2024-06-03

Caregiver and Clinician Impressions on the Development of Spoken Language in Autistic Cochlear Implant Users

Courtney Dawn Marks
Brigham Young University

Caregiver and Clinician Impressions on the Development of Spoken Language in Autistic Cochlear Implant Users

Courtney Dawn Marks

A thesis submitted to the faculty of
Brigham Young University
in partial fulfillment of the requirements for the degree of

Master of Science

Garrett J. Cardon, Chair
Connie L. Summers
Tyson G. Harmon

Department of Communication Disorders

Brigham Young University

ABSTRACT

Caregiver and Clinician Impressions on the Development of
Spoken Language in Autistic Cochlear Implant Users

Courtney Dawn Marks
Department of Communication Disorders, BYU
Master of Science

The current literature reports that oral language development is not a realistic goal for autistic cochlear implant users (ACI). This is curious, due to the general success of cochlear implants and the fact that many autistic individuals develop strong spoken language skills. Therefore, this study aimed to examine clinician and caregiver insights into spoken language development in the ACI population with the aim of elucidating the factors that influence the poor outcomes reported in the literature. We predicted that cochlear implant function would not be the limiting factor in ACI language development, but that device use and sensory atypicalities might have significant impacts.

Participants were two speech-language pathologists (SLPs), six audiologists (AuDs), and one dually certified SLP/AuD. Additionally, six mothers and their ACI children participated in the study. All participants, excepting one ACI parent, took a survey and then participated in a 30–60-minute interview about their experience either caring for or working with ACIs and their insights into their language development. We also collected language samples in ACI participants. Quantitative analysis included computing descriptive and inferential statistics, where appropriate, concerning language abilities, sensory processing, anxiety, and survey responses regarding factors associated with cochlear implant (CI) use. We also engaged in qualitative thematic analysis of caregiver and clinician interviews. Quantitative and qualitative results were then integrated to triangulate findings.

In important ways, our results are inconsistent with current literature concerning ACIs. For instance, caregiver and clinician statements, as well as quantitative results suggested that it was very possible for ACIs to develop spoken language, when given the right conditions. Qualitative themes that shed light on the factors important to positive language-related outcomes in ACIs included: a) finding (and advocating for) access to care; b) sensory processing difficulties; c) differentiating between autism and hearing loss, and the spectra of both conditions. One specific and novel finding showed that sensory profiles—especially sensory seeking versus sensory averse—may have a significant impact on a child's oral language development following cochlear implantation and should be considered when counseling families on possible outcomes. These findings provide new insight into and concrete future directions for supporting the ACI population.

Keywords: autism spectrum disorders, cochlear implants, assistive technology, mixed methods research

ACKNOWLEDGMENTS

This thesis was truly a huge undertaking, and I could not have done it without the support of so many - first and foremost my Savior, Jesus Christ. My faith in Him has supported me throughout my life and this thesis will certainly not be the last thing I need Him for.

I want to thank my parents. Mom, Dad – you have always believed I could do anything I put my mind to. Your love, support, and belief has meant the world to me, and I would not be the person I am or where I am today without you. Dad, thanks for sharing a love of learning and reading with me. Thanks for encouraging my curiosity as a kid and for always letting me talk about stuff. Mom, thank you for always picking up the phone. Thanks for being the best adventure buddy and for always doing the most for your kids. You two have always been my biggest cheerleaders. I love you both.

I also want to thank my husband, Greg. You are my rock and I have depended so much on your support and love throughout my education. Thank you for believing in me and supporting my dreams – and for buying me SO many burritos. I love you.

I would be remiss if I didn't thank Dr. Garrett Cardon – this thesis would have so many run-on sentences without you! It also would never have happened, so thank you so much for believing this project was worth exploring, believing we could pull it off, and for all the late-night Zoom calls, emails, and GIFS so we could get the thing done - you rock.

Additionally, I want to thank those at the McKay School of Education who approved our research grant and provided funding to this project.

Finally, I want to thank Rey. Thanks for all the late nights, good talks, comfort, laughs, and for lighting up my life. I couldn't have done this without you.

TABLE OF CONTENTS

LIST OF TABLES

LIST OF FIGURES

DESCRIPTION OF THESIS STRUCTURE AND CONTENT

This thesis, *Caregiver and Clinician Impressions on the Development of Spoken Language in Autistic Cochlear Implant Users*, is written in a hybrid format. The preliminary pages of this thesis reflect requirements for submission at the university level. However, the thesis report is presented as a journal article and conforms to the style requirements for submitting research reports to scientific journals. Identity-first language (e.g., "autistic children") is used throughout the report due to this terminology's growing favor over person-first language in autism communities and published research data supporting its use (Bottema-Beutel et al., 2021). However, we also recognize and respect many people's preference for person-first language, including individuals with hearing loss (Nicks et al., 2022). Language preferences for each group have been considered as this paper includes both autistic individuals and people with hearing loss. Appendix A contains the consent forms/Institutional Review Board Approval letter, Appendix B contains Instruments, Appendix C contains the Annotated Bibliography, Appendix D contains Supplementary Data, and Appendix E includes a list of participants' quotes.

Introduction

For many, speech production and perception are primary mechanisms of communication. However, some find speech production difficult or impossible, necessitating alternate methods of communication. These various methods of communication, such as American Sign Language or the use of augmentative and alternative communication (AAC) are no less valuable than the production of spoken language. Still, many persons with hearing loss and their caregivers set their sights on the development of speech. Included in this group are many with congenital deafness diagnosed with autism spectrum disorder (ASD, i.e. autistic person or autistic individual*)*. Unfortunately, the existing data suggest that spoken language development is significantly more difficult for autistic people than non-autistic individuals with hearing loss who are treated with cochlear implants (CI). Curiously, however, approximately 70% of normal hearing autistic people develop spoken language by age 5 (Tager-Flusberg & Kasari, 2013), and CIs have proven to be highly successful in driving typical speech and language development overall. The present study aims to examine variables that underlie spoken language development difficulties in autistic cochlear implantees (ACIs).

Autism and Hearing Loss

Due to the advent of newborn hearing screenings, infants with hearing loss are being diagnosed and treated much earlier than they would be otherwise (Faistauer et al., 2022). In fact, in the United States, 3 months of age is the gold standard for a confirmed diagnosis of hearing loss (Hamzah et al., 2021). Upon such a diagnosis, for children with profound hearing loss, cochlear implantation is regularly offered as a treatment option and can be performed as early as 9 months old in the United States. Additionally, candidacy requirements have become somewhat more liberal for older children and adults, with severe and moderate hearing loss, respectively.

Together with the success of cochlear implantation in the recent past, these factors have led to a steep increase in implanted individuals (Fan-Gang, 2004).

The prevalence of autism is also increasing in the United States. While statistics vary among researchers, two studies found extremely similar numbers with 1/69 people and 1/68 people being autistic in the United States, and numbers being even higher among males with 1/46 (Hansen & Scott, 2018; Tavares et al., 2021). A more recent study found the prevalence of autism to be 1/36 of 8-year-old children in the United States, 4% male and 1% female (Maenner et al., 2023). Furthermore, in recent years, more attention has been given to females with autism. Autism is traditionally diagnosed at a ratio of 4:1, with males being diagnosed with a much higher rate than females (Dworzynski et al., 2012). Some researchers believe that the true ratio is likely much smaller and that females with autism may be underdiagnosed (Dworzynski et al., 2012; Hull et al., 2023; Russell et al., 2011). With research into female presentations of autism increasing and the continuing education of clinicians, it is likely that the recorded prevalence of autism will increase.

Among those with hearing impairment, the prevalence of autism is higher than in the general population, although it should be noted that hearing loss does not cause autism (Demopoulos & Lewine, 2016; Mathew et al., 2022). For instance, one study found that 20–40% of children with hearing loss have an additional disability and estimate that for 1/59 of those children with hearing loss, that additional disability is autism (Brown et al., 2021). On the other hand, it should be noted that some studies report that the rates of hearing loss in autistic individuals resemble the rates of hearing loss in non-autistic individuals (Mathew et al., 2022). Demopoulos and Lewine (2016) found that after examination of autistic individuals' auditory functioning, using methods such as tympanometry, uncomfortable loudness level, and auditory

brainstem response, there were significantly higher rates of peripheral audiological abnormality, among autistic individuals (55%) versus their non-autistic control group (6%). Other research suggests that up to 3.5% of individuals with autism have hearing loss, and the Gallaudet Annual Survey found that of the 8-year-old children receiving services under Inclusion, Diversity, Equity, and Access (IDEA) during the 2009–2010 school year, 1/59 of the children receiving services for hearing loss were also receiving services for autism (Brown et al., 2021; Szymanski et al., 2012). Brown et al. (2021) also reported in their case studies of children with hearing loss and other co-occurring medical disorders that it was the children with severe to profound hearing loss bilaterally that were most likely to be later diagnosed with autism, and Meinzen-Derr et al. (2014) reports that in autistic children with hearing loss, a profound loss is most common. In summary, while not completely understood, individuals with hearing loss represent a significant and growing portion of the autistic population. As such, it is all but certain that audiologists, SLPs, and other professionals serving on cochlear implant teams will work with ACIs and their families. For these reasons, improving our understanding of this population is vital.

One complication that arises when hearing loss and autism co-occur is that there is currently no validated way to properly diagnose autism in hearing-impaired individuals, that is, the Autism Diagnostic Observation Schedule 2 (ADOS-2; Lord et al., 2012) is not yet validated on children who are deaf or hard of hearing (Camarata, 2013; McFayden et al., 2023; Szarkowski et al., 2014; Wiley et al., 2014). As the ADOS-2 is one of the most used assessments when diagnosing a child with autism, research is being done on how it can be used more effectively with children who have hearing loss. Preliminary findings on sensitivity and specificity on the ADOS-2, adapted for Deaf children, are promising, although widespread use may be limited by

the fact that the administering clinician must be able to communicate with the child without using an interpreter (Phillips et al., 2022).

Autism and hearing loss share many of the same symptoms, and diagnosis of one can often delay diagnosis of the other (Tavares et al., 2021). One study found that children with hearing loss are diagnosed with autism 10 months later, on average, than their normal hearing peers (Mandell et al., 2005). This overlap of symptoms can cause diagnostic overshadowing, that is, delays or missed diagnoses that occur when a person has two diagnoses with similar features (Szymanski et al., 2012). Some of these similar characteristics include overall language delays and difficulties with language functioning, delayed theory of mind, failure to respond to one's name, and pragmatic language difficulties (Szarkowski et al., 2014). The comorbidity of hearing loss and autism is alarming because it can cause the diagnosis of autism to be delayed or missed in children with hearing loss as compared to children with typical hearing (Brown et al., 2021; McFayden et al., 2023; Mikic et al., 2016). Many parents and clinicians seeing such symptoms in a child known to have a hearing loss may believe that the symptoms are arising from the hearing loss itself and not seek additional diagnoses. For children with profound hearing loss whose caregivers have decided on cochlear implantation, autism is not commonly diagnosed or discussed before surgery and many parents of Deaf autistic children feel that clinicians do not offer sufficient support or have appropriate experience (Szarkowski et al., 2014; Wiley et al., 2014). For these reasons, including the growing prevalence of newborn hearing screenings, it is less common for a child to receive a diagnosis of autism before a diagnosis of hearing loss. This phenomenon is evidenced by the average difference in age of diagnosis between hearing loss and autism. Similarly, in the minority of cases where a child has received an autism diagnosis first, hearing evaluations are often delayed since behaviors that often prompt such assessments, like

speech or language delays, might be attributed to their autism (Mandell et al., 2005; Meinzen-Derr et al., 2014). Thus, due to the very early identification of most pediatric hearing losses and the relatively later average age of autism diagnosis, and in conjunction with the difficulty of diagnosing ASD in individuals with hearing loss due to the lack of validated testing, as well as the tendency for autistic individuals to present with severe to profound hearing loss, many will receive CIs before parents are aware of a potential autism diagnosis. (Brown et al., 2021; Demopoulos & Lewine, 2016; Hansen & Scott, 2018; Mathew et al., 2022; Szymanski et al., 2012). Many of these parents likely have high hopes that their child will develop spoken language following their implantation, and, given the immense success of cochlear implants (Baron et al., 2019; Blanc et al., 2022) in the non-autistic population, may even have been encouraged in their hoping by the clinicians on their CI team.

Autism and Cochlear Implantation

Cochlear implantation is often the treatment of choice for children born with profound hearing loss. In non-autistic children, typical/near-typical spoken language development is often achieved with cochlear implantation (Baron et al., 2019; Blanc et al., 2022). Factors that influence the outcome of cochlear implantation include age of implantation, family influences and support, level of compliance with habilitation programs, and some unchangeable factors such as the presence of auditory neuropathy or later diagnosed syndromes, and unilateral versus bilateral implantation (Baron et al., 2019; Black et al., 2012; Blanc et al., 2022). For instance, a study by Baron et al. (2019) found that for bilaterally implanted individuals, benefits included better speech intelligibility and discrimination. Their study also found that children who had bilateral CIs had accelerated language development when compared to their progress before receiving their second implant.

Before a child can be implanted, their candidacy must be evaluated by a CI team, consisting of, at least, both their surgeon and an audiologist who will continue to monitor their aural health and hearing after implantation and surgical recovery has taken place. In some cases, additional professionals such as aural rehabilitation specialists or SLPs may also be members of the CI team (American Speech-Language-Hearing Association [ASHA], 2004). CIs have been approved by the FDA for children 12 months and older, although one model was approved in 2020 to be used in children as young as 9 months old (ASHA, 2020). These children must have experienced no or limited benefit from hearing aids, be in good physical health, and have appropriate family and social support to justify implantation. Children who are implanted early and receive early intervention typically have better speech and language outcomes than those who wait (Faistauer et al., 2022; Yoshinaga-Itano et al., 1998). In contrast, children implanted after age 10 had statistically significant poorer outcomes than children implanted earlier (ASHA, 2004; Sharma et al., 2002a, 2002b). These spoken language outcomes for non-autistic CI users emphasize the importance of early implantation but also necessitate the experience and close observation of clinicians working with them to identify signs of autism when that diagnosis isn't typically made until a few years later.

While common benefits for non-autistic children with CIs include improved speech perception, speech production, and language skills, for autistic children, benefits of CI use are often vastly different. The limited available literature shows that spoken language is not commonly developed to the same extent in ACIs, even after a surgically successful and early implantation (Donaldson et al., 2004; Lachowska et al., 2018; Mathew et al., 2022; Mikic et al., 2016; Tavares et al., 2021).

Many autistic children (up to 70%) without hearing loss develop spoken language, as do most children with CIs if they are implanted early enough and receive appropriate rehabilitation (Tager-Flusberg & Kasari, 2013). When autism and cochlear implantation co-occur, however, the likelihood of a child developing spoken language seems to drastically decrease (Donaldson et al., 2004; Lachowska et al., 2018). For instance, Lachowska et al. (2018) emphasized this decrease in likelihood when they declared that based on their study and results from other studies, autistic children with CIs do not develop speech and language even after many years of habilitation. Donaldson et al. (2004) made a similar statement in their paper when they emphasized that speech production sufficient for oral communication is not a realistic goal for a child with a CI diagnosed with autism.

Although spoken language may not occur after cochlear implantation for autistic children, that does not mean that a CI would have no benefit. Some researchers report that additional benefits for ACIs include improved eye contact, improved interactions with family members, the ability to identify sounds in their environment, increased vocalizations, responses to requests, response to their name, and reduced anxiety (Donaldson et al., 2004; Lachowska et al., 2018; Tavares et al., 2021). These benefits alone may justify implantation for autistic children, even if spoken language is never acquired. They also emphasize the importance of setting realistic expectations for parents who may be expecting their child to acquire spoken language and may not recognize these other benefits as positive outcomes resulting from implantation.

A study published by Jenks et al. (2022) was, at the time, the largest study with the longest follow-up available on the range of outcomes experienced by autistic children following cochlear implantation. They found that of the 30 ACI participants, 31% used spoken language

exclusively, 14% used a mix of spoken language and sign, 14% used sign language alone, and 28% had no mode of communication. These numbers more closely resemble those found by Tager-Flusberg and Kasari (2013) when they reported that approximately 30% of autistic children do not develop oral communication even in the absence of hearing loss. Of note, four ACIs from the Jenks et al. study were able to participate and graduate from a mainstreamed high school. Apart from those four, however, the remainder of the participants did not have age-appropriate communicative abilities, were limited in what and how they could communicate, and many used AAC in conjunction with their preferred form of communication. Notably, this study did not describe what oral communication looked like for their participants or give specific information about how their ability to learn and use oral communication changed post implantation.

Additionally, two previous studies have shown that some, albeit few, autistic children have developed spoken language after cochlear implantation. For instance, Meinzen-Derr (2014) reported that, of the 14 autistic participants in their study who had received cochlear implants, four used oral communication. Of those four, two used oral communication in conjunction with sign and behavior to supplement their speech production and two used oral communication as their primary method of communication. This development raises questions about why only 2/14 ACIs were able to primarily use oral communication and what was different about them compared to their peers. Similarly, Eshraghi et al. (2015) reported that 10/15 of their subjects improved their verbal expression and could afterwards use simple phrases and commands or produce sentences. Some (4/15) of their subjects, however, continued to have only some vocalizations. The non-autistic control group, in contrast, resulted in all 15 implantees being able to either produce sentences or in one child's case, primarily use short phrases and commands.

Thus, a significant difference in spoken language development between autistic and neurotypical cochlear implant recipients exists, with the former having much more difficulty than their non-autistic peers.

Many autistic individuals develop spoken language (Tager-Flusberg & Kasari, 2013). It should be noted, however, that approximately 25–30% of autistic children, without accounting for hearing loss, use no or minimal verbal expressions by the time they enter kindergarten (Tager-Flusberg & Kasari, 2013). These data suggest that for ACIs it should be expected that there will be some who would have spoken language difficulty independent of their hearing loss. However, given the success of cochlear implantation among neurotypical implantees in driving auditory development and spoken language acquisition, these data also point to the notion that ACIs should be capable of developing spoken language at a rate that mirrors such development in the autistic population overall. The lack of clarity regarding the most likely cause(s) for delayed development of spoken language establishes the motivation for this study.

Given the above, it is unclear if part of the reason for the results of implantation to be so drastically different between non-autistic and autistic children is that there are few studies done about implanted autistic populations. Another possible explanation may be in part, due to the small, but significant, population size, available subjects to participate in studies have been limited. These findings emphasize the importance of studying this population and understanding why spoken language development is so seemingly uncommon in ACIs. For instance, it's reasonable to speculate that poor outcomes could be related to poor CI usage patterns, inadequate therapy due to limited clinician experience or education, delayed implantation, or delayed diagnosis of hearing loss. It is also possible that sensory processing differences in the autism population play a role in poor speech and language outcomes in ACIs.

The Relationship Between Autistic Traits and Hearing Loss

Spoken language delays are not the only challenges associated with autism and hearing loss, other difficulties include heightened fear and anxiety. In their study, van Steensel and Heeman (2017) found that autistic children had a higher prevalence of anxiety than non-autistic children as well as found that for all children with anxiety, autistic children had more severe manifestations. They also emphasized the need for future research, including the effects that bullying or other stressful life events may have on anxiety in autistic children. Similarly, children with CIs also experience heightened levels of anxiety (Ghaly et al., 2022). This may be due, in part, to increased bullying as van Steensel and Heeman (2017) pointed out. Feijóo et al. (2021) reported a higher prevalence of bullying among children with CIs and found that most of the bullying was attributed to wearing their implant. Other factors included asking others to repeat themselves and not always understanding what was said. Given this, it may be possible that anxiety, autism, and cochlear implantation also overlap although no studies focused exclusively on that relationship to our knowledge. One study did report that reduced anxiety was a positive outcome of CI use for ACIs, (Lachowska et al., 2018) indicating some connection between autism and cochlear implantation.

Many autistic children have trouble with sensory processing, which may contribute to their difficulty with spoken language development as speech and language are both multisensory processes. Sensory processing and language difficulties have been shown to be related in previous studies (e.g., Cooper, 2021). As emphasized by Stevenson et al. (2014), successful speech and communication is reliant on multisensory perceptual binding, or the ability to combine information from various senses into one coherent whole. For ACIs, this may be especially relevant. Many non-autistic cochlear implant users (NACI) rely on visual signals in

addition to the auditory information they perceive through their implant to understand spoken language—this was only made more evident by the recent pandemic where masks limited available visual feedback to CI users and their communicative abilities declined (Homans & Vroegop, 2021; Sönnichsen et al., 2022). ACIs, in contrast, do not seem to combine visual and auditory information as readily as NACIs. An example of this phenomena comes from the McGurk effect, with autistic individuals being less influenced than non-autistic individuals (Bebko et al., 2014; Stevenson et al., 2014). Given the above, it is possible that language difficulty in ACIs could be associated with multisensory integration and sensory processing difficulties.

Understanding Parent Experience

Due to the reported difficulty experienced by many ACIs in communication, much of the available information on how ACIs have benefited or not from cochlear implantation has been gathered via parent survey or interview. Additionally, parent involvement is vital following cochlear implantation as they participate in aural rehabilitation with their child (Zaidman-Zait & Curle, 2018). One study conducted by Zaidman-Zait and Curle (2018) focused on the experiences of ACI mothers, and interviews were conducted over the phone with nine mothers of male ACIs. Of those children, only two reported using oral communication, although the meaning of oral communication was not made clear. Additionally, mothers from this study reported that difficulty communicating with their child is among some of their greatest difficulties in parenting their ACI. Much of the reported benefits experienced by ACIs such as improved eye contact, improved interactions with family members, the ability to identify sounds in their environment, increased vocalizations, responses to requests, response to their name, and

reduced anxiety come from parent report (Donaldson et al., 2004; Lachowska et al., 2018; Tavares et al., 2021).

Understanding Clinician Confidence

One study emphasized that due to the difficulty of differentiating symptoms of autism from other health conditions, such as hearing impairment, generalist care providers may not immediately turn to a diagnosis of autism (Mandell et al., 2005). These delays in autism diagnosis may also be caused by the unawareness about the common co-occurrence of these two pathologies by the surgeons, audiologists, and speech language pathologists who have these children on their caseload. Diagnostic delays in the autistic population with hearing loss are additionally impacted not just by diagnostic overshadowing and lack of diagnostic tools as mentioned above, but also by the struggle to find competent clinicians (McFayden et al., 2023). Many professionals who work with autistic children express low levels of confidence and training (Bono et al., 2022). A greater understanding about the confidence, experience, and training that professionals in each of these groups have when working with implanted autistic individuals is vitally important. These providers are the gatekeepers of care and should be the first to recognize when a child they are working with is experiencing something that is more than hearing loss so that they can create realistic expectations for their patients and their patient's caregivers (Mathew et al., 2022). Most of these professionals, as well as parents, will likely indicate the development of spoken language as one of the results of successful cochlear implantation, but what happens when spoken language is not developed as expected? It is also vitally important to understand the confidence of clinicians in providing intervention for this population once they have received both an autism and hearing loss diagnosis.

Understanding clinician confidence, experience, and training is an important step towards ensuring that implanted autistic children are diagnosed quickly and accurately and receive necessary intervention to support their development following both their autism diagnosis and cochlear implantation.

Aims

This study aimed to explore why spoken language development for ACIs seems to be more difficult when compared to NACIs and autistic children with typical hearing (ATH), by investigating the relationships between autistic traits, sensory (esp. auditory) function, anxiety, and oral communication. We also aimed to gain detailed insight into the nuances of ACI spoken language development by examining the experiences of caregivers and clinicians relative to cochlear implantation in autistic individuals.

The current study employed a convergent, mixed methods design, completed in two phases. Phase 1 focused on gathering and analyzing quantitative information, while Phase 2 focused on the qualitative. Quantitative and qualitative data were integrated during analysis. The first phase included surveys for caregivers and clinicians and the second phase included follow-up interviews with caregivers and clinicians, in addition to language sampling with ACI participants. See Figure 1 for detailing on phases and the measures completed in each phase. Participants were recruited through social media, flyers, referral from associates of the research team, and word of mouth. All procedures were approved by the Institutional Review Board of Brigham Young University and were in accordance with the Declaration of Helsinki.

Methods

Participants

Group 1: Caregivers

The first group of participants included 49 caregivers of ACIs (n = 6), NACIs (n = 4), ATHs (n = 4), and non-autistic children with typical hearing (NATH; n = 34). Additionally, five ACI caregivers were included in Phase 2 interviews. To be included as a caregiver of an autistic child, their child must have been diagnosed according to the ADOS-2 and criteria from the *Diagnostic and Statistical Manual of Mental Disorders 5th Edition* (DSM-V; American Psychiatric Association, 2013). Diagnoses were verified by parents and degree of autistic trait expression was determined via the Social Responsiveness Scale, 2nd Edition (SRS-2; Constantino, 2013). To be included as the caregiver of a child with a CI, their child must have received their implant(s) at least 6 months prior to their research involvement. Caregivers were recruited by word of mouth, through their doctor's office, and from participating clinicians. Caregivers in this study came from several locations across the United States.

Group 2: Clinicians

The second group of participants included six audiologists (AuD), two speech-language pathologists (SLP), and one dually licensed AuD and SLP from various locations across the United States. See Table 1 for full demographic details. These subjects filled out an online survey during Phase 1. Participants then participated in in-depth, semi-structured qualitative interviews as part of Phase 2. Clinicians were recruited through word of mouth and referrals from contacts of the thesis chair. Clinicians came from several locations across the United States.

Group 3: Children

The third group of participants included five ACIs, see Table 8 for a full summary. These individuals were recruited through their audiologist's office using an email or survey to determine interest in participating in this study, and by word of mouth. Participants were three females ages 03.06, 05.06, and 14.03, as well as two males ages 04.02 and 07.08. Autistic subjects were diagnosed according to the ADOS-2 and criteria from the DSM-V. All participants had received their implants at least 6 months prior to their research involvement. Exclusion criteria for this group included neurological impairment, seizure history, head injury with loss of consciousness, and/or substance abuse. Though for most studies, five participants would be considered a small sample, given the scarcity of ACIs and the qualitative methods planned, this sample size is appropriate for the current study as indicated in multiple studies including those by Donaldson et al., (2004), Hayman and Franck (2005), Lachowska et al., (2018) and Zaidman-Zait and Curle (2018).

Instrumentation

Phase 1

Measure 1. An online survey was sent to caregivers of all participants included a measure of expressive and receptive language, sensory processing, anxiety, autistic traits, and questions surrounding cochlear implant use. Some responses were descriptive, and others were scored, see Table 2. The language measure included questions adapted from the Bilingual English Spanish Assessment Inventory to Assess Language Knowledge (BESA ITALK; Peña et al., 2018) and from Eshraghi et al., (2015). See Appendix B for full list of questions.

To examine sensory processing, we used the Short Sensory Profile (SSP; Dunn, 1997). This instrument is a 38-item caregiver questionnaire that is meant to assess areas of sensory

processing, modulation, and behavioral and emotional responses in children ages 3–10. Questions utilize a Likert Scale of 1–5 to assess if a child's sensory experience is "different" or typical. The SSP was chosen for our study due to its widespread use in assessing autistic children (Crasta et al., 2020; Glod et al., 2019; Simpson et al., 2019) and ease of administration, particularly given that caregivers are the main participants in this study. Additionally, it has moderate to strong internal consistency ranging from 0.70–0.90 in the different sections (Tomchek & Dunn, 2007). Different sections include tactile sensitivity, taste/smell sensitivity, movement sensitivity, under-responsive/seeks sensation, auditory filtering, low energy/weak, and visual/auditory sensitivity (Tomchek & Dunn, 2007).

To measure levels of anxiety, we used The Screen for Child Related Anxiety Disorders (SCARED; Birmaher et al., 1999). This is a 41-item questionnaire for use in children ages 8–18 that is filled out by a parent, rating common anxiety-inducing situations on a 3-point Likert scale. Parents respond from 0–2, with zero indicating "not true or hardly ever true" and two indicating "very true or often true." The questionnaire screens for anxiety disorders in four domains: panic/somatic, separation anxiety, generalized anxiety, and school phobia. The SCARED has been shown to have generally high internal consistency ($\alpha = .91$) as well as moderate sensitivity (.71) and specificity (.67) (Hale et al., 2011). Stern et al. (2014) found moderate to strong psychometric properties in the SCARED for identifying anxiety disorders in autistic children, (Cronbach's alpha = 0.92). This instrument was chosen both for its overall validity in identifying anxiety disorders, and for its ability to identify these disorders in autistic children.

Questions about CI use allowed us to create a CI user profile for each applicable participant and evaluate if inconsistencies or difficulty with the device itself contributed to poorer language outcomes. To informally assess cognition, parents were asked questions

regarding school performance, additional disabilities, additional services being received, and various cognitive skills. Researchers recognize that this is not a comprehensive evaluation of cognition and note a full understanding of each child participant's cognitive ability to be a limitation of this study.

Measure 2. A pre-interview survey was sent to clinicians. This survey gathered demographic and caseload information, as well questions about their experience in providing care to ACIs.

Phase 2

Measure 1. Caregivers of ACI participants joined in a semi-structured interview, which included administration of the Auditory Skills Checklist (Meinzen-Derr et al., 2004) and open-ended questions related to the following topics: a) what they felt their biggest challenges were in caring for their child, b) what a successful outcome of cochlear implantation means to them, c) if they were aware that their child may be autistic before receiving their CI, and d) factors contributing to success of language development. For a full list of questions, see Appendix C, although the research team maintained the ability to ask to follow-up questions not specifically listed.

Measure 2. A qualitative, semi-structured interview was given to clinicians by the research team. These clinicians commented on their definitions of what a successful cochlear implantation for an autistic person looked like, what they viewed as predictors of success for this population, as well as reported on their confidence in providing specialized high-quality care for these individuals. Other questions included the following, based on initial survey responses: a) provide further information about clinician confidence during treatment, b) how often they participate in interdisciplinary collaboration, and c) how often they are aware of an autism

diagnosis or notice indicators of autism before implantation. For a full list of survey and interview questions see Appendix B.

Measure 3. All five ACI participants were invited to participate in an additional language assessment, with three returning a language sample. The language sample was elicited at home by a caregiver, either through story retell or as an expository language sample, depending on participant age. Children ages 3.6–12.8 participated in the story retell and children ages 12.9–18.9 participated in the expository sample. These samples were analyzed using Systematic Analysis of Language Transcripts (SALT; Miller & Iglesias, 2020; Miller & Nockerts, 2024). Children ages 3.6–7.5 used *Frog, Where Are You?* Children ages 7.6–8.11 used *Pookins Gets Her Way*. Children over the age of 12.8 were allowed to give an expository language sample on the topic of their choice. Samples were then transcribed, coded, and analyzed through SALT. We extracted measures of intelligibility (% intelligible words), measures of syntax and morphology (MLU in morphemes, % utterances with verbs), measures of semantics (number of total words, number of different words), and errors (% utterances with errors).

Procedures

Phase 1

Caregivers participated in the initial survey online at their convenience via Qualtrics. Surveys took approximately 30 minutes to complete. All participants who completed a survey were entered into a drawing to receive a cash prize. Clinicians also completed an online survey via Qualtrics. After filling out the pre-interview survey, clinicians were asked if they would be interested in participating in a short interview for Phase 2.

Phase 2

Caregiver and clinician interviews were completed virtually, using *Zoom* (Version 5.17.11). Participant responses were then transcribed and coded for qualitative analysis. Caregiver interviews lasted approximately 60 minutes, while clinician interviews were limited to 30 minutes. All participants were compensated for their time.

Language sampling for ACIs was completed by caregivers for children younger than 12.8 with explicit instructions from the research team. Children older than 12.8 participated via Zoom. For the children completing a story retell, caregivers were provided with a digital version of the story, a script, and video instructions from the research team. Those participating in the expository sample were minimally affected by remote participation. All participants who participated in language sampling received compensation for their time through their caregiver.

Data Analysis

Phase 1: Statistical Analysis

We performed the following statistical analyses on the quantitative data collected in both caregivers and clinicians: Descriptive statistics, such as means and standard deviations, were computed for appropriate questionnaire items. Such descriptive statistics were used for general observation of questionnaire data and were particularly useful in determining clinician rankings of obstacles to CI success in ACIs. Also, given that data were not normally distributed, we employed non-parametric statistics for all between-group comparisons and within-group correlations. We used Mann-Whitney U, Kruskall-Wallis one-way ANOVA testing, and proportions testing to assess between-group and across group differences for our language measure and each standardized behavioral questionnaire. Partial correlations, controlling for age, were calculated between the total scores of the language measure, Auditory Skills Checklist,

SSP, and SCARED to determine their relationships. Additionally, we compared the strengths of these correlations between groups of ACI and NACI children using a general linear model method, to assess potential statistical differences in these associations.

Phase 2: Qualitative Analysis

Following video and audio recording of all interviews via Zoom, they were orthographically transcribed and coded using the Framework Method (Gale et al., 2013). We used the ATLAS.ti software package to facilitate code organization and analysis (Version 24 Mac). Each question was either descriptive or scored, see Table 3.

A team of one graduate and three undergraduate students coded each interview. We employed a multi-step iterative process to code caregiver and clinician interview transcripts. First, each of the four team members familiarized themselves with interview data by watching videos multiple times while reading and correcting the transcripts. Team members recorded their thoughts, impressions, and insights throughout this and all subsequent coding steps. Following the readying of the transcripts, the coding team created an initial codebook during a meeting held to discuss themes initially observed in the interviews. In this meeting, the team discussed and defined these first codes and refined the codebook into a concise list ready to be used in coding. Then, team members each iteratively coded select transcripts, met together to discuss their coding and memos, and refine the codebook (i.e., modify, merge, add, or delete codes). Once consensus was reached in each meeting about coding and codebook modifications, each team member returned to coding select transcripts. This iterative process occurred over three rounds until consensus was reached on a final codebook structure and all unanimously agreed that codes were appropriate and captured the essence of the interviews. An audit trail of code modifications was kept in ATLAS.ti. Finally, we met to organize codes into salient themes and sub-themes.

Based on this organization, we then completed the reporting of these constructs by summarizing themes and selecting quotations to illustrate and support these notions (Braun & Clarke, 2006, 2021; Gale et al., 2013; Harmon, 2020).

Language sample analysis was done through SALT. Samples were transcribed and coded, and SALT was used to generate a Standard Measures Report that included measures of intelligibility (% intelligible words), measures of syntax and morphology (MLU in morphemes, % utterances with verbs), measures of semantics (number of total words, number of different words), and errors (% utterances with errors).

Positionality

I am a 25-year-old White woman born and raised primarily in the United States of America, with some years spent abroad due to my father's ongoing service in the United States Air Force. I hold a Bachelor of Science in Communication Disorders and am currently a master's student at Brigham Young University in Provo, Utah, studying speech-language pathology. My religious affiliation is a member of The Church of Jesus Christ of Latter-Day Saints. I am hearing and have one close family member with pediatric hearing loss. Although I have no close friends or family who are diagnosed with autism, my interest in studying ACIs stems from an interest in audiology, language development, and how professionals can provide neuroaffirming care. I acknowledge that my background provides certain biases in the interpretation of participants' interviews, and I strive to be cognizant of these biases and how they shape my research.

Results

ACI Participant Profiles

Some non-essential biographical and demographic data has been modified or removed to maintain participant confidentiality.

ACI Participant 1: Owen

At the time of this study, Owen was a 4.17 year-old male. Owen was born with a parent-reported bilateral moderate sensorineural hearing loss (SNHL) with a family history of hearing loss. Following his hearing loss diagnosis at the time of his newborn hearing screening, Owen's caregivers immediately brought him to the audiologist for an auditory brainstem response test (ABR) and began hearing aid use. At 10 months, the audiologist recognized that Owen's presentation was not typical of a child who only had a hearing loss and recommended that he be evaluated for autism. Owen's parent did not specify what behaviors the clinician had noticed. Owen's parents immediately joined the year-long wait list for an autism assessment. During their time on the wait list, Owen's hearing loss continued to progress, and his parents decided to pursue bilateral cochlear implantation (CI) when he was 21 months old. Owen received his autism evaluation at 27 months and began applied behavior analysis (ABA) via the Early Start Denver Model (ESDM) a month after diagnosis.

At the time of the study, Owen's mother reported that he had bonded with his CIs. He understood that when he wore them, he could hear and vice versa. Owen was wearing his CIs for approximately 12 hours a day and both his play skills and intense behaviors had improved since implantation. Owen's mother reported that he had strong sensory seeking tendencies. Owen was producing 4–5-word sentences, using Total Communication, and had an Auditory Skills Checklist score of 68/70.

Owen completed a language sample, participating in a modified story-retell of *Frog, Where Are You?* (Mayer, 1969). During Owen's language sample, his parent would read him one page at a time and ask him what he remembered from that page. It should be noted that Owen's responses indicated generative speech production and were not echolalic in nature. Owen's language sample was approximately 10 minutes. Owen was 60% intelligible at the utterance level and 88.4% intelligible at the word level. Intelligibility milestones include 75% at 36 months and 100% at 48 months in connected speech (Paul et al., 2018). Additionally, Owen's MLU in words was 4.95 and his MLU in morphemes was 5.45. According to Pence Turnbull and Justice (2017) the MLU in morphemes of a 42-month-old should be 2.96–4.6 while a 54-month-old should be between 3.96–6.08. In this sample Owen produced 109 words and 46 different words (NDW), with his moving-average NDW being 43. According to the SALT Performance Report, the moving-average NDW of preschoolers and kindergarteners was 50 (Miller & Nockerts, 2024). See Table 8 for a summary.

ACI Participant 2: Claire

At the time of this study, Claire was a teenage female (14.3 years of age). Claire passed her newborn hearing screening. However, following a pediatrician visit and a subsequent referral to the audiologist at age 3, Claire was diagnosed with a profound SNHL across all frequencies in her left ear and a mild-moderate flat SNHL in her right ear which was immediately aided with hearing aids. Claire developed oral communication skills and was "very verbal" with her hearing aids. Around first grade, Claire's parents noticed that her hearing was declining and that she had lost significant hearing capability in her right ear. They decided to pursue cochlear implantation and by her seventh birthday she had received bilateral implants. A few years later, at age 12, Claire was diagnosed with autism.

Today, Claire loves her implants and wears them during all waking hours unless she is engaged in water-based activities (e.g. swimming, showering). Claire's parents reported that she regularly exhibits sensory seeking behaviors and that she is very socially motivated to wear her implants. Claire uses oral language fluently to communicate with family and peers and has an Auditory Skills Checklist score of 66/70.

Claire completed a language sample, participating in a conversation with examiners and a parent. Claire was 95.1% intelligible at the utterance level and 99.2% intelligible at the word level. Intelligibility milestones include 100% intelligibility by age 4 in connected speech (Paul et al., 2018). Additionally, Claire's MLU in words was 6.53 and her MLU in morphemes was 7.31. According to Pence Turnbull and Justice (2017) the MLU in morphemes should be between 3.96–6.08 at age 5. It should be noted that MLU is not commonly used in teenagers but has been reported in young school-aged children as 4.99 MLU in words and 5.51 MLU in morphemes for children aged 8.6–8.11 (Rice et al., 2010). In this sample Claire produced 359 words and 157 different words (NDW), with her moving-average NDW being 62. According to the SALT Performance Report, the moving-average NDW seventh graders was 60. (Miller & Nockerts, 2024). See Table 8 for a full summary.

ACI Participant 3: Nora

At the time of this study, Nora was a 3.06-year-old female. At birth, Nora was referred on her newborn hearing screening. Her parents were told that it was likely fluid in her ears and waited the recommended time to schedule an audiology appointment. At that time, ABR confirmed profound SNHL in both ears. Nora's parents pursued a second opinion and received the same diagnosis, at which point Nora began a hearing aid trial as part of the CI qualification process. At 9 months, Nora received bilateral CIs, which were activated at 10 months. Nora's

parents reported seeing progress in oral language development and auditory skills in the months that followed, but that shortly before she turned 2 years, she experienced a language regression. Nora's parents began traveling with her significantly to access appropriate services. A professional acquaintance noted that there seemed to be an additional factor that was complicating her hearing loss and language development. Shortly after this suggestion, Nora received a diagnosis of autism spectrum disorder at age 2. Nora then began a full-time autism preschool and received play-based ABA.

At the time of the present study, Nora is a consistent CI user and enjoys routines related to CI care such as putting them on, taking them off, and turning them on if they disconnect. Nora's parents report that she has aversive responses to some tactile stimuli (i.e. her hands must be clean, dislikes clothing textures and long sleeves) but that she does enjoy her CI. Nora also has sensory seeking tendencies for movement and does not exhibit hypersensitivities to sound. Her Auditory Skills Checklist score was 58/70.

Nora did not participate in a language sample.

ACI Participant 4: Sophie

At the time of this study, Sophie was a 5.06-year-old female. Sophie was diagnosed with a profound bilateral hearing loss at 2 months. Upon diagnosis, Sophie began hearing aid trials in preparation for CI and began auditory verbal therapy (AVT) shortly thereafter. At 11 months, Sophie received bilateral CIs, which were activated a month later, all the while continuing to receive AVT. Due to the COVID-19 pandemic, Sophie began virtual therapy, which reportedly had little effect. When COVID precautions had relaxed, Sophie began receiving inpatient therapy where she began making rapid progress in her spoken language development. At this time, Sophie's speech therapist recommended that she be evaluated for autism. Although Sophie's

parents weren't convinced that Sophie was autistic, they felt that services would be more available for autistic children, so they supported the evaluation. This testing resulted in Sophie's eventual diagnosis at age 3.

Following Sophie's autism diagnosis, Sophie attended a preschool for the deaf where signing was not allowed. CI use was physically enforced at this school, often against Sophie's will. Unfortunately, this method of treatment created a trauma response in Sophie, leading to decreased CI use and concomitant lessening of oral language.

Today, Sophie uses Total Communication, and her parents are beginning to reintroduce her CI after 2 years of non-use, due to the aforementioned trauma. Sophie craves movement (i.e., sensory seeking), has vestibular differences, and has no intense sensory aversions. Sophie has an Auditory Skills Checklist score of 2/70. Sophie did not participate in a language sample.

ACI Participant 5: Luke

At the time of this study, Luke was a 7.08-year-old male. Luke was referred for a full audiological diagnostic session when he didn't pass his newborn hearing screening. Luke's parents were told that it was likely fluid buildup in his ears that led to this result. At 3 weeks, however, Luke visited an ENT and was diagnosed with a bilateral severe-profound SNHL via ABR. Subsequently, Luke received amplification at 4 weeks and later was approved for CIs. Due to physician concerns about sedating an infant over a long period of time, Luke received his implants sequentially—the right device at 9 months and the left CI at 14 months. In Luke's home state, there was limited access to oral schools for deaf children. So, at 18 months, Luke's parents decided to relocate to an area with better access to services. Shortly thereafter, Luke began receiving services more regularly. Additionally, Luke's speech therapist advised his parents that

he may also be autistic. At age 3, after a referral to an autism clinic, Luke was diagnosed with autism.

At the time of the current study, Luke enjoys hearing and wearing his CIs for approximately 16 hours a day. Luke has sensory seeking tendencies, particularly to gross motor activities (i.e. pressure "squeezes"). Luke has some sensory sensitivities with clothing textures and eating. Luke is an oral communicator, producing greater than five-word sentences. Parents also report that Luke most often understands conversations. He presented with an Auditory Skills Checklist score of 65/70.

Luke participated in a language sample, completing a modified story retell of *Pookins Gets Her Way* (Lester, 1987). Luke's parent read Luke the story and asked him to retell it at the end, however, they did provide visual support by turning back through the pages of the book. Luke's language sample lasted 1 minute and 11 seconds. It should be noted that Luke was not enthusiastic about participating in this portion of the study and his parents report that this was a poor representation of his true abilities. Luke was 100% intelligible at the utterance level and 100% intelligible at the word level. Established intelligibility milestones include 100% intelligibility by age 4 in connected speech (Paul et al., 2018). Additionally, Luke's MLU in words was 4.83 and his MLU in morphemes was 5.17. According to Pence Turnbull and Justice (2017) the MLU in morphemes should be between 3.96–6.08 at age 5. Again, MLU is not commonly used to measure language in older children. One study found, however, in children aged 7.06–7.11 an MLU in words of 4.92 and MLU in morphemes of 5.45. In this sample Luke produced 29 words and 20 different words (NDW), with his moving-average NDW being 20. According to the SALT Performance Report, the moving-average NDW of first graders was 51 and second graders was 56. (Miller & Nockerts, 2024). See Table 8 for a full description.

Quantitative

This section reports measures recorded for the various participant groups, including language scores, sensory processing as reported by the SSP, and anxiety indicators reported using the SCARED.

Between-groups comparison (Kruskal-Wallis One-Way ANOVA, with pairwise comparison of all groups) of the total score of our language measure revealed no significant differences between any of the groups in our sample (see Table 5). Of particular interest were how the cochlear implantees compared to each other, as seen in Table 6. Again, no significant differences were identified, and this pattern was consistent with both the expressive and receptive language scores as well.

When comparing the scores of our various groups to the normed scoring of the SSP, NACIs scored as having typical performance, NATHs scored as having a probable difference, and both autistic groups scored as having definite differences in sensory processing. Interestingly, the mean of those with autism who were not implanted showed greater sensory difficulties than those with autism who were implanted (see Table 4). The percentages of participants from the various groups showing a "probable difference" or "definite difference" in sensory processing abilities, according to the scoring of the SSP, can be seen in Table 7. Proportions testing showed that the ACI group presented with a trend of differing from the NATH group in the proportions of participants exhibiting a definite difference ($Z = 1.8$; $p = 0.07$). All other proportions tests revealed non-significant differences.

Sensory processing was significantly correlated with language measure scores in NATH participants ($r = 0.50$; $p = 0.00$). Notably, participants with CIs showed an SSP and language measure correlation of similar strength but that did not reach significance ($r = 0.62$; $p = 0.10$; see

Figure 4). The NATH participants also showed that receptive language was more strongly correlated with SSP ($r = 0.63$; $p < 0.001$) than expressive language ($r = 0.32$; $p = 0.034$). The CI users in our sample also showed this pattern, though their correlations were not statistically significant (receptive: $r = 0.44$; $p = 0.28$; expressive: $r = 0.24$; $p = 0.58$). Non-significant results in these cases are likely due to small sample size, even when CI participants were combined for correlational analysis. Taken together, these data support the idea that sensory processing and language outcomes, particularly related to receptive language, may be related.

On the measure of anxiety among participants, the ACI ($M = 21.83$; $SD = 18.7$) group differed significantly from the NATH group ($M = 8.88$; $SD = 7.80$; $U = 7.5$; $p = .037$). The proportions of subjects in each group that scored above the cut off for a possible anxiety disorder were as follows: ACI-33%, NACI-25%, ATH-25%, NATH-3%. Proportions testing revealed that the ACI did not differ from either the NACI group ($Z = 0.27$; $p = 0.79$) or the ATH group ($Z = 0.27$; $p = 0.79$). However, the ACIs showed a significant difference in the proportion scoring above the SCARED cutoff score when compared with the NATH group ($Z = 2.57$; $p = 0.01$), suggesting elevated anxiety in the ACI subjects as a group.

Overall, these data suggest relationships between sensory processing and language, as well as differences in anxiety between our participant groups.

Qualitative

Interview responses from clinicians and parents were distilled into four key themes. Each theme is summarized using a participant quote with relevant subthemes described beneath it.

Theme 1: "[We're] talking about a population of autistic users who need more support."

The subthemes included advocacy (1.1), navigating the system (1.2), lack of professional support (1.3), and lack of professional training (1.4). Each subtheme will be discussed in further detail below.

Theme 1: Parent Perspective

Parent participants were extremely passionate about the struggle they experienced working to find care for their ACI child. Many noted the importance of, and challenges related to, *advocacy* (subtheme 1.1) and *navigating the system* (subtheme 1.2). They emphasized the role they had to fill, not only as parent for their child, but also as advocate and defender. "We've found the caregivers and experts to [provide services to our child] but it has been extremely challenging," (Quote 1) reported one parent, while another said, "Parents know their child more than any other person and I think that parent's voices get lost within the system" (Quote 2). Many parents, when asked about their journey with autism and hearing loss, became emotional, particularly regarding the loneliness they felt throughout their difficult journey to find access to care. One mother said, through misty eyes, "I don't want to start crying, but it was difficult" (Quote 3). Some parents even reported advocating for their clinicians to participate in continuing education and fighting for the clinician's employers, whether it be a medical center or school district, to fund and offer release time to receive the necessary training to care for their child.

> "I essentially convinced [the clinic] to [pay for further education] and I was like, you know, some of these concepts might be helpful for other kids in the program too." (Quote 4)

Navigating the United States healthcare system, such as finding providers and handling insurance, can be a challenge for anyone, but it can become particularly difficult for parents who

have a child with multiple disabilities. Parents from multiple U.S. states recounted similar stories related to this point. For instance, one parent, while talking about her extensive experience with children with special needs said, “I’m a mom that knows how to navigate [the system] and knows the language to put up the fight to get the right people at the table” (Quote 5). This quote not only emphasizes the work this parent has put into advocating for her child, but also implies that the system likely has negative impacts on other parents who are not familiar with the United States medical system, advocacy, and/or “fighting” for their child. One parent articulated this challenge particularly well, saying:

> What about the other hundred kiddos who don’t have an educated mom? Even my husband says all the time, “If it wasn’t for [parent name], I’d have been twiddling my thumbs 4 years ago, being like, what the hell do I do?” (Quote 6)

Notably, many of the parents who participated in this study relocated to find better access to care for their child, with many of them moving across state lines or leaving spouses for months or years to find their child access to care that was unavailable at home. This is another aspect of navigating the system that is more extreme for the ACI population than some.

As children grow, navigating the healthcare system often also turns into navigating the school system. One parent noted that even when their child was provided with a 504-plan, granting them some accommodations, they still struggled to know how to use them or what they could continue requesting of the school. One parent said, “The school is kind of like, well, if she needs something, just let us know. It’s hard for her to … remember what her accommodations are to ask [to use them], and she feels embarrassed” (Quote 103). This parent further emphasized the difficulty they encounter when a child has accommodations, but teachers are not proactive about helping them access those saying:

> One of her accommodations is to sit close to the front and I talk to all her teachers at the beginning of the year and her math teacher was like, 'Well, she said she can hear me fine if she sits in the back of the class by the air conditioner,' and yet she has a D in math. (Quote 102)

Parents also emphasized that they experienced a *lack of professional support* (subtheme 1.3; i.e. speech pathologists, audiologists, physicians, behavioral therapist, autism providers, etc.). One parent reported, "There are not speech providers that understand hearing loss and autism and how to bring those two together" (Quote 7). Another said, similarly, "There really aren't services out there targeted I would say for kids with hearing loss and autism" (Quote 99). Another reported, "Someone has to advocate for these little guys to get the proper services because nobody … really knows what to do with them" (Quote 8). Still another said, "[It's hard] just finding that right therapist that knows how to tackle both components" (Quote 98). Another defended a clinician with whom she had had a poor experience saying, "It wasn't like she was like, "oh, [they're] autistic. But I'm not going to be flexible at all." I think she just didn't know what to do" (Quote 67). During our interview process, one parent even asked our research team if we had learned of any clinics that specialized in autism and hearing loss over the course of this study saying,

> It's a constant work-in-progress to try and find. I mean, I've looked all over. I don't know if you guys know of a program that specializes in working with kids with both of these things going on, like I don't. It seems like it's all kind of one or the other. (Quote 9)

Parents also reported difficulties with clinicians working together or giving them differing information. One said, "We had four therapists, and nobody was on the same page" (Quote 96).

Similarly, a parent said, “Everybody wanted to do their own approach. Everybody had their own expertise” (Quote 97).

In contrast to the above sentiments, some parents discussed the significant benefits associated with working alongside clinicians who had experience with the ACI population. Though finding these clinicians was often extremely challenging, some of the parents we interviewed were eventually able to find speech therapists, audiologists, and other professionals for their child who provided high levels of care. Their praise for these professionals highlights the relief and gratitude they felt upon finding someone that could help their child and the contrast to working with inexperienced clinicians. One parent said, “Along our journey we were lucky enough to have a strong audiologist team where it was [two provider names] and between the two of them they collaborated really, really well” (Quote 50). Another said, “The one person … that I had to really fight for was [provider name] and without [them], we would not be the parents that we are and [child name] would not be doing as well as [they are] without [their] expertise” (Quote 51). Another praised their speech therapist saying, “[They’re] very willing to learn … and [they’re] super motivated, too. I like [them] a lot” (Quote 52). Still another said, “I feel like the professionals that we have worked with have all been wonderful” (Quote 53).

These subthemes emphasize the point that parent advocacy for their children seemingly never ends. Parents are desperate for providers who understand both autism and hearing loss and how they interact with each other. Additionally, parents seek easier access to professionals with knowledge and experience working with ACIs.

Theme 1: Clinician Perspective

The clinicians interviewed had many similar perspectives as parents on some of the above points, albeit with some key differences. Many of the clinicians we interviewed

recognized that parents struggle to find high quality access to care for their children, with one reporting, “[We’re] talking about a population of autistic users who need more support” (Quote 10). Professionals also highlighted the *lack of professional training* (subtheme 1.4) and preparation received in graduate programs with regards to treating children with multiple disabilities. One audiologist described their training with ACIs as, “I think unless you do some extra program, you’re not going to get a lot at all” (Quote 11), while an SLP described their trainings with the statement, “We don’t get a lot of instruction in autism in our graduate programs. We don’t get a lot of instruction on hearing loss in our graduate programs” (Quote 12). Another clinician reported, when asked if they felt that graduate students were trained appropriately on how to work with ACIs, “If you just do the general grad program, no, I’d probably say not” (Quote 13). Another said, “I definitely was not prepared for these kids who have ASD and CIs” (Quote 14).

Of course, these clinicians and the authors of the present study recognize that it is impossible for graduate programs to teach their students everything about every condition, along with their numerous variations and the various ways they can present in clinical practice. One clinician put this dilemma well saying, “When you think about SLPs and everything they have to learn in a 2-year program, then you can’t really go very deep because you have to go really broad” (Quote 15). To this end, we asked our clinicians about continuing education and how they might improve the training of future clinicians. One replied, “I don’t know that that’s something really that can be changed other than the professionals taking upon themselves the opportunity to specialize” (Quote 16). Another said:

> How do we make continuing education really valuable in our specialties? And really accessible, making sure that if you’re gonna be working with children who have autism,

> you are able to get more training because you're probably not gonna be able to get it in your AuD program (Quote 17).

Another clinician emphasized the importance of experience when working with this population saying, "On the job experience is … big and I think finding somebody to … mentor you that has those skills, and that experience is … big too" (Quote 18).

The clinicians we interviewed did recognize the immense impact that their training and experience, or lack thereof, would have on their clients. One said, "If you're not good at what you do, that kid's gonna pay for it" (Quote 19), and another said, "your familiarity with autism and um yeah, your skills, your experience, with that does … play a part absolutely. That's a big part of the kid's success, which definitely puts a lot of pressure on you" (Quote 20).

Theme 1: Conclusion

While their perspectives are different, both the parent and clinician participants recognized the struggle that comes with finding services for an ACI and their family. Parents feel strongly about access to care. Clinicians, meanwhile, recognize the deficits in their fields and are doing the best they can. Clinicians additionally recognize that training could be improved but seem to have little insight on how to make reasonable adjustments to graduate programs or postgraduate training.

Theme 2: "There's a few hurdles that we oftentimes run into, sensory … being a huge one."

The subthemes included sensory averse (2.1), sensory seeking (2.2), behaviors & dysregulation (2.3), and the struggle to treat (2.4) Each subtheme will be discussed in further detail below.

Theme 2: Parent Perspective

Concerning sensory processing, parents spoke about encouraging their child to wear their devices, the sensory profiles of their individual children, and school participation. A primary parent concern was how they could encourage their child to keep their implant on, particularly for those with sensory aversions (sub-theme 2.1) to things being on their head. One parent reported, "[Child name] wears [their] implants Neptune style. So, clips on the back [because they] have sensory adverse behaviors over the ear" (Quote 22). Another, whose child has struggled to wear their implants said, "It's a work in progress right now, getting them over the ear" (Quote 23). Yet another said, "[They're] definitely sensory aversive when it comes to touch" (Quote 25). In this respect, some parents praised their clinicians saying, "[The audiologist] turned on [their] implants and programmed them and mapped them out and really set [child's name] up to be successful and not have sensory overload to [their] environment" (Quote 24).

Of note was that most of our participants were *sensory seeking* (subtheme 2.2). This means that they enjoy and often seek out sensory stimulation (Dunn, 1997). One parent labeled her child very clearly saying, "[They're] a seeker" (Quote 29). Another described their child's response to sensory input saying, "There's not a whole lot that [they're] super averse to" (Quote 30). Another parent noted that this sensory seeking tendency was having a direct influence on her child's CI use patterns saying, "If it coils off, you know [they try] to put it back on, [they don't] like it when they're not on" (Quote 31).

One such point of emphasis included how sensory differences were affecting their child's *behaviors* (subtheme 2.3) and subsequent participation in school. One parent said, referencing sensory processing, "[Their] hearing loss is not affecting [their] academic career, [their] autism

component is" (Quote 21). Another noticed that sensory regulation was vital to her child's success in school saying, "I've even asked the school to, I've had a letter from an OT to say, you know, if you can incorporate [sensory activities] in the morning and afternoon, that would be great" (Quote 28). One parent noted an accommodation provided to their child that has made a big difference in regulating their sensory differences and helping them have better success in school. "[Child's name] has a special chair with a TheraBand at the bottom of it where [they're] kicking it instead of sitting on the carpet kicking [their] peers" (Quote 32).

Other parents reported sensory difficulties that didn't necessarily or directly influence implant use. However, given that sensory difficulties are associated with maladaptive behaviors and anxiety (among other characteristics), we report them as possibly having indirect effects on CI use. Some examples include, "Hands have to be clean. [Child's name] doesn't like wearing long sleeves and generally dislikes a lot of clothing" (Quote 26), or, "[They are] definitely aversive to overwhelming social environments" (Quote 27).

Theme 2: Clinician Perspective

In contrast to our parent participants, most of our clinicians seemed to have worked with ACIs who mostly exhibited sensory aversions. Many of them reported *the struggle to treat* (subtheme 2.4) brought on by sensory differences. "They certainly are challenging in terms of testing" (Quote 36), said one clinician, specifically referring to the difficulties that come with performing audiological testing with a patient prone to sensory hypersensitivities. Another clinician said, "Doing otoscopy on [ACIs] is impossible" (Quote 33). Other clinicians reported challenges during audiological testing including, "Most of the time, I'm convinced that what I'm getting is minimal response. Like that's not your true threshold, but I don't know what your true threshold really is" (Quote 35), and "They're certainly challenging, but I think it's rewarding"

(Quote 37). Another emphasized the importance of objective measures with this population saying, "[There's a] focus on objective testing, but then you run into those sensory difficulties, saying how much objective testing can I do or how much will they tolerate" (Quote 39). Still another reported, "We rely so much on speech perception testing, whether it be words, whether it be sentences, whatever that is. And very often, these kids are nonverbal or limited, or echolalic … and so it's like, how do we measure awareness?" (Quote 41).

Another provider discussed the difficulty that sensory differences have not only on treatment, but on maintaining progress saying, "If there was a loud sound or something highly sensory, as far as auditorily speaking, that would put [the patient] kind of into a downward spiral. And we had to start over again" (Quote 34). When considering sensory differences in their treatment plans, many clinicians mentioned strategies that have worked. One said:

> The majority of my kids with autism prefer their off-ear processors, umm, and I don't know if it's because it's a little less of a tone hook touching their pinna … but I have four kids that just don't tolerate an on-the-ear processor and thankfully we have the ability to have an off-the-ear processor. (Quote 42)

Another clinician had a similar experience with an ACI who needed an off-the-ear processor, saying, "He wears that thing like 12 hours a day. As long as nothing is touching his ear" (Quote 44). Another said, concerning off the ear processors, "I think that's an important thing to like kind of keep in the back pocket too, is knowing that there are different styles" (Quote 49). Another strategy clinicians have reported includes, "We take the batteries out of the device. But just put it on the head, right. So there's no sound. And sometimes we start there" (Quote 46). Another strategy still was creating visuals to help ACIs know what to expect from their

appointment. This clinician said, “Especially with kids with autism who really, really, need and thrive better with that predictability being able to know what’s going on [is huge]” (Quote 101).

Clinicians have also noted a relationship between their sensory seeking and sensory adverse clients. For the sensory averse, one clinician pointed out, “It’s gonna impact wear time. It’s gonna impact compliance of use” (Quote 45). Another said:

> We certainly have kids who are on the spectrum, who have sensory defensiveness that is not related to sound, which is not impacting their … spoken language. But I would say for sure, if you’re sensory defensive related to auditory input, then that’s going to be a problem. (Quote 47)

Another reported while reflecting on her caseload of sensory seeking versus sensory adverse clients and their language outcomes that “There’s got to be something to that. Those sensory seekers are more okay with more sound” (Quote 48).

Theme 2: Conclusion

Parents and clinicians have different focuses when it comes to sensory processing differences. While parents seem to focus more on what their child needs to be regulated and do well in school, clinicians are focused on how to mitigate the impact that sensory differences have on device use and treatment sessions. Both groups have noticed trends between sensory seeking and sensory averse ACIs, noting that it seems that the former are more likely to use their device and have improved language outcomes.

Theme 3: "These two worlds [autism and hearing loss] for the most part, don't get along very well."

The subthemes included diagnostic delays (3.1), inconsistent performance and missed milestones (3.2), behaviors and dysregulation (3.3), and counseling (3.4). In general, clinicians spoke more about this theme than parents.

Theme 3: Parent Perspective

For each of the children in our study, hearing loss was diagnosed prior to autism. This was due, in great part, to newborn hearing screenings capturing their hearing loss so early in life. When asked if they thought their child's behaviors were signs of autism, one parent said "[They have] the hearing loss. [They have] ADHD and [disruptive mood dysregulation disorder]. I kind of felt like any of the concerns that we were having could kind of be explained by some of these other diagnoses" (Quote 54). Another parent said, "[They] would have been diagnosed as autistic like you know a lot earlier had the hearing loss aspect been taken aside" (Quote 55). Another parent said of their dual diagnosis journey, "When there's so many overlapping things having to weed out and you know, oftentimes even the professionals didn't always know, I feel like it was a lot of guess and check" (Quote 56). This highlights the influence of *diagnostic delays* (subtheme 3.1) particularly in reference to autism.

Throughout their journeys, some parents reported *inconsistency in their child's performance* (subtheme 3.2) with respect to their hearing. One parent said, "[Their] sound booth testing didn't match [their] sedated ABR test" (Quote 57). Another parent said, "[They] would pick up on Ling sounds then [they] wouldn't pick up on Ling sounds. [They were] babbling the Ling sounds then [they weren't] babbling the Ling sounds" (Quote 58). Similarly, another parent said, "I was becoming more concerned, like, what's going on here, like [they] used to say, like,

at least a few things. And then [they] completely stopped" (Quote 59). Other parents did not report as much on inconsistency; however, clinicians emphasized this subtheme (see below).

Parents did, however, focus a great deal on developmental milestones. For some, missing milestones was their first indicator that their child may have something else interacting with their hearing loss. One parent said she remembers thinking, "There's something else going on here that's complicating this, [they] should be further along than this" (Quote 60). One parent reported "wasting" a year in early intervention because clinicians were working on hearing loss related skills when their child's communication difficulties "ultimately ended up being [the] autism component, umm, and not [the] hearing loss because the hearing loss component was ruled out because we knew [they were] hearing" (Quote 63). In this case, it appeared that the CI was functioning well and that spoken language difficulties were associated more closely with autism.

Other parents reported that they wished clinicians had been more milestone focused with their counseling. This effect surfaced in the interviews, perhaps, because milestones, for many parents, are how they compare their child's development to what is typical. For instance, one parent said, "[The clinicians] never really told us how far behind [they were] or they never told us, you know, they just say 'Oh [they're] making progress and that's what we want to see'" (Quote 61). Another parent had a similar concern saying, "The slope of the trajectory was never part of the conversation. And they were always very, very vague" (Quote 62).

Also related to counseling is parent coaching. One parent reported her experience during her child's therapy sessions saying, "There was zero parent coaching. There was zero, like, telling me what to do at home. So, I'd just be sitting there" (Quote 100).

Regarding *dysregulation* (subtheme 3.3), parents reported having difficulty knowing if their child's behaviors stemmed from access to sound issues, sensory issues, or something unrelated. One parent said about their child's behavior:

> [They] would bang [their] head … [they] would spin in circles … behaviors as far as hitting, kicking, biting, hair pulling, head banging, at that point, [they were] throwing [their] hearing aids and actually right after for about 6 months or so, [they were] throwing [their] implants too. (Quote 64)

Another parent reported:

> [They're] in this, like, screaming stage.… It's not associated with how his implants are programmed or the environment of it being too loud or not. We've done so much … to ensure that it's not a sound input sensory behavior. It's an autism attention seeking behavior. (Quote 65)

These quotes highlight the difficulty that parents experience knowing if the behaviors their child is displaying are related to sensory difficulties inherent in autism or associated with hearing loss or their implant. In addition to sensory differences being associated with autism, sensory issues are more common in children with hearing loss than their typical hearing peers (Alkhamra & Abu-Dahab, 2020). Additionally, these quotes also hint at the possibility that such behaviors are a form of communication. Regarding her frustration with trying to understand her child as best she can and find them the support they need, one parent said, "These two worlds for the most part, don't really get along very well" (Quote 66).

Theme 3: Clinician Perspective

Like parents, clinicians also reported difficulties with diagnostic delays, as well as with inconsistent performance during assessment and treatment, milestones, and behaviors and dysregulation.

Concerning *diagnostic delays* (subtheme 3.1*)*, clinicians reported both noticing signs of autism as well as trying to provide services to a child who didn't have a diagnosis yet. "Some of these kids are not getting diagnosed with autism until well after their hearing loss has been diagnosed, potentially after they're implanted, depending on age of implantation" (Quote 78). One clinician said, referencing treatment for hearing loss beginning so early for most children following a newborn hearing screening, that "the real obvious autism signs don't show up, you know, at 2 months old or whatever" (Quote 81). Another said, "I would say more often than not, we know that they have a hearing loss before we know they have autism" (Quote 82). When asked if knowing about an autism diagnosis makes a difference in how a clinician moves forward with treatment, one said, "I wish I could say that I had ever gotten a child that I had that benefit" (Quote 75). Conversely, one said that when a child comes to their clinic having already been implanted, "I think that when I already have a diagnosis of autism for a patient, I think it makes it easier" (Quote 69).

Given the similarity of presentation between autism and hearing loss, one clinician noted why diagnostic delays may happen with this population. They said, "[That child] is already carrying [a hearing loss] diagnosis. So, until they start to see things that don't look like hearing loss then we don't really make any headway in appropriate [autism] diagnostics" (Quote 71). Still another said, "Children have hearing loss so much earlier oftentimes professionals with

good intentions default to this is a behavior indicator that is associated with hearing loss" (Quote 70).

Another clinician's opinion differed, reporting that they felt rather confident in their field's ability to recognize when something else was at play saying, "Those of us who know what hearing loss looks like, we watch them for a couple of days and we think, "Mmm, there's more going on here" (Quote 72). Another clinician commented on the importance of having good information because of the similar presentations saying:

> They do always show similar. So, I always tell [families], I need you to have a good hearing test done first, because if you don't have access to the language, a lot of the behaviors that we'll see mimic each other. (Quote 77)

When asked about the impact of newborn hearing screenings on diagnostic delays for autism, one said, "Does early newborn hearing screening, early intervention help or hurt [the autism] diagnostic process? I think it depends … generally we're moving to the implantation before we're moving to the diagnosis of autism because of early intervention" (Quote 73).

Clinicians also presented a theme of having trouble knowing how or when to *counsel* (subtheme 3.4) parents about autism. One said, "It's hard for us to say, 'Well, I can kind of help you understand why your child won't be speaking, because I know he has autism,' but nobody has gone that far to tell you that yet" (Quote 74). Another shared a similar sentiment saying:

> Sometimes, you know [they have autism], even before the parent knows which is hard. Because then you feel like you're keeping a secret from them when you're not. You just, it's not your place. It's not, you know, you're there to treat the hearing loss. You're there to support in whatever way you can. (Quote 68)

Another clinician reported that, “I’ve had families that did not [have an autism diagnosis], and I had to counsel families saying, ‘This is not my specialty, but of the behaviors that I’m noticing, I think it would be good to get a developmental evaluation’” (Quote 76).

One clinician reported optimistically that, “As time has passed, we have seen both [diagnosis for hearing loss and autism] move younger and younger” (Quote 79). Another said, “One of the things that we’ve seen for sure is younger and younger diagnosis of both and earlier intervention for both” (Quote 80).

Theme 3: Conclusion

The similar presentations of autism and hearing loss is challenging both for the parents and the clinicians. Counseling is something that parents wished had been done better, and something that many clinicians reported feeling unsure about. Happily, diagnosis and early intervention are improving for both disorder subtypes.

Theme 4: “The biggest thing [ACIs] have in common, is that they have nothing in common.”

Subthemes included blanket statements (4.1) and imposed limitations (4.2). Each subtheme will be discussed in further detail below.

Theme 4: Parent Perspective

Parents recognized the heterogeneity of autism, and some pushed back against the stereotypical presentation or *blanket statements* (subtheme 4.1) that accompany this diagnosis. One said, “I don’t think that there should be a stereotypical autistic person because you know, autism is such a [spectrum]” (Quote 83). Similarly, one parent said, “Autism is so different to begin, like it’s, you know, the characteristics present differently” (Quote 85). Another reported on how clinician’s expectations of what autism looks like has affected them saying, “[My child]

wasn't what they envisioned as, you know, an autistic kid and so, um, they just thought, you know, [they're] being difficult" (Quote 84).

Other parents had very strong feelings on the *limitations imposed* (subtheme 4.2) on their child by some clinicians or clinics based on their dual diagnosis. Surprisingly, various parents reported experiences in which some clinicians or centers were hesitant to provide autistic children that met audiologic CI candidacy requirements with implants, due to what they expected would be a poor outcome. One parent said, "[There's] implicit bias. I think they really are big on wanting their numbers to look good of success" (Quote 86). Another with a similar experience said:

> It's rage-provoking in me that [centers] are denying these kids the opportunity [to have access to sound]. Like why? Even if they don't have expressive language in the way that they want or define it, why don't they have the opportunity to have access to sound? (Quote 89)

This passion, resulting from recognition that their children were on a spectrum and capable of learning deeply, even if outcomes didn't match pre-defined norms, influenced parent's *advocacy* (sub theme 1.1) described in Theme 1.

Theme 4: Clinician Perspective

Clinicians were also quick to comment on the wide variety of skills and abilities they have seen among their ACI patients. One said, "I think because [autism and hearing loss] are a spectrum, [language development] just really depends on the child" (Quote 87). Still another said, "Putting them into one group is quite a challenge" (Quote 95). Another stated eloquently, "The biggest thing they all have in common, is that they have nothing in common" (Quote 88).

All the clinicians in the present study were passionate about providing high quality care and access to sound for all their patients. Unfortunately, like the parents, some had heard of clinics or providers who do not share their same fervor and instead counseled parents against cochlear implantation for their autistic child. One said she feared the following attitude was prevalent among professionals:

> A lot of typical hearing kids that have autism end up signing anyways or using an AAC device anyways, so it's kind of like why are we going to put [autistic children] through this surgery if we're not even going to be able to measure benefit? (Quote 38)

Another clinician shared a similar sentiment saying, "I think they're a population that probably gets written off very quickly" (Quote 40). One clinician said that while imposing limitations is not best practice, it's still important to create reasonable expectations for parents. Regarding counseling on language outcomes, another provider said, "It would be just more of a cautious prognosis because communication is already compromised as a part of the autism diagnosis" (Quote 90). Another clinician had a differing opinion saying, "Reasonable expectations … is such a stupid phrase" (Quote 91). She continued by saying, "We always want to presume confidence" (Quote 92). Another clinician agreed with this sentiment saying, "Don't limit yourselves. Don't predisposition the families" (Quote 93).

In contrast to some of those attitudes, our clinicians for the most part seemed to be grappling with their own implicit biases or expectations when they treated ACIs. One said, "I am not oblivious to the fact that there's plenty that I can still [learn]. But I also have intended to learn as much as I can and to reach out to people um who I know have more experience than I do" (Quote 94).

Theme 4: Conclusion

Both parents and clinicians agree that it is unfair to limit ACIs based on the former diagnosis, particularly because both autism and hearing loss have such variability. Unfortunately, parents and clinicians both are aware of individuals or clinics where ACIs are not being provided appropriate (or any) care because of implicit bias and concerns about what success may look like in their population.

Discussion

The present study aimed to examine why spoken language outcomes seem to be poorer in ACIs compared to NACIs and ATH individuals. Through analysis of both quantitative and qualitative data, we found that finding access to appropriate support, sensory differences, diagnostic and treatment difficulties between autism and hearing loss, and the nature of existing along two spectra all have an influence on language outcomes in this population. The following discussion will elaborate on the above points, triangulating quantitative and qualitative data, as well as provide recommendations for future researchers and clinicians as they continue working with this important, but understudied, population.

Access to Appropriate Support

Overall, our ACI participants exhibited favorable language outcomes. This is partially in contrast to the existing literature (Donaldson et al., 2004; Eshraghi et al., 2015; Jenks et al., 2022; Lachowska et al., 2018; Mikic et al., 2016). Our data showed that one of the most influential factors for the language outcomes of our participants was intense family support. In general, our results are in keeping with previous findings that family support significantly impacts successful cochlear implantation (Black et al., 2012). However, given the high level and unique types of support needs of our ACI subjects, the limited availability of trained

professionals in their local communities (subthemes 1.3 and 1.4), and difficulties navigating both the medical and school systems (e.g., use of IEP / 504 plans; subtheme 1.2), family support that exceeded typical levels was needed. Our qualitative Theme 1 emphasized the importance of parent advocacy (subtheme 1.1) for our ACI families and how access to care would have been different had the parents not been extremely involved. Additionally, clinician survey responses indicated that "Parent Compliance with Rehabilitation Efforts" ranked fourth out of ten possible factors that influence language development and treatment (see Figures 2 and 3), providing further evidence for the important role of family support for positive outcomes in ACIs.

The parents in this study went to great lengths to find their child support (e.g., advocating for further education for providers, relocating to areas with better services). The positive language outcomes, both self-reported and present in our quantitative analysis, are likely underpinned by the advocacy and support provided to these children by their parents. In contrast to the existing literature and recommendations, the current study showed that when parents were able to find experienced providers, their children were successful and responded well to treatment. It should be noted that not all parents may have the means or resources to provide that same care or opportunities for their children. This may influence ACI language development and help explain why the ACIs in this study had better use of oral language than ACIs in other studies previously mentioned (i.e., Donaldson et al., 2004; Lachowska et al., 2018; Mikic et al., 2016).

Clinician experience and skill is another vital aspect of care, perhaps especially for ACIs. Throughout our interviews, parents reported a lack of professional support (subtheme 1.3), and clinicians reported a lack of professional training and preparation from their graduate programs (subtheme 1.4). Lack of professional training and preparation is a theme reported in the literature

as influencing the treatment of individuals with multiple disabilities, including autism and hearing loss (Bono et al., 2022; McFayden et al., 2023). Many of our clinicians indicated that they felt unprepared and often unsure of how exactly to support ACIs (Quotes 12, 13, 14, 15). They also indicated the importance of gaining experience and participating in continuing education so that they could be better suited to provide high quality treatment to these patients (Quotes 16, 17, 18). Interestingly, while both parents and clinicians mentioned that clinician experience was key; clinicians ranked their lack of training and inexperience as being seventh and eight respectively out of ten factors that affected language and treatment outcomes (see Figures 2 and 3). Taken together, these findings highlight the immense influence that access to services has on language outcomes, particularly with ACIs.

The difficulty reported by parents and clinicians with access to care caused us to speculate about other autistic children with hearing loss who may not have the same support as the children in this study did. Caregivers in this study, for example, went to great lengths to find support for their children, some went so far as to relocate. Caregivers in this study were also highly educated, familiar with the healthcare system, native English speakers, and of middle to upper socioeconomic status (SES). For other parents, however, providing their child quality access to care may prove difficult. Low SES, unfamiliarity with the healthcare system, or in some cases limited English proficiency are just a few potential barriers. These, and other, barriers may make it extremely difficult or impossible for some caregivers to provide high quality care for their child, even if they wanted to. In turn, such phenomena could have a significant impact on their child's outcomes.

Sensory Differences

Sensory processing difficulties are more common among those with hearing impairments and particularly among CI users (Alkhamra & Abu-Dahab, 2020; Bharadwaj et al., 2009) and are a fundamental characteristic of autistic individuals (American Psychiatric Association [APA], 2013). Findings from our proportions testing were consistent with these previous reports. Additionally, sensory processing and language outcomes have a known relationship (Cooper, 2021; Marco et al., 2011). This association was evident in the outcomes of our ACI participants. Clinicians ranked sensory processing difficulties as the second and third most impactful factors on treatment and language development. Patient behavior and cooperation during sessions was ranked as the most influential factor in treatment and language development, which may also have links to sensory differences (Gundogdu et al., 2023). Both quantitative data from parent surveys and qualitative interview results suggest a relationship between sensory processing and language development. Furthermore, correlational analysis in both NATH and NACI participants suggested a trend toward significant association between sensory difficulties and language outcome.

In conjunction with the general relationships between sensory difficulties and language development, our analysis uncovered a novel finding which, to our knowledge, has not been reported previously in the literature. That is, several clinicians commented on their observations that ACIs who tend to be “sensory seeking” (subtheme 2.2) have better language outcomes than those who are “sensory averse” (subtheme 2.1). Further support for this finding is evident in the success of our ACI participants in developing spoken language, and their parents’ own evaluations of these participants as having sensory seeking tendencies. It may be that people who exhibit sensory seeking behaviors/needs are more apt to accept and want auditory stimulation via

a CI, leading to more consistent use, whereas those who have sensory hypersensitivities are more prone to find such stimulation uncomfortable, overwhelming, or otherwise undesirable, leading to diminished or non-use. Given these converging findings, we propose that it might be beneficial for AuDs and SLPs to use sensory screenings in their practices. These screenings could help clinicians know what to expect and how to plan intervention that would best suit their client's sensory profile. These screenings could also give clinicians additional insight into what future language development may look like and could help clinicians improve their counseling efforts. Alternatively, or additionally, CI teams should engage in interdisciplinary collaboration with occupational therapists when autistic persons are exploring cochlear implantation as a treatment option.

Autism and Hearing Loss as Dimensions, not Categories

Unfortunately, diagnostic delays of autism are common among those with hearing loss as reported by our clinician and parent participants, and as reported in the literature (Mandell et al., 2005). However, many of the ACI participants in our sample received autism diagnoses quickly following implantation and were able to begin receiving services immediately. This timing was due, in large part, to the advocacy of their parents. Delays in diagnoses also often create deferment of some services, which, in turn, can impact a child's progress. Early intervention seems to be a key to ACI language development (Yoshinaga-Itano et al., 1998).

One reason for differences during diagnosis in autistic individuals with hearing loss that has been discussed both previously (Mandell et al., 2005; Szarkowski et al., 2014; Tavares et al., 2021) and in the present findings is that autism and hearing loss both exist along spectra that overlap with each other (National Institute on Mental Health Research Domain Criteria; RDoC; Cuthbert & Insel, 2010). That is, autism and hearing loss share many characteristics, such as

language delays, failure to respond to one's name, and pragmatic difficulties, among others (Szarkowski et al., 2014). In addition, such individuals can have varied degrees or presentations of both conditions. As such, each ACI is an individual with unique characteristics across multiple dimensions (e.g., autistic traits, sensory differences, anxiety, hearing loss, etc.; Dwyer, 2022; Kotov et al., 2017). Unfortunately, the general, categorical statements and limitations imposed both by the existing literature and some clinicians upon this population are overwhelmingly negative (Donaldson et al., 2004; Lachowska et al., 2018; Mathew et al., 2022). To us, this trend is curious, since the ACIs in the current study all developed fluent oral language, apart from one who experienced a regression following a traumatic experience, which led to stark reduction in CI use. Even the latter participant had reportedly developed strong spoken language abilities prior to this incident. Our assertions about positive oral language outcomes are supported by parent questionnaire responses, language samples from three of our five ACI participants, and qualitative interviews. Moreover, data from the Auditory Skills Checklists that we completed with our participants suggest a high level of auditory skills in the majority of our ACI participants. It is possible that our methodology, since it was different than previous studies, may have contributed to the notable differences in results reported here compared to other studies. Some of those differences include the use of language sampling and parent report rather than standardized audiologic and language assessments to assess ACI participant hearing and expressive/receptive language abilities. Additionally, recruiting methods may have influenced outcomes as some ACI caregiver and child participants were referred by their clinicians. Some clinicians may have exhibited bias in their referrals, perhaps referring only their best performing patients or clients. Conversely, clinicians may have referred parents to this study who they felt were most likely to participate which may have contributed to the demographics of our sample.

Though such methodological issues may have had some impact on the results, taken together, we believe that differences in methodology are not the major driver of the differences in results reported by the present study.

Importantly, and in answer to our original research question(s), taken together, our results suggest that spoken language development is more dependent on securing appropriate supports and autism-related characteristics than those associated with hearing loss or Cis themselves. Thus, in addition to measuring the language outcomes of our participants, the results reported herein provide insight about some of the most important factors associated with the performance of our sample of ACIs. Such notions may have bearing on the support of other ACIs. Overall, while we acknowledge that setting and maintaining reasonable expectations and using one's best judgement to make life-altering clinical decisions is essential, we submit that future researchers and practicing clinicians should recognize the high degree of variability among ACIs and realize that many can develop oral language, given appropriate support.

Limitations

While the present researchers aimed to be as thorough as possible, this study had several limitations. First among them was sample size, however, given the specificity of the group six participants is not unreasonably small. Notably, six ACIs is comparable to other studies in the literature (Donaldson et al., 2004; Hayman & Franck, 2005; Lachowska et al., 2018; Zaidman-Zait & Curle, 2018), a larger sample would have added depth and increased reliability and generalizability of our findings and added statistical power to the quantitative measures. To mitigate the effects of a small sample, quantitative statistics most appropriate for small sample sizes and non-normally distributed data were used. Additionally, ACI language samples and other qualitative information were treated more as "case studies" rather than as representative of

the population. Furthermore, we took great care to examine and report the mutual substantiation of our qualitative and quantitative data (i.e., triangulation).

In connection to being limited by size, this study was also limited by the age of ACI participants. Those participating in the study spanned a wide range of ages from 3–14 years old. With this wide a range, it is difficult to truly make comparisons between participants, especially in terms of language, due to the varied stages of development each participant is in. Additionally, none of our NACI participants were perfectly matched pairs for our ACIs creating additional difficulty with comparison. Thus, in all quantitative analysis in which it was possible, we controlled for age. Additionally, ACI participant language samples were not compared to each other. Rather, they were compared to previously collected developmental norms appropriate for their age.

We also recognize that all ACI participants were White and living in the United States, and that their caregivers were highly educated. Future studies would benefit from participants from varying cultures and ethnic backgrounds, as well as differing socio-economic backgrounds.

Access to audiological files and cognitive abilities was another limiting factor of this study. To compensate, participants were given the Auditory Skills Checklist and other basic audiological, clinical, and school related history was collected from the ACI parents during interviews. Additionally, data collected from the parents during interviews suggested that autism was more related to their children's difficulty with language development than the particulars of their hearing loss.

Implications and Future Directions for Clinicians and Other Stakeholders

Based on the results of the current study, we urge fellow clinicians to presume patient competence when working with the ACI population. This study demonstrated that oral language

development is possible given the right circumstances, and perhaps, given a sensory seