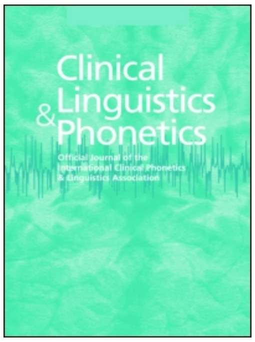

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Audiometric profiles in children with speech sound disorder: Subclinical hearing loss as a potential factor

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ABSTRACT

In the present study, hearing sensitivity in children with speech sound disorder (SSD) is scrutinized. Middle ear function (wideband tympanometry and acoustic stapedial reflexes, ASR) and inner ear function (audiometric thresholds in the conventional$_{1\text{-}8\ kHz}$ and extended$_{10\text{-}16\ kHz}$ high frequency (EHF) range, and distortion product otoacoustic emissions (DPOAEs$_{2\text{-}10\ kHz}$) were investigated. Hearing results were analyzed in relation to speech discrimination of phonemic contrasts (quiet and in noise conditions) and reproduction. Thirty-two children with SSD and 41 children with typical development (TD) ages 4–5 years participated. Children with SSD exhibited significantly less sensitive hearing compared to children with TD. This was demonstrated as more absent contralateral ASR (right ear SSD 43.7%; TD 22.0%), a higher prevalence of minimal hearing loss (MHL, > 15 dB HL at one or more frequencies or ears$_{1\text{-}8\ kHz}$ and PTA ≤ 20 dB HL, SSD 53.1%; TD 24.3%) and EHF hearing impairment (EHF HI, > 20 dB HL at one or more frequencies or ears$_{10\text{-}16\ kHz}$, SSD 31.3%; TD 24.3%). At 2 kHz bilaterally, children with SSD showed significantly higher hearing thresholds than children with TD (mean difference, left ear 3.4 dB: right ear 4.3 dB), together with a significantly lower SNR in DPOAEs at 2.2 kHz (left ear 5.1 dB mean difference between groups). In all children, audiometric thresholds at the key-frequencies for speech, 2 and 4 kHz and DPOAEs within similar spectral regions, predicted 7–12% of the variance in phonemic discrimination and reproduction. Overall, these results suggest that hearing should be more fully investigated in children with SSD.

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Speech sound disorder; key-frequencies for speech; acoustic stapedial reflexes; extended high-frequency hearing thresholds; dpoaes

Introduction

Hearing – a foundation for a listening, speaking and reading brain

For more than 100 years, hearing is considered an important contributor to language and to general cognitive development (Romey, 2013; Spearman, 1904). Early hearing experience is essential for the acquisition of speech perception fundamentals, such as prosodic and phonetic sensitivity (Kuhl, 1994, 2009; Mampe et al., 2009; Moon et al., 2013). In conjunction with socially cognitive mechanisms, which make human infants specially targeted for speech within time-limited phases (Locke, 1997), hearing lays a foundation for a listening, speaking and reading brain (Flexer, 2017). Early untreated hearing loss has negative impact on the development of the auditory cortex, essential for speech language learning (Cardon

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Supplemental data for this article can be accessed on the publisher's website.

et al., 2012; Kral & Lenarz, 2015; Sharma et al., 2002). Even minimal hearing loss (MHL ≥15 dB HL and PTA <20 dB) may cause impaired speech-in-noise hearing and reduced phonological working memory (Moore et al., 2019). Considering the large impact of hearing on speech and language acquisition, it is surprising, that there is still limited knowledge in how hearing contributes to speech sound disorders (SSD), a condition in which speech perception, phonological processing and speech production, is compromised. SSD is estimated to affect 3.4 to 6.4% of 4- to 8-year-old children, and as such, is one of the largest communicative disabilities in childhood (Beitchman et al., 1986 as cited in Eadie et al., 2015; Shriberg et al., 1999; Wren et al., 2013). In the present study, sub-clinical hearing loss, is proposed as a potential factor in children with SSD. Due to today's dichotomized classification of clinical audiology, where less than mild symptoms (subclinical hearing loss) are not detected, there's a risk of overseeing hearing factors which may restrain speech perception, phonological processing and speech production. By examining middle ear sound absorbance, acoustic stapedius reflexes (ASR), and cochlear amplification function, as DPOAEs, a fuller understanding of how hearing contributes to speech and language is accomplished. Importantly, by acknowledging hearing as an integrated system, which is communicating with the environment (for example, through noise exposure, ear infections), a fuller picture may be achieved.

Importance of sensitive and objective hearing measures

More than twenty years ago, Stackhouse and Wells (1997) found that children with SSD have pervasive speech processing problems, including reduced speech discrimination skills, imprecise storage of composition of words (phonological representations), and/or difficulties producing speech (articulation) (Nathan et al., 1998). More recently, Krueger and Storkel (2017) addressed the importance of considering the often-occurring overlap and interaction between these domains, to enable a fuller understanding of SSD, since this condition arises from deficits in multiple, interrelated systems. These researchers stressed the need of obtaining more sensitive data, which does not rely on behavioural testing alone. It is widely known that a major challenge in assessing children less than 12 years of age, is their ability to perform behavioural tests such as speech perception and audiometry (Mendel, 2008; Moore et al., 2008). Thus, it is expected that limitations in working memory and selective attention may influence children's results (DeBonis, 2015; Gathercole et al., 2004; Tamm, 1912). This could be one of the reasons, for the lack of studies investigating hearing in young children with SSD. For this reason, and to learn more about possible hearing sites of origin of SSD, a comprehensive hearing assessment with three objective physiological measures was used in the present study; wideband tympanometry – including single-frequency tympanometry at 226 Hz and 1000 Hz – acoustic stapedial reflexes (ASR) and distortion product otoacoustic emissions (DPOAEs).

Middle ear sound energy transmittance – developmental aspects and connection to DPOAEs

External and middle ear functioning is commonly evaluated with single frequency tympanometry (226 Hz and 1000 Hz), wideband tympanometry/acoustic immittance (WAI), and acoustic stapedial reflexes (ASR). Single-frequency tympanometry has been used comparably longer than WAI, due to calibration being easier for these frequencies (Hunter, 2020). The 226 Hz tympanometry has been useful for detecting middle-ear dysfunction and to study developmental changes (Roush et al., 1995). But, clear predictive relationships between tympanometric measures and conductive hearing loss (CHL) – a common pediatric condition associated with otitis media with effusion – have not been identified (Dempster & Mackenzie, 1991, as cited in Sanford et al., 2013). Therefore, since the early work on eardrum acoustic impedance (Allen, 1986), researchers have worked in improving measurements of how sound energy is transmitted through the middle ear. As a result of this work, WAI tympanometry is favoured as an objective tool in detecting middle-ear disorders and CHL. Still, the use of WAI for children with SSD is scarce.

WAI includes impedance, admittance, reflectance and absorbance, which are grouped in the catch-all term of *immittance measures* (Rosowski et al., 2013). Immittance measures are influenced by the dynamic properties of the peripheral auditory system (Kei et al., 2013). In healthy adults, largest absorbance (amount of energy transmitted into the middle-ear cavity) is between 1 and 4 kHz. It is also in this frequency range, a middle-ear infection has its largest effect (Hunter, 2020). At birth, the external and middle ear of a neonate is immature. With maturation, there is ossification of the inner two thirds of the external auditory canal, increasing its stiffness. With age, the external auditory canal length increases, its mass decreases and the middle-ear stiffness increases. This results in a decrease in resonance frequency of the ear canal (Kei et al., 2013). The physical size of the middle ear and mastoid air cell system (volume) increases from 1 to 6 years of age, thereafter reaching an average adult size. The Eustachian tube, a pressure equalizer of the middle-ear cavity, matures slowly and reaches adult functioning at 7 years of age (Kei et al., 2013). It is likely, that together, these maturational changes affect WAI measures in preschool children as in the present study. Sanford and Feeney (2008) showed that WAI morphology had a double-peaked pattern, with two maxima between 2 and 6 kHz in infants, 4, 12 and 24 weeks, in contrast to adults, who showed a single peak at around 3 kHz. It is of interest to investigate whether WAI morphology may differ in children with SSD compared to children with typical development (TD).

