beyond the scope of this study to investigate the mechanisms of impairment underlying listening difficulties in children with FASDs, the preliminary finding that ADHD was related to atypical auditory filtering scores suggests that attention may play an important role in such auditory dysfunction. A relationship between the features of ADHD and presumed (C)APD has been previously reported (reviewed in de Wit et al., 2018), leading to a suggestion that (C)APD may, in some cases, simply reflect amodal attentional deficits (absent any auditory-specific processes) already captured by ADHD diagnosis. Of three studies reviewed by de Wit et al. that compare performance on various standardized assessments between children diagnosed with presumed (C)APD and those with ADHD, one observed auditory and visual duration pattern test performance differences between groups (Bellis, Billiet, & Ross, 2011). Although it is not possible based on the present results to ascertain whether auditory-specific or more general attentional deficits are related to listening difficulties in FASDs, previous evidence suggests that auditory filtering ratings do capture a construct distinct from ADHD. In a study of 46 children with FASDs, Carr et al. (2010) found that some auditory filtering scores differed according to FASD diagnosis, yet only one participant had ADHD, although all had received formal assessment. Moreover, Abele-Webster et al. (2012) found no correlation between the attention-deficit/hyperactivity index of the Conners' Parent Rating

Scales and any SSP domain scores and concluded that sensory processing problems in children with FASDs are distinct from ADHD. Although rates of comorbid ADHD and FASDs are typically high—53.9% of the larger FAS DPN population was diagnosed with ADHD (Astley, 2010, Table 11), and other researchers have estimated that up to 95% of children with FASDs have ADHD (Fryer, McGee, Matt, Riley, & Mattson, 2007)—Kodali et al. (2017) report that patterns of brain activity related to executive function differ between the two disorders. A meta-analysis by Kingdon, Cardoso, and McGrath (2016) also showed distinct patterns of executive function behavior deficits, with more extensive set-shifting impairments in FASDs. Such set-shifting impairments may be associated with attentional switching deficits that could negatively impact active listening behaviors.

To get a better sense of the specific behaviors driving the low auditory filtering scores observed in this study, we explored the overall caregiver-response profile across individual auditory filtering items (22–27; see Figure 3). Although not formulated to specifically probe sound-in-noise listening, some of the SSP items at least partially capture these behaviors. Items 23 (“appears to not hear what you say [for example, does not ‘tune in’ to what you say, appears to ignore you]”) and 26 (“doesn’t respond when name is called but you know the child’s hearing is OK”) do explicitly assess listening behaviors, and although there is no mention of the acoustic environment, it is reasonable to assume that noise may play a role in the scenarios caregivers envision when rating their child’s behavior. In contrast, Items 24 (“can’t work with background noise [for example, fan, refrigerator]”) and 25 (“has trouble completing tasks when the radio is on”) appear to interrogate behaviors related to sound sensitivity or distractibility. As can be seen in Figure 3, the percentage of caregivers responding that their child “always or ‘frequently’” exhibits the described negative behavior was higher for Items 23 and 26 (black bars) than for Items 24 and 25 (gray bars), across FASD diagnoses. This suggests that problematic listening behaviors more strongly impacted the low auditory filtering ratings observed than did sound sensitivity/distractibility. Interestingly, Tomchek and Dunn (2007) reported similar results in children with ASD, with 73% and 51% responding “always/frequently” to Items 23 and 26, respectively, versus 13% and 16% to Items 24 and 25. Other auditory filtering items that consistently received atypical ratings for the children with FASDs in the study sample were Items 27 (“has difficulty paying attention”) and 22 (“is distracted or has trouble functioning if there is a lot of noise around”), neither of which explicitly probes listening or even (in the case of Item 27) sound-related behavior but does clearly address attentional abilities. Negative behaviors on these items were also similarly elevated in Tomchek and Dunn’s sample of children with ASD, with 79% responding “always/frequently” to Item 27 and 58% to Item 22.

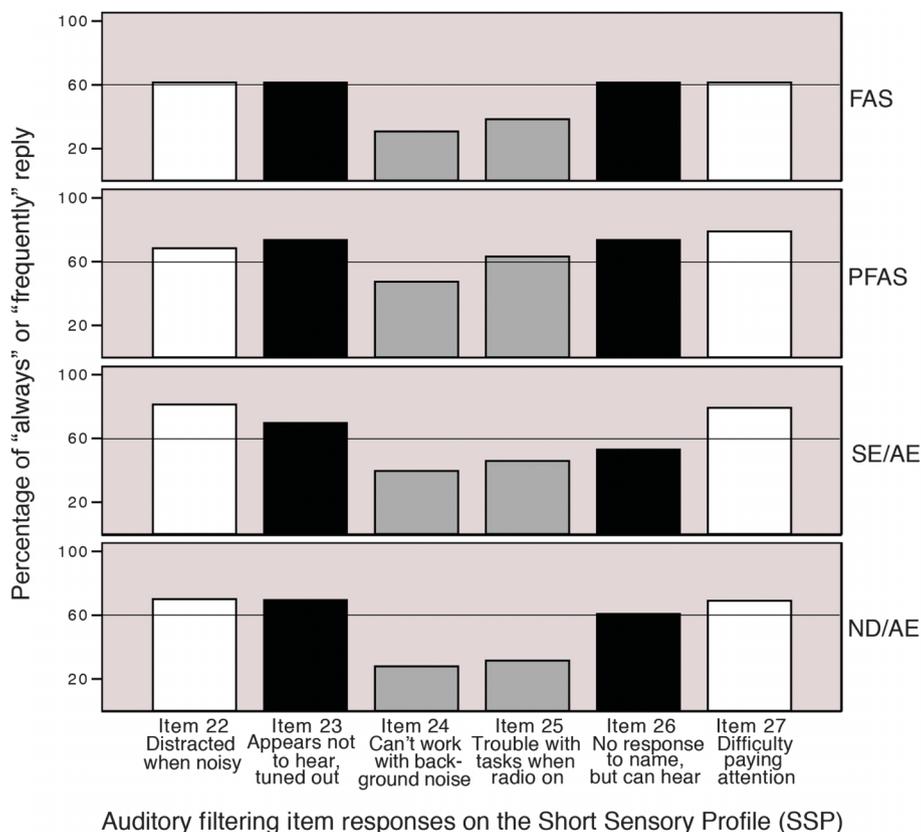
No relationship was observed across the sample between auditory filtering scores and the other putative measures of CNS-mediated processing examined: cognition (FSIQ)

and language function. The lack of correlation with IQ is consistent with previous studies, which reported that overall SSP scores are independent of IQ outcomes (Carr et al., 2010; Jirikowic et al., 2008). However, the lack of observed relationship across the sample between auditory filtering scores and language function is more surprising but is potentially related to the aggregate nature of the data used to derive this measure. “Language” is a complex behavioral domain, and the relatively gross clinical rankings of language function in the data set (based on available clinical measures that differed depending on the examined age range) potentially lacked appropriate resolution to capture the impact of problematic, higher order auditory behaviors on language functioning, particularly in younger children. It is also possible, as suggested by our findings, that the relationship between atypical auditory filtering behaviors and language function is not apparent until children are old enough that higher order, more subtle aspects of language are expected to be mastered. For example, Thorne and colleagues (Thorne, 2017; Thorne & Coggins, 2016) have observed impaired cohesive referencing—which, in English, relies on correct use of the articles “a” and “the,” two similar sounding terms that can require subtle auditory processing to discriminate—to be highly prevalent in older children with FASDs. If, as we suggest, degraded linguistic input due to impaired sound-in-noise listening processes (reviewed in Bronkhorst, 2015) makes it challenging to learn the fundamental linguistic rules (White-Schwoch et al., 2015) involved in functional use of these two terms, atypical auditory filtering behaviors may have an impact on language functioning that is not captured by a global clinical ranking of language function in younger children but reveals itself when children are older. Prospective research that systematically explores the relationship between auditory function in FASDs and more specific measures of language function, particularly those that rely on subtle auditory processing, and particularly in older children, is needed.

### **Potential Limitations**

This study represents a preliminary step in investigating auditory dysfunction in children with FASDs using retrospective SSP auditory filtering data, clinical measures of CNS function, and caregiver-provided peripheral hearing information. These existing data served as expedient but imperfect proxies for more targeted, objective, and systematic assessments of suprathreshold listening abilities and prospective audiologic evaluations. Due to the observational nature of the data, it is possible that hearing loss in the sample was underreported and that the imprecise behavioral measures available representing sound-in-noise listening and language function obscured the observation of important relationships. Moreover, the subjective caregiver ratings on the SSP were susceptible to bias, as with any caregiver rating, although it is important to note the caregivers completed the SSP prior to their child’s FASD diagnostic evaluation, so their responses were not biased by knowledge of their child’s FASD diagnosis. Systematic, prospective investigation

**Figure 3.** Profile of caregiver ratings on Short Sensory Profile auditory filtering items across fetal alcohol spectrum disorder diagnoses (abbreviations [Astley, 2004]: fetal alcohol syndrome [FAS], partial FAS [PFAS], static encephalopathy/alcohol exposed [SE/AE], and neurobehavioral disorder/alcohol exposed [ND/AE]). The percentage of “always” or “frequently” ratings with respect to the described negative behavior was higher, across fetal alcohol spectrum disorder diagnoses, for items related to sound-in-noise listening (Items 23 and 26: black bars) than for items related to sound sensitivity/distractibility (Items 24 and 25: gray bars).



employing carefully controlled objective measures of sound-in-noise listening abilities, detailed audiologic assessment, and more targeted neuropsychological testing in children with FASDs, grouped by different age bands, is needed.

### Conclusions and Future Directions

Results from this study indicate that, although children with FAS exhibit a higher-than-normal prevalence of hearing loss, it is listening difficulties in the absence of hearing loss, likely related to suprathreshold processing deficits, that are strikingly prevalent across the spectrum of fetal alcohol disorders. Such listening difficulties are impactful—affecting life function adversely and potentially contributing to lifelong difficulties with linguistic exchange and/or social interaction—and are likely widespread, given that prenatal alcohol exposure is the leading preventable cause of birth defects and intellectual and neurodevelopmental disabilities (Williams & Smith, 2015). The present results related to listening difficulties in FASDs point to the importance of systematic clinical assessment of this domain of functioning whenever prenatal alcohol exposure is part

of the clinical profile of an individual being assessed, so that common and relevant impairment is not missed.

Although it is unclear whether listening difficulties in FASDs derive from the similar etiologies as listening difficulties observed in children with other neurodevelopmental disorders or in children presumed to be typically developing, the physiological bases of listening difficulties, in general, remain unclear (DeBonis, 2015; D. R. Moore, 2018). FASD offers a unique opportunity to explore their etiology relative to the involvement of a known and well-characterized teratogen, alcohol. The present results suggest an important role for attention-related processes in listening difficulties in children with FASDs. However, a detailed and systematic investigation of auditory dysfunction in FASDs is needed before firm conclusions are reached. It will be important to characterize auditory behavior and objectively assess suprathreshold listening abilities in FASDs, using detailed audiologic and psychoacoustic assays, along with advanced multimodal neuroimaging methods to assess the functional integrity of the auditory pathway—from periphery to cortex—in order to better parse out the physiological bases of auditory dysfunction from among the multitude of candidate auditory and cognitive processes. Although there are few evidence-

based interventions available to date to ameliorate listening difficulties related to suprathreshold deficits, enhanced understanding of the physiological underpinnings of auditory dysfunction will ultimately improve diagnoses and better inform therapy options available to these listeners.

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