With higher absorbance in the middle ear cavity, the higher the likelihood of a DPOAE response (Sanford et al., 2009, as cited in Sanford et al., 2013), which is the byproduct of cochlear amplifier processing (Madell & Flexer, 2014; Sanford et al., 2013). The great frequency specificity of DPOAEs with high reliability above 1000 Hz (Shiomi et al., as cited in Bendo et al., 2015) make them an objective test often used together with pure tone audiometry. Otoacoustic emissions (OAEs) are linked to the normal functioning of the outer hair cells. They reflect mechanical responses and are considered "preneural" (Madell & Flexer, 2014).

DPOAEs are generated when two sinus tones (called primaries) are presented to the ear simultaneously (Madell & Flexer, 2014). The lower frequency primary is referred to as $f_{1,}$ and the higher frequency primary is $f_{2.}$ The byproduct (produced by outer hair cells rapid length and shape changes, and/or nonlinearity of the outer hair cells stereocilia bundle in a healthy ear) produce intermodulation distortion products. These distortion products are mathematically related to the frequencies of the primaries (Madell & Flexer, 2014). DPOAE levels change as the child develops. They are higher in newborns than in adults. Moreover, DPOAE levels are higher in children aged 1–5 years than older children and adults (Madell & Flexer, 2014).

DPOAEs are often used together with extended high frequency (EHF) audiometry to detect noise-induced hearing loss in adults (Laffoon et al., 2019; Mehrparvar et al., 2014) and to evaluate characteristic quantities of the cochlear-impaired ear (Janssen, 2013). DPOAE audiograms ("DP-grams") are able to reveal a transitory sound-conductive hearing loss because of Eustachian tube dysfunction and/or amniotic fluid in the tympanic cavity. They may also confirm a persisting cochlear hearing loss because of outer hair cell dysfunction in babies (Janssen, 2013). Reduced DPOAE responses have been observed in clinical groups of children with severe communication disorders, such as autism (Bennetto et al., 2017) and Williams syndrome (Silva et al., 2018). However, few studies have incorporated DPOAEs in their hearing evaluation of children with SSD.

Acoustic Stapedius Reflexes – sensory overload preventor and sound discrimination enhancer

The importance of the attenuation reflex, the ASR, for hearing and language will now be considered. The ASR is a response of the auditory system to high levels of sound. As the stapedius muscle contracts, it pulls on the stapes and stiffens the annular ligament in the oval window of the cochlea (Madell & Flexer, 2014). The ASR is a bilateral effect. Its contraction is a result of a cascade of events from the middle ear to the brainstem. These events involve activation of fibers in the auditory nerve and brainstem, which triggers a response from the motor nucleus of the facial nerve, to activate the facial nerve which in turn contracts the ASR. For this cascade of events to occur, each station along the way must be functional (Madell & Flexer, 2014).

The ASR increases the stiffness of the middle-ear linkage, hereby diminishing the masking effect of low-frequency sounds on high-frequency sounds (Borg & Counter, 1989). The ASR also enhances one's ability to hear soft sounds while one speaks, mainly by diminishing the influence of the comparably higher intensity of vowels to that of softer sounds, consonants. In young infants, when a contralateral ASR occurs, there is a maximal negative shift in admittance at 1000 Hz, compared with a maximal positive shift for adults. Thus, there is a frequency-dependent shift of admittance through development. Still, the time course for an adult-like ASR shift is not known (Madell & Flexer, 2014).

Borg and Counter (1989) showed that an inactive ASR, and exposure to noise, resulted in significantly worse hearing within the key-frequencies for speech (.25–4 kHz), and at 1 and 2 kHz in particular. Borg and Counter stressed that the evolution of the ASR and its associated structures (nuclei of the brainstem) effectively suppresses loud internal and external noise, allowing relevant soft sounds to be separated from irrelevant loud sounds. Neural circuits responsible for the ASR, share elements of neural pathways that control muscles of the larynx during speech. Hence, there is reason to believe that reduced function of the ASR may impact speech discrimination, both when listening to speech from others, and when hearing oneself speaking. Two Brazilian studies (Attoni & Mota, 2010; Attoni et al., 2010) found decreased or absent ASR in 5-7-year-old children with SSD. In one of the studies, a positive relationship between changes of the ASR and severity of SSD, was found (Attoni & Mota, 2010).

Key-frequencies for speech

A well-known concept in audiometric testing is the “key-frequencies for speech”, 0.25 to 4 kHz. The original work by Lidén and Fant (1954) and Fant (1960) showed that when the formants, which are the spectral peaks of the speech spectra, are plotted on an audiogram, their boundaries take a banana-shaped form. Within these boundaries, all phoneme formants are assembled. The properties of our middle ears allow most absorbance of sound energy to occur between 1–4 kHz, this is why human ears are most sensitive in this frequency range (Hunter, 2020). The importance of the key frequencies for speech is demonstrated in individuals with a so-called U-shaped audiogram, a rare condition, which eventually leads to a need for hearing amplification (Shah et al., 2005). The effects of subclinical hearing losses in the key frequencies for speech are less known. However, two recent studies on clinical populations of children with autism, have found dips within the key frequencies of speech. For example, Demopoulos and Lewine (2016) found reduced hearing thresholds at 2 kHz in children 5–18 years with autism, and Bennetto et al. (2017) found reduced otoacoustic emissions at 1 kHz in 6–17-year-old boys with autism. Demopoulos and Lewine (2016) also demonstrated a relationship between pure-tone auditory thresholds within the key-frequencies for speech, and expressive and receptive language measures. Bennetto et al. (2017) reasoned that attention to specific-frequency deficits may be important in clinical groups of children with auditory processing impairments. Still, there is limited knowledge about hearing sensitivity within the key frequencies for speech in children with SSD and how this is connected to middle-, and -inner ear functioning, speech discrimination and production.

Rational for the present study

In a recent study, phonemic discrimination (quiet and speech shaped noise) and reproduction (American Listen-Say test, Nakeva von Mentzer, 2020) and sensitive measures of hearing, i.e., a combination of behavioural and objective hearing measures, were assessed in 41 healthy children 4–5 years of age (Nakeva von Mentzer, 2020). In this sample of children, differences with respect to hearing was not expected. Nonetheless, elevated extended high frequency (EHF) thresholds were found in 24.3% of the children, and deviant DPOAEs signal-to-noise-ratios (SNRs) in 14.6%. There was a significant association between these variables. Moreover, phonemic discrimination was compromised in noise, and there was a moderate and significant correlation between phonemic discrimination in noise and EHF hearing thresholds. In light of these findings, the aim of the present study was to investigate hearing sensitivity in 4-5-year-old children with SSD and to compare their results to the children with TD. A comprehensive hearing assessment with three objective physiological measures was used; wideband tympanometry – including single-frequency tympanometry at 226 Hz and 1000 Hz – acoustic stapedial reflexes (ASR) and distortion product otoacoustic emissions (DPOAEs). Results in hearing were analyzed in relation to phonemic discrimination and reproduction. The hypothesis was that children with SSD would have reduced hearing sensitivity compared to children with TD, and in particular, in the $EHF_{10\text{-}16kHz}$ range.

Materials and methods

Participants

Seventy-three children (36 girls, 37 boys) with a mean age of 4.8 years (min = 4.1 years, max = 5.9 years) participated in the study. Forty-one children had typical speech and language development (TD, 21 girls, 20 boys), meaning they had passed developmental evaluations for speech language, motor and cognition during their early years, and had a Crystallized Cognitive composite score (Picture Vocabulary Test which assesses receptive vocabulary skills, and Oral Reading Recognition Test from the NIH toolbox, Weintraub et al., 2013) within normal limits (Nakeva von Mentzer, 2020), see Table 1. Thirty-two children (15 girls, 17 boys) had been diagnosed with SSD by a group of certified SLPs at Cincinnati Children's Hospital Medical Center, CCHMC (diagnosis code F80.0, Phonological disorder, International classification of disease, ICD 10, 2020). To ensure that these SLPs used the same diagnostic criteria the author took part of one of their monthly meetings at the start-up of the study, where the diagnostic criteria for a phonological disorder was carefully examined. Thus, the inclusion criteria for F80.0 were phonological disorder with language comprehension and non-verbal cognitive ability within normal limits. Exclusion criteria were hearing loss or impaired neurological status (ICD 10, 2020; Krueger & Storkel, 2017). Documentation in the medical records of the children showed that 4 children besides F80.0 had another diagnosis; dyspraxia of speech ($n = 3$) and slurred speech ($n = 1$).

Inclusion criteria for all children were normal hearing thresholds (≤ 20 dB HL, 1–8 kHz) and no current tympanic membrane pressure equalization (PE) tubes (grommets) at time of testing. Children with TD were recruited via postings at local community networks and children with SSD through the Division of Speech Language Pathology at Cincinnati Children's Hospital Medical Center. Children received an economic reimbursement of 50 USD for their participation. Fifty-four children were White (39 TD, 15 SSD), 15 children were African-American (1 TD, 14 SSD) and four children were biracial (1 TD, 3 SSD). The groups were not balanced with respect to race (TD; White 95.1%, African American 2.4%, Bi-racial 2.4%, SSD; White 46.8%, African American 43.8%, Bi-racial 9.4%). These proportions (SSD:TD White 2.6:1 (15:39), African American/Biracial 8.5:1 (17:2) are not representative of actual prevalence of SSD for these ethnic groups (Tomblin et al., 1997; Specific language impairment, White 7%, African American children 11%). Table 1 presents descriptive statistics for the cognitive tasks from the NIH Toolbox (Weintraub et al., 2013). Children with TD had scores within normal limits on all cognitive tasks. They outperformed children with SSD on the Picture Vocabulary Test and Oral reading. Processing speed did not differ between groups. These results verify specific difficulties

Table 1. Cognitive test performance from the NIH-toolbox (Weintraub et al., 2013) in children with TD and children with SSD.

	TD	SSD	
	Mean (SD)	Mean (SD)	*p*-value
Vocabulary	113.1 (12.5)	93.8 (17.2)	.000
Oral reading	109.6 (9.3)	101.7 (9.6)	.001
Processing speed	79.8 (16.3)	77.4 (20.2)	.59

Note, TD = typically developing, SSD = speech sound disorder with speech and language skills and the clinical diagnosis of SSD in the children with SSD in the present study. Within the group of children with SSD, there was no significant difference with respect to race on the cognitive tests (Picture Vocabulary test p =.15, Oral reading p = .11, Processing speed p = .57).

General test procedure

Participants attended one test session, approximately 2 hours in duration, in which they completed a series of audiometric, perceptual and cognitive tests, including the American Listen-Say test, with short breaks between tests and one longer snack break. Beyond audiometry and DPOAE testing, middle-ear function was assessed through wideband tympanometry, with results calculated at 226 and 1000 Hz, number of peaks counted between 0.226 kHz to 8 kHz, and acoustic stapedial reflexes (ASR).

Hardware and software

The American Listen-Say test was written in MATLAB R2016b (2016) and delivered on a laptop computer with a touchscreen (ViewSonic TD2220 55 cm). Speech stimuli were five monosyllabic CV words 'D, tea, key, see and she'. Pictures of objects to represent words were avoided to circumvent the risk of semantic misunderstandings, and discrimination was separated from reproduction. For detailed description of development of the American Listen-Say test and selection of speech stimuli, see Nakeva von Mentzer (2020). Speech stimuli were routed through an external USB-soundcard (SoundBlaster Omni, surround 5.1, Model SB 1560) and delivered through lightweight Sennheiser HD 25–1 on-ear headphones. Calibration was completed with a sound level meter (Larson Davis Model 84; B&K microphone and artificial ear) to set the default, long-term sound level at 65 dBA.

The American Listen-Say test – speech discrimination and reproduction

Speech discrimination

In the speech discrimination task of the American Listen-Say test a word stimulus is auditorily presented (X) followed by two further word stimuli (A and B), see Figure 1(a). The child identifies which of A or B matches the initial stimulus X. To keep the child's attention, each auditory presentation was accompanied with a cartoon picture of a woman's face; a large face at the top of the screen (X) that represents the initial stimulus, and two smaller faces at the bottom of the screen (A and B), that represent the alternative response stimuli (for more details, see Nakeva von Mentzer, 2020). A visual timeline with squares coloured in blue, green, red and yellow, was placed in front of the child. Children were instructed that the face background on the screen would change colour in order of the visual timeline, with the yellow colour presented last. All possible combinations of word (x5 per colour) and position (x2, A or B) were presented in two overall repetitions, generating a maximum correct score of 20. The order of correct stimuli (A or B) for each participant was randomized from trial to trial. Words followed a carrier phrase "Listen to". Children's response accuracy and reaction times were recorded in MATLAB (2016). Only accuracy scores are presented in the present study. Children listened to the CV-tokens in two conditions; quiet and in speech shaped noise.

For the noise condition, the individual words were perceptually homogenized with respect to speech reception thresholds (SRT) in noise following the procedure of Leensen et al. (2011), Sheikh Rashid et al. (2017), and Vlaming et al. (2014). The SRT chosen for the study was the SNR at 90% speech intelligibility of 10 normally hearing adults (≤ 20 dB HL; 1–8 kHz; 1 male; Mean age 30 yrs, Min = 24, Max = 59 yrs). For detailed test procedures, see Nakeva von Mentzer (2020).

Speech reproduction

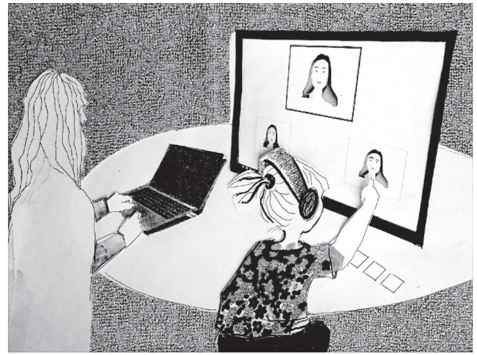
The reproduction task of the American Listen-Say test presented the same five words (D, tea, key, see and she) auditorily in word pairs, see Figure 1(b). Each possible word pair was presented once, generating a maximum score of 10, thus each correctly reproduced word pair generated a score of 1. The stimulus order was randomized for each participant. The reproduction task was explained to the children with the following instruction: "You're going to listen to two words. First, two faces on the screen will say the words, for example, 'toe-so'. Second, a microphone will appear on the screen. When you see the microphone, you repeat the words back in this microphone" (instructor pointing to the table-stand microphone). After successful completion of two initial trials off-line where the child correctly repeated two consecutively presented words, a computer-delivered trial was presented. Children's voice responses were typed by the researcher and scored as correct or incorrect in MATLAB (2016). Voice responses were simultaneously recorded in Audacity for later off-line verification of reproduction accuracy. After each testing, the recording was checked with the researcher's typing. Any discrepancies were corrected. All children were offered to listen to the recording of their own voice as a reward after completing the game. Children's response accuracy was recorded in MATLAB (2016).

Reliability of the American Listen-Say test was certified in the following ways; 1. the first author and test developer who is an SLP by training, tested all children, 2. test duration was shorter (7–8 min per condition) compared to an earlier Swedish version of the test (Nakeva von Mentzer et al., 2017), diminishing the influence of attention abilities (Mendel, 2008), 3. the same set up was used for all children (laptop, headphones, microphone, visual timeline). To examine construct validity, Spearman correlation analysis (N = 73) was performed. The results showed significant correlations with a medium effect between the three conditions (quiet-noise, rs, = .37, p = .001, quiet-reproduction, rs = .33, p = .004, noise-reproduction rs = .32, p = .33), suggesting they assess a similar underlying construct. For further considerations regarding selection of test stimuli, see section 2.2 in Nakeva von Mentzer (2020). Figure 1(a,b) shows the setup of the American Listen-Say test.

Middle-ear measures

Normal middle-ear function was defined as a static acoustic admittance within the normal range (Study Protocol, 2016, see Supplement) and presence of an ipsilateral acoustic reflex at or below 100 dB HL. Seven children with TD and 2 children with SSD had a history of pressure equalization (PE)-tubes. Thus, in the present sample of children 12.3% had an history of middle-ear infection which was treated with PE-tubes. This number represents comparable prevalence of PE-tubes treatment that has been reported by the Swedish Agency a.

"You're going to listen to three words. First, the big face on the top of the screen says one word, for example 'cat'. Second. two small faces at the bottom of the screen say one word each. for example. 'cat' and 'hat'. ***You'll touch one of the small bottom faces that says the same as the big top face".***



b.

"You're going to listen to two words. First, two faces on the screen will say the words, for example 'toe-so' (1). Second, a microphone will appear on the screen (2). ***When you see the microphone, you repeat the words back in this microphone****" (3) (instructor pointing to the table-stand microphone).*

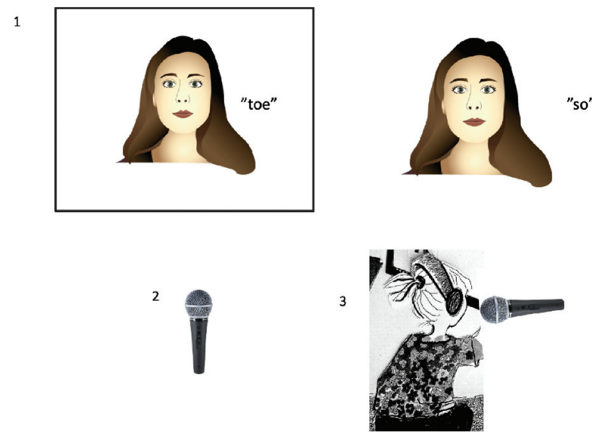


Figure 1. A.*"You're going to listen to three words. First, the big face on the top of the screen says one word, for example, 'cat'. Second. two small faces at the bottom of the screen say one word each. for example. 'cat' and 'hat'.* ***You'll touch one of the small bottom faces that says the same as the big top face".***
B.*"You're going to listen to two words. First, two faces on the screen will say the words, for example, 'toe-so' (1). Second, a microphone will appear on the screen (2).* ***When you see the microphone, you repeat the words back in this microphone****" (3) (instructor pointing to the table-stand microphone).*
Figure 1(a,b). Experimental setting for the Listen-Say test phonemic discrimination and reproduction task

for Health Technology Assessment and Assessment of Social Services (SBU, Swedish Agency for Health Technology Assessment and Assessment of Social Services, 2008).

Otoscopy

Otoscopy was completed to ensure that the ear canal was clear enough to allow the insertion of an admittance probe tip of appropriate size, free from excessive cerumen, and free from pressure equalization (PE)-tubes (Madell & Flexer, 2014). It was also ensured that the tympanic membrane looked healthy (normally set, pale but slightly yellowish, relatively transparent with fine reflex in front lower quadrant (SBU, Swedish Agency for Health Technology Assessment and Assessment of Social Services, 2015). In children with excessive cerumen, a clinical audiologist, was consulted in removing it. Only children with a free ear canal continued with the testing.

Tympanometry

Wideband tympanometry (click stimulus) with individual extraction of tympanograms at 226 Hz and 1000 Hz were completed with an Interacoustics Titan immittance system (Middelfart, Denmark) to investigate middle-ear function. Normal tympanometry criteria at 226 Hz were the following: equivalent volume (ml) 0.3–1.5 cc, peak pressure −150-50 daPa, static acoustic admittance, 0.3–1.5 mmho (Study Protocol, 2016, see Supplement) and tympanogram width (daPa) 50–250 daPa (Hunter, 2020). Absorbance graph values (0.226 to 8 kHz) at peak pressure were inspected (Titan, 2019), and number of peaks were counted. Children in whom it was not possible to obtain a seal despite consulting the clinical audiologist at the lab or had flat tympanograms, were excluded from the study.

Acoustic stapedial reflexes (ASR)

Acoustic stapedial reflexes (ASR) thresholds were measured immediately after tympanometry with the ear-canal pressure maintained at tympanometric peak pressure to maximize the possibility of obtaining a response. A broadband noise stimulus from 60 to 100 dB HL was delivered in 5 dB increments. The ASR threshold was defined as the lowest intensity at which change in admittance of 0.02 mmho was detected (Kei & Zhao, 2011, chapter 4).).

Inner ear measures

Audiometric hearing thresholds

Pure tone air conduction thresholds were obtained for each ear separately at 1.0, 2.0, 4.0, 8.0, 10.0, 12.5 and 16.0 kHz using an Interacoustics Equinox Audiometer (Middelfart, Denmark) and Sennheiser HDA 300 circumaural earphones in a sound-attenuating booth. Since EHF hearing thresholds were also being tested, 0.25 and 0.5 kHz were not included to avoid an excessively long test for these young children (Mendel, 2008). Also, wideband tympanometry was used to evaluate middle ear function at low frequencies.

For conventional and EHF audiometry, the headphones were positioned by the examiner and the child was instructed to put a colored peg on a board whenever he/she heard the auditory stimulus, thus condition play audiometry. Participants were familiarized with the task prior to measuring thresholds. The pure tone air-conduction threshold was determined using the Hughson-Westlake bracketing technique (Jerger, 2018), starting at 30 dB and descending in 10 dB intervals until the point where the child no longer responded to the sound. Starting at that intensity, the ascending technique was used at 5 dB intervals until the child responded. The hearing threshold was established when the participant was able to correctly identify at least two out of three tone presentations. Reliability check at 1 kHz was done in all participants. The criterion of minimal hearing loss (MHL) was one or more thresholds > 15 dB $HL_{1\text{-}8\ kHz}$ in at least one ear. The criterion of EHF hearing impairment, HI was > 20 dB HL at one or more frequencies or $ears_{10–16\ kHz}$.

DPOAEs

Outer hair cell activity was measured using distortion product acoustic emissions, DPOAEs with the Interacoustics Titan system (Middelfart, Denmark) with the 440 module. Recording parameters included primary tone stimulus levels of 65 dB SPL (L1) and 55 dB SPL (L2) with an f_2/f_1 frequency ratio of 1.22. DPOAEs were measured at ambient pressure with the DPOAE response reliability set at 98% and a 7-dB level tolerance. DPOAE test frequencies were measured in loops with a maximum test time of 90 seconds where each DPOAE test frequency was measured for approximately 3 seconds before continuing to the next one. DPOAE signal level and noise level were measured at $2f_1$-f_2 in descending order at ten f_2 frequencies from 2–10 kHz including (10.0, 9.1, 8.3, 7.5, 6.2, 5.1, 3.9, 3.2, 2.7 and 2.2 kHz). The signal to noise ratio (SNR) was calculated by subtracting the mean DPOAE noise level from the mean DPOAE signal level at each f_2 test frequency. A cut off criterion of ≥ 6 dB was used for normal SNRs (Konrad-Martin et al., 2017).

Statistical analysis

Data were analyzed statistically using IBM SPSS (2013) version 26. The Mann-Whitney U-test was used for between-group comparisons. Corrections for multiple comparisons were made with the Bonferroni test. Fischer's exact test was used for comparisons of categorical data. A composite score of the American Listen-Say test was calculated as an average score of the speech discrimination tasks (quiet and noise) and reproduction task. A multiple linear regression analysis was performed with The American Listen-Say test as dependent variable (quiet, noise and reproduction) and hearing measures as independent variables. Corrected significance values were p = .004 for audiometric thresholds and p = .005 for DPOAEs.

Results

Middle ear measurements

Overall, normal middle ear function was defined as static admittance within the normal range (Study Protocol, 2016, see Supplement) and presence of ipsilateral reflex. Normal middle ear functioning was observed in 62 children on the left ear (TD n = 35, 85.4%; SSD n = 27, 84.4%) and in 55 children on the right ear (TD n = 33, 80.5%, SSD n = 22, 68.8%). The slightly higher number of normal middle ear responses in children with TD compared to children with SSD did not represent a statistically significant difference.

Tympanometry

Table 2 displays the results for children with TD and children with SSD. For static admittance, left and right ear mean values varied between .45 and .57 mmhos (min = .2, max = 1.9) in the children. There was no significant difference between the groups, U = 487.0, $z = -.51.8$, $p = .60$. Deviant static admittance results at 226 Hz were observed in eleven children unilaterally (TD $n = 8$; SSD $n = 3$). No child had deviant results bilaterally. Results at 1 kHz were then analyzed. Left and right ear mean values varied between 1.7 and 2.0 mmhos (min = .2, max = 7.4). No significant differences between the groups were observed, $U = 406.5$, $z = -.96.3$, $p = .60$. Since norms for preschool children are lacking, analysis of deviant values was not conducted.

Number of peaks between 0.226 kHz to 8 kHz were counted. One or two peaks were observed in 60 children on the left ear (TD $n = 35$, 85.4%; SSD $n = 25$, 78.1%) and in 56 children on the right ear (TD $n = 36$, 87.8%; SSD $n = 20$, 62.5%). Three to four peaks were observed in 13 children on the left ear (TD $n = 6$, 14.6%; SSD $n = 7$, 21.9%). Corresponding number of peaks for the right ear were observed in 15 children (TD $n = 5$, 12.2%; SSD $n = 10$, 31.3%, two missing values). The mode score for both groups was 2, and so was the median score. The slightly higher number of peaks in children with SSD did not constitute a significant difference.

Acoustic stapedial reflexes (ASR)

Table 3 displays ASR in children with TD and children with SSD. The analysis showed that there was a 10.2% difference between the groups with respect to overall present ipsilateral ASR, children with TD having more present reflexes (TD = 91.4%, SSD = 81.2%). Corresponding difference for overall contralateral ASR was 9.6%, again children with TD having more present reflexes than children with SSD (TD = 76.8%, SSD = 67.2%). Neither of these differences were statistically significant. However, when ears were inspected separately, children with SSD showed a significantly higher number of absent contralateral ASR than children with TD on the right ear (present reflexes TD, 78.0%; SSD, 56.3%, X^2 (1) = 4.0, $p = .047$). The mean level of each ASR and ear did not differ between the groups.

Absent tympanometric responses and/or absent ASR

In 5 children with TD there were absent tympanometric responses on the right ear at 226 Hz or at 1000 Hz. In two of these children absent responses coincided with absent ASR ipsilaterally and contralaterally. Correspondent analysis revealed that 7 children with SSD had absent tympanometric responses at 226 Hz or at 1000 Hz. Two of these children had absent responses bilaterally at 1000 Hz, and one child had absent responses bilaterally at both 226 and 1000 Hz. In 5 of the children either one or both ASRs were absent on either ear or side.

Inner ear measurements

Audiometric thresholds

Mean audiometric thresholds for each ear for the two groups are listed at each tested frequency in Table 4. Between group comparisons corrected for multiple comparisons ($p = .004$) revealed a significantly higher threshold in SSD children at 2 kHz bilaterally. Left ear ($U = 410.0$, $z = -2.8$, $p = .004$, $r = .34$), right ear ($U = 386.5$, $z = -3.09$, $p = .002$, $r = .36$). Mean difference between ears

Table 2. Tympanometric results in children with TD and children with SSD.

	TD		SSD	
	Left Ear	Right Ear	Left Ear	Right Ear
226 Hz	Mean (range)	Mean (range)	Mean (range)	Mean (range)
Equivalent Volume[1]	1.1 (.8 – 1.4)	1.09 (.9 – 1.3) [b = 2, c = 2]	1.1 (.9 – 1.5) [b = 2, c = 3]	1.1 (.6 – 1.5) [c = 3]
Peak Pressure (daPa)	–.36.4 (–261 – 15) [a = 4]	–36.1 (–258 – 80) [a = 5, b = 2]	–37.3 (–242 – 46) [a = 1, b = 3]	–40.8 (–286 – 74) [a = 5, b = 3]
Static Admittance (mmho)	.57 (.2 – 1.9) [a = 4]	.53 (.2 – 1.6) [a = 4, b = 2]	.45 (.2 – .8) [a = 1, b = 3]	.47 (.2 – 1.1) [a = 2, b = 3]
Tympanometric Width (daPa)	115.4 (64 – 246) [a = 1]	117.0 (70 – 210) [a = 1, b = 2]	119.2 (83 – 166) [a = 1, b = 2]	124.5 (75 – 202) [a = 2, b = 4]
1000 Hz	Mean (range)	Mean (range)	Mean (range)	Mean (range)
Peak Pressure (daPa)	–20.1 (–305 – 42)	–23.1 (–258 – 70) [b = 3]	–14.0 (–242 – 58) [b = 5]	–26.6 (–351 – 94) [b = 5]
Static Admittance (mmho)	1.8 (.2 – 5.1)	1.7 (.4 – 7.4) [b = 3]	1.8 (.3 – 5.1) [b = 5]	2.0 (.5 – 6.0) [b = 5]
Tympanometric Width (daPa)	102.8 (51 – 386)	96.4 (64 – 198) [b = 3]	92.8 (57 – 132) [b = 5]	97.2 (55 – 181) [b = 5]

Note, TD = typical development, SSD = speech sound disorder, [1] = same values obtained for 1 kHz, [a] = out of normal range, [b] = missing responses, [c] = missing responses for 1 kHz

Table 3. Acoustic stapedial reflexes in children with TD (n = 41) and children with SSD (n = 32). N shows number of present reflexes between 60–100 dB SPL. Mean shows threshold in dB SPL of present reflexes.

Children with present acoustic stapedial reflexes				
	TD		SSD (n = 32)	
IPSILATERAL	*n*	Mean level (SD, Min – Max)	*n*	Mean level (SD, Min – Max)
Left (count, %)	39; 95.1	80.6 (11.2, 60– 100)	28; 87.5	82.0 (8.8, 65– 100)
Right (count, %)	36; 87.8	82.4 (10.1, 70– 100)	24; 75.0	85.2 (9.3, 60– 95)
Percent ears	91.4		81.2	
Percent total ears	**87.0%**			
CONTRALATERAL				
Left (count, %)	31; 75.6	87.6 (8.4, 70– 100)	25; 78.1	89.0 (7.6, 70– 100)
Right (count, %)	32; 78.0	86.6 (9.3, 70– 100)	18; 56.3	89.4 (8.7, 65– 100)
Percent ears	76.8		67.2	
Percent total ears	**72.6%**			

Note, TD = typical development, SSD = speech sound disorder

was 3.4 dB (left ear) and 4.3 dB (right ear). This difference corresponds to a doubling of sound energy, i.e. children with SSD needed a doubling of sound energy to hear the tone compared to children with TD.

The analysis of MHL of the whole sample of children (thresholds > 15 dB HL at one or more frequencies or $ears_{1\text{-}8\ kHz}$) showed that 10.6% (62/584) of the thresholds were elevated (TD 21/328 thresholds 6.4%; distributed among 10 children, SSD 41/258, 16.0%, distributed among 17 children). A Chi square test of independence revealed that there was a significant difference between the groups. More thresholds were elevated (> 15 $dB_{1\text{-}8\ kHz}$), in children with SSD than in children with TD, (X^2 (1) = 14.0, p = .0003). In sum, 10 children with TD (24.3%) had either unilateral MHL (n = 6) or bilateral MHL (n = 4). Correspondent numbers were 17 children with SSD (53.1%), unilateral MHL (n = 9) or bilateral MHL (n = 8).

The analysis of EHF thresholds of the whole sample of children (left and right $ear_{10\text{-}16\ kHz}$, TD and SSD) showed that 11.7% (50/426) were elevated (> 20 dB HL, TD 21/240 thresholds, 8.8% distributed among 10 children, 24.3%; SSD 29/186 thresholds, 15.6% distributed among 10 children, 31.3%). A Chi square test of independence revealed that there was a significant difference between the groups. Children with SSD having more

Table 4. Hearing thresholds (mean and standard deviations, left and right) in children with TD (n = 41) and children with SSD (n = 32).

	TD			SSD		
	Left Ear	Right Ear		Left Ear	Right Ear	
Freq.	Mean (SD)	Mean (SD)	Left ear comparisons	Mean (SD)	Mean (SD)	Right ear comparisons
kHz	dB HL	dB HL	p-values	dB HL	dB HL	p-values
1	11.1 (4.8)	11.3 (4.6)	.04	13.4 (5.0)	12.0 (5.9)	.43
2	10.4 (4.8)	7.6 (5.5)	.004	13.8 (4.4)	11.9 (5.8)	.002
4	8.1 (5.9)	8.1 (6.1)	.31	9.7 (6.7)	9.5 (6.4)	.33
8	5.1 (7.5)	6.5 (7.8)	.32	3.9 (7.4)	3.8 (8.6)	.15
10[a, b]	11.2 (11.8)	10.1 (9.8)	.84	11.8 (13.1)	10.8 (9.2)	.65
12.5[b]	5.5 (10.3)	7.7 (9.2)	1.0	7.7 (14.7)	13.9 (15.4)	.10
16[b]	1.7 (11.5)	2.0 (11.0)	.48	2.1 (16.1)	4.4 (14.2)	.53

Note, TD = typical development, SSD = speech sound disorder, [a] n = 38 in children with TD, [b] n = 31 in children with SSD. Significance value after correction for multiple comparisons was .004.

elevated EHF thresholds, (X^2 (1) = 4.5, p = .033). Four TD children (3 uni- and 1 bilaterally) and 6 children with SSD (3 uni- and 3 bilaterally) met the strict criterion of EHF HI (> 20 dB average EHF on at least one ear). When comparing groups with respect to number of children meeting this strict criterion, no significant difference was found. When inspecting asymmetric HI ≥ 15 dB difference between ears, an equal number of children in each group, 3 TD and 3 SSD children, fulfilled this criterion. Figure 2 displays EHF hearing thresholds with confidence intervals in the TD and SSD group. Additionally, Figure 2 shows, that at 12.5 kHz right ear, children with SSD had a significantly higher hearing threshold.

DPOAE

Complete data were derived from 76 ears in the TD group (left ear, n = 39, right ear, n = 37). Corresponding number in the SSD group was 64 ears, thus, in this group complete data were derived from all children. Figure 3 displays SNR mean levels in TD children and children with SSD. Group comparisons showed overall comparable SNRs except at 2.2 kHz left ear, where children with SSD exhibited significantly lower SNRs than children with TD (SSD, M = 19.5 dB, SD = 8.4/Mdn = 21.2 dB, min = 5.7, max = 34.7, TD, M = 24.6 dB, SD = 5.7/Mdn = 25.3 dB, min = 9.2, max = 34.3, U = 380.5, z = −.2.8, p = .005, r = .34), mean group difference between was 5.1 dB, see Figure 3 for overall comparisons and 95% confidence intervals.

Eight children with SSD had low DPOAE SNRs (< 6 dB SNR, Konrad-Martin et al., 2017). Two of these children had one or more elevated EHF hearing thresholds and six children had EHF hearing thresholds within the normal range (≤ 20 dB HL). Fischer's exact test did not reveal any significant interaction between these variables. The association between elevated EHF hearing thresholds and low DPOAE SNRs observed in the normalization study (Nakeva von Mentzer, 2020) in TD children was not confirmed in children with SSD.

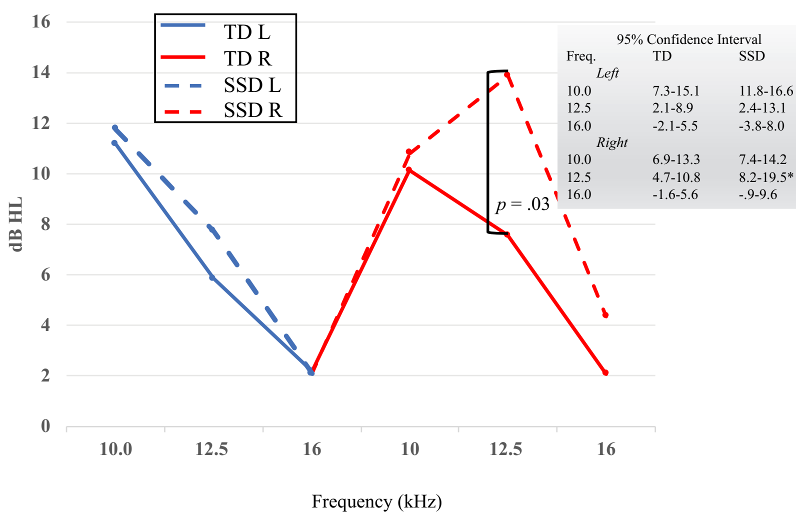
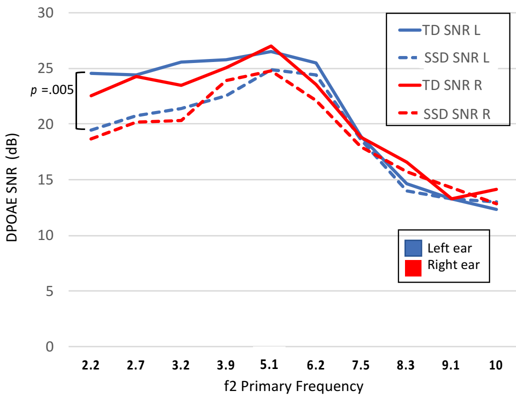


Figure 2. Extended high frequency thresholds in children with TD (continuous lines) and children with SSD (crosshatched lines).



Confidence Intervals (Left, Right)				
Freq.	TD	SSD	TD	SSD
2.2	22.8-26.7	16.4-22.5*	20.1-24.9	15.0-22.3
2.7	22.2-26.9	17.9-23.6	21.6-26.1	17.4-23.0
3.2	24.2-28.1	17.7-25.0	20.8-25.9	17.3-23.4
3.9	23.9-28.7	19.3-25.7	22.1-28.1	21.3-26.4
5.1	24.3-30.3	22.3-27.5	23.6-29.3	22.0-27.6
6.2	22.8-28.5	20.9-27.9	19.7-26.5	18.6-25.5
7.5	16.4-21.0	15.5-21.7	15.9-21.5	14.2-21.7
8.3	12.2-16.8	10.3-17.7	13.7-19.1	12.4-19.1
9.1	11.9-15.2	10.1-16.4	10.1-15.9	10.4-18.2
10.0	10.4-14.4	10.1-15.9	11.6-16.3	9.9-15.8

Figure 3. DPOAE mean dB SNR in children with TD and children with SSD.

Phonemic discrimination and reproduction

Children with TD had overall higher performance than children with SSD on the composite score of the Listen-Say test (TD M = 75.3% SD = 10.9, SSD M = 56.9% SD = 16.2, p = .000, r = .57). Separate comparisons (discrimination; quiet and noise, reproduction) revealed that there were overall higher scores in children with TD. Only the reproduction score reached a statistically significant difference between the groups, see Figure 4 and Table 5. Thus, children with SSD showed comparably more difficulty reproducing CV-tokens than discriminating them.

Table 5 also displays comparisons in relation to children having elevated EHF hearing thresholds (> 20 dB HL 10–16 kHz). In sum, overall lowest scores were found in the ten children with SSD with elevated EHF hearing thresholds. Within group comparisons (SSD ≤ 20 dB n = 22; SSD > 20 dB n = 10) showed no statistically significant difference (composite score, p = .34, quiet, p = .06, noise, p = .41, reproduction p = .06).

Investigating relations between hearing measures and phonemic discrimination and reproduction

Before performing the linear regression analysis, all variables were examined to check if there were significant correlations between the variables. When significant correlations were observed among variables within the same domain, those variables were summarized to create a sum score. To avoid multicollinearity, right and left ear were analyzed separately. In sum, seven summed scores were created for each ear. These were: Tymp_comp_226, Tymp_comp_1000, Audiom_ lowfreq, Audiom_ highfreq, DPOAEs_SNR lowfreq; DPOAEs_SNR middlefreq and DPOAEs_SNR highfreq. The left ipsilateral ASR was kept as single variable since making a composite score reduced the strength of the correlation.

Table 6 displays the results of the linear regression analyses with the American Listen-Say test as outcome variable and hearing measures as predictors for left and right ear respectively.

The American Listen-Say test was weakly and significantly correlated with Audiom_lowfreq and DPOAE_SNR lowfreq (left ear, phonemic reproduction; right ear, all three conditions). The quiet condition left ear showed only one significant

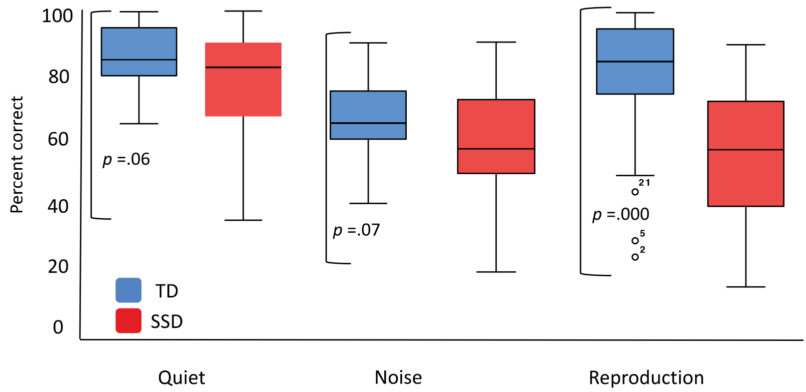


Figure 4. Phonemic discrimination (quiet and noise) and reproduction in children with typical development (TD) and children with speech sound disorder (SSD).

Table 5. Phonemic discrimination and reproduction in children with TD and children with SSD, and children categorized into EHF ≤ 20 dB HL and EHF >20 dB HL.

	TD		SSD		p-value	
	Mean, Mdn (SD, range)		Mean, Mdn (SD, range)			
Composite score[1]	75.3, 76.7 (10.9, 43–93)		56.9, 55.8 (16.2, 18–92)		.00	
Quiet	85.7, 85.0 (10.0, 65–100)		76.6, 82.5 (17.8, 35–100)		.06	
Noise	66.3, 65.0 (13.0, 40–90)		59.4, 57.5 (17.4, 20–90)		.07	
Difference score[2]	19.4, 20.0 (12.8, −10-50)		17.2, 20.0 (19.3, −25-70)		.56	
Reproduction	81.2, 85.0 (21.0, 25–100)		56.1, 57.5 (21.0, 15–90)		.00	
EHF hearing	EHF ≤ 20 dB (*n* = 31)	EHF >20 dB (*n* = 10)	EHF ≤ 20 dB (*n* = 22)	EHF >20 dB (*n* = 10)	TD	SSD
Composite score	76.1, 78.3 (11.0, 43–93)	73.0, 75.8 (10.0, 60–85)	59.3, 55.8 (15.9, 33–92)	51.5, 54.2 (16.1, 18–68)	.31	.34
Quiet	85.3, 85.0 (10.3, 65–100)	87.0, 87.5 (11.1, 70–100)	80.0, 85.0 (16.1, 45–100	69.0, 75.0 (19.8, 35–100)	.069	.058
Noise	68.1, 65.0 (13.1, 40–90)	61.0, 60.0 (11.9, 40–85)	61.1, 60.0 (18.0, 20–70)	55.5, 55.0 (16.0, 20–80)	.11	.41
Reproduction	80.8, 85.0 (20.4, 25–100)	82.5, 85.0 (13.0, 60–100)	57.5, 60.0 (22.4, 15–90)	53.0, 50.0 (18.3, 20–75)	.80	.059

Note, TD = typical development, SSD = speech sound disorder, EHF = extended high frequency. [1] Composite score = average score of the three tasks of the American Listen-Say test, [2]Difference score = quiet accuracy score subtracted by noise accuracy score

correlation, which was with DPOAE_SNR lowfreq. Finally, on the left ear, the noise condition was weakly and significantly correlated with left ipsilateral ASR and DPOAE_SNR lowfreq.

The model with two predictors Audiom_lowfreq and DPOAE_SNR lowfreq, was significant for the quiet and noise condition on the right ear, explaining 8 and 7% of the variance respectively. For the quiet condition left ear, the simple regression analysis showed that DPOAE_SNR low explained 10% of the variance. For the noise condition left ear, the model was not significant. Lastly, for phonemic reproduction right ear, only Audiom_lowfreq was significant, explaining 11% of the variance. On the left ear, only DPOAE_SNR lowfreq was significant, explaining 12% of the variance.

In sum, the analyses for all children (TD and SSD combined) showed these inner ear hearing measures in the low-frequency range, explained 7 to 12% of the variance in the right and left ear models. Overall, the present study´s regression models showed a generally weak relationship between hearing measures and phonemic discrimination and reproduction in the children.

Table 6. Results of the linear regression analyses with the American Listen-Say test as outcome variable.

Outcome variable	Ear	N	Model Adjusted R^2	F	*p*-value for model	Explanatory Variable (Standardized Beta Coefficient)	
						Audiom_low	DPOAE_SNR low
Quiet	L[1]	71	.10	8.4	.005		.33*
	R	71	.08	4.1	.02	-.14*	.25*
Noise	L[2]	65	n.s.	n.s.	.06	n.s.	n.s.
	R	71	.07	3.3	.04	-.17*	.20*
Reproduction	L	71	.12	5.8	.005	−.21	.28*
	R	71	.11	5.4	.007	-.25*	.20

Note, [1] = simple regression, only DPOAE_SNR low served as predictor. [2] = ipsilateral acoustic reflex served as second predictor, * = p <.05

Discussion

Unbalanced ethnic distribution

The aim of this study was to examine hearing in 4-5-year-old children with a diagnosed speech sound disorder (SSD) in comparison with children with typical development (TD). Hearing results were analyzed in relation to phonemic discrimination and reproduction of the American Listen-Say test (Nakeva von Mentzer, 2020). First, the over-representation of African-American children in the SSD group will be acknowledged. The causes for this, are probably multifaceted. One aspect is that the city of Cincinnati, where the data collection was made, has the 12th largest population of African Americans in the US (42.7% compared to 13.4% in the US, Indexmundi, 2020) which could explain the relatively higher number of African American families signing up for the study. Another aspect is the health care situation in the US, where families need health care insurance to receive care (Children's hospitals, 2020). By participating in this study, children were economically reimbursed and received an hearing evaluation, which could have increased some families' willingness in participating. All participating children with SSD had been diagnosed by a clinical SLP at CCHMC. Due to the relatively higher proportion of African Americans in Cincinnati compared to the United States at large, we should expect a higher awareness about the African American dialect. Thus, it should not be the case that dialectal differences should result in a SSD diagnosis. With culturally fair language measurements (Washington & Craig, 2004), there is little reason to believe children were not accurately diagnosed. Also, the scores of the NIH-toolbox (Weintraub et al., 2013) in the present study, confirmed their clinical diagnoses.

Middle ear measurements

WAI results

Middle ear status was within normal limits for the majority of children. Only a slight number of children had deviant peak pressure, suggesting these children were recovering from an upper respiratory tract infection. The cause is commonly Eustachian tube dysfunction creating negative middle ear pressure (Revai et al., 2008). Measurements with respect to single frequency tympanograms at 226 Hz and 1000 Hz, showed no difference between the groups. Static admittance values at 226 Hz varied between .45 mmhos and .57 mmhos. These values are comparable to those for healthy children 3 to 5 years of age (Shanks et al., 1988). The mean static admittance values obtained at 1000 Hz varied between 1.7 and 2.0 mmhos (min = .2, max = 7.4). Low cutoff norms (compensated admittance magnitude values) for newborns are provided by Margolis et al. (2003) and Kei et al. (2003) (as cited in Madell & Flexer, 2014, p. 127). Margolis et al. suggested .60 mmho (5th percentile for peak-to-negative-tale compensated admittance), and Kei et al. suggested .39 mmho (5th precentile for peak-to-positive-tail compensated admittance). As age is a significant factor in 1000 Hz tympanometry, admittance values increasing with age (Kei & Zhao, 2011), the conclusions drawn from the present study, is that a certain number of children in both groups show deviant admittance values. But overall, mean admittance values at 1000 Hz in the present study, resemble the mean values presented for young infants by Kei and Zhao (2011, Table 1–3, see authors discussion on differences connected to different instrumentation and different test protocols), but with higher upper limit values (Kei & Zhao, 2011, 1.7–3.1 mmhos, this study 5.1–7.4 mmhos). Shanks et al. (1988) report peak compensated static admittance values for adults at 678 Hz (90% normal range) which varied between 1.06 and 6.31 mmhos. These upper limit values resemble the values obtained at 1000 Hz in the present study. The immittance of the ear is frequency dependent. Results of the present study clearly show that at higher frequencies, more sound energy is transmitted into the middle ear cavity compared to at lower frequencies. Since norms for static admittance values at 1000 Hz are still lacking for preschool children, the present values may be used (Calandruccio et al., 2006; Kei & Zhao, 2011).

WAI morphology showed a slightly higher number of peaks in children with SSD compared to children with TD, but these values did not reach statistical significance. Sanford and Feeney (2008) found a double-peaked pattern in young infants between 2 and 6 kHz, as compared to a single peak in adults at 3 kHz. Thus, as the ear canal grows longer, the ear channel resonance frequency decreases, as does number of peaks. If, number of peaks is related to maturation of the peripheral auditory system, a higher number of peaks could be a sign of a less mature system. However, the present study could not confirm any such results.

Acoustic stapedial reflexes

More children with SSD than children with TD had absent contralateral acoustic stapedius reflexes (ASR) on the right ear. This result indicates that in children with SSD, the middle-ear system may less effectively allow relevant soft sounds to be separated from irrelevant loud sounds, contributing to less precise speech discrimination abilities (Borg & Counter, 1989). Evidence from children with SSD having reduced or absent ASRs is scarce. However, there are two Brazilian studies investigating this. Attoni et al. (2010) measured ASR between .5 and 4 kHz (cut-off range at 90 dB HL, this study 100 dB HL) in children with TD and children with SSD. Higher ASR thresholds and/or more absent reflexes, were found in children with SSD. Attoni et al. (2010) reported that the phonological system was compromised if ASR was absent. Furthermore, voiced consonant phonemes – requiring vocal fold vibration – were affected in children who had most impaired phonological systems and compromised ASR. In the other study, Attoni and Mota (2010) studied the contralateral ASR in children with SSD and found deviant results in all children. In their study, no relationship was found between the severity of the SSD and changes in the ASR. In an upcoming study, qualitative and quantitative phonological analyses will be conducted (Nakeva von Mentzer, ongoing) to understand if certain phonological processes, – as the voicing distinction – is relatively more affected, and if impaired phonological reproduction, is correlated with absent ASRs, in accordance with the findings of Attoni et al. (2010).

Conditions of absent contralateral reflexes have been observed in other clinical populations with language disorders, for example, children with Williams syndrome (Attias et al., 2008; Silva et al., 2018). In the study by Attias et al. (2008) the middle olivo-cochlear (MOC) efferent system was tested together with ASR. Absent ASRs in 62–86% of the participants and hyperexcitability of the MOC was reported. Attias et al. reasoned that hyperexcitability of the MOC efferent system, coupled with absence of ASR, may contribute to the often observed hyperacusis in Williams syndrome. In the review by Silva et al. (2018), associations between absent ASR and complaint of hyperacusis was reported. In their review, mild to moderate sensorineural hearing loss mainly in the high-frequency range, was common, together with absence of OAE and acoustic reflex. Few studies have investigated hyperacusis in children with SSD and in the present study, families were not asked about whether their child showed any such symptoms.

Pure tone audiometry within the conventional and EHF range

Children with SSD exhibited less sensitive hearing thresholds than children with TD, both evident as a larger presence of MHL, and a higher number of elevated EHF hearing thresholds. Thus, the auditory insensitivity, as proposed by Tamm (1912), was confirmed in the present study. Moore et al. (2019) investigated auditory perception, cognition and communication in children with MHL (≥ 15 dB HL and < PTA 20 dB HL)) relative to children with normal hearing. Speech-in-noise hearing and phonological working memory were impaired in children with MHL, the latter comparably more in children with minimal *symmetric* hearing loss. Logistic regression showed that cognitive skills (working memory, language and phonological reading) were particularly sensitive to hearing loss. Investigation of a compensatory effect of enhanced cognition on hearing loss, did not reveal a significant effect. Moore et al. regretted that a great many, possibly the majority of the children with problematic hearing loss around school entry age, are not currently detected. Thereby, detrimental consequences of hearing loss on cognitive performance may be insufficiently understood, and available treatment methods for better speech-in-noise hearing and learning, as FM systems (Mendel et al., 2003) and sound field systems (Dockrell & Shield, 2012) may be disregarded.

The hypothesis that EHF hearing thresholds in children with SSD would be comparably more affected than in children with TD, was confirmed. Monson (as cited in Hunter et al., 2020) has shown that individuals with EHF HI experience deficits in recognizing speech in complex listening environments. Monson stresses that EHF hearing has ecological utility for speech perception. Relatedly, White-Schwoch et al. (2015) showed brain-behaviour relationships between the integrity of the neural coding of speech in noise and phonology in 3–14-year-old-children. These authors suggested neural processing of consonants in noise a fundamental mechanism for language and reading development. Consequences for preschool children's learning in a kindergarten setting are clear, since generally these environments represent complex listening environments. It is important to acknowledge that many children with SSD have weak vocabulary skills (Gierut, 2016), resulting in fewer successful mappings between the acoustic structure of the speech signal and stored phonological representations in long-term memory. For children with a 'double hit' (EHF HI and vocabulary deficit) learning capacity in complex listening environments may be more hampered than in children with SSD with normal EHF hearing.

Key-frequencies for speech

Elevated hearing thresholds at 2 kHz bilaterally were found in children with SSD compared to children with TD. This coincided with a significantly lower SNR in DPOAEs at 2.2 kHz (left ear 5.1 dB mean difference between groups). Here, a clear benefit of combining behavioral and objective hearing measures when testing children is evident (Krueger & Storkel, 2017; Mendel, 2008). Thus, claiming that reduced memory and attention are the simple causes for the impairment, would be incorrect (Tamm, 1912). Rather, the most probable explanation for this finding, is reduced cochlear amplification function for this key-frequency. Possible consequences for children with SSD would be less detailed acoustic phonetic perception of speech sounds that rely on acoustic cues around 2 kHz. Lindblad (2002) defines the acoustic peak for velar plosives /k/ and /g/ at around 1.5 to 4 kHz. Hence, the acoustic information of velar sounds may be comparably more difficult to perceive, if CA of these frequencies is reduced. It is well known that velar fronting is a common process in early speech development (Vihman et al., 1986: Strömbergsson, 2014) and many children with SSD exhibit particular difficulties acquiring velar sounds (Hansson & Nettelbladt, 2002; Strömbergsson, 2014). Future studies (Nakeva von Mentzer, ongoing) may reveal whether the children with SSD in the present study, exhibit comparably more problems with discrimination and production of velar sounds. If so, a link between impaired CA and phonemic discrimination and production, may be confirmed. Another line worth investigating, is the development of the frequency-dependent shift of admittance when an ASR occur. As Madell and Flexer stress, only small-n studies have been conducted thus far (Feeney & Sanford, as cited in Madell & Flexer, 2014) and more knowledge is needed as to when in development a positive peak admittance at 1000 Hz occurs.

In all children, hearing thresholds at the key-frequencies for speech, at 2 and 4 kHz and/or DPOAEs within similar spectral regions, significantly predicted 7–12% of the variance in the American Listen-Say test. The regression model with two predictors was significant on the right ear for the quiet (predicted variance 8%) and noise condition (predicted variance 7%). On the left ear, only DPOAE SNRs was significant, for the quiet condition (predicted variance 10%) and for phonemic reproduction (predicted variance 12%). On the right ear, only hearing thresholds was significant, predicting 11% of the variance. Overall, the present study´s regression models showed a generally weak relationship between hearing measures and phonemic discrimination and reproduction in the children. As only one tool for speech discrimination and reproduction was used, the American Listen-Say test, future studies should investigate how hearing measures relate to standard audiometric tests for speech perception or kindergarten classroom perception performance.

Practical implications

Overall, the present study suggests that subclinical hearing loss could be one of the challenges children with SSD are facing. Thus, further investigation of hearing, including objective tests for middle and inner ear functioning, is warranted for this population. Merely relying on pure-tone-audiometry screening is not sufficient to understand how hearing contributes to speech perception, speech production and cognitive performance in SSD. Interdisciplinary work between speech language pathology and audiology is crucial to enable a fuller understanding of SSD, since it arises from deficits in multiple, interrelated systems (Krueger & Storkel, 2017). Further investigation of subclinical hearing loss and SSD should focus either on risk factor models where subclinical hearing loss places a child at a greater risk (likelihood) of SSD, and association models where subclinical hearing loss could co-occur with SSD, but the likelihood of one is not dependent on the likelihood of the other. The present study's findings do not support focusing on single distal cause models where sub-clinical hearing loss would cause SSD.

Limitations of the study

The present study had several limitations. First, there was little possibility to balance the test and control group with respect to ethnicity. As a result, the findings of this research may be difficult to generalize to the broader population of children with SSD. Since all children with SSD were diagnosed by a professional SLP, accuracy of the diagnosis is not questioned, but other factors, such as socioeconomic background, may also have influenced children's performance. The methodology for WAI morphology analysis needs further improvement, both in establishing criterion as for when a peak is present, and at which frequencies it appears. As for missing middle ear values, children with SSD were over represented. Analysis of missing values could further guide the conclusions drawn in research (Kang, 2013). However, this was beyond the scope of the present study.

Conclusions

Children with SSD exhibited significantly less sensitive hearing compared to children with TD. This was demonstrated in behavioural and objective hearing test results. Absent contralateral acoustic stapedius reflexes in conjunction with significantly less sensitive hearing thresholds, may negatively affect these children's speech perception abilities in complex listening environments, such as at kindergarten where the main learning activities of a preschool child takes place. Reduced hearing sensitivity and reduced cochlear amplification function at 2 kHz could impede the acquisition of specific speech sounds, such as velar sounds. Fronting of velar sounds (k-t, g-d) is a common phonological process observed in both children with TD and children with SSD. An ongoing study will reveal if velar phonemes are indeed more affected than others, in the children with SSD.

Future studies

More studies are needed to investigate developmental aspects of the frequency-dependent shift of admittance when a contralateral ASR occurs. Norms for typical children are warranted so these may be used when analyzing results in children with SSD.

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Data availability

The data may be available upon request from the author.

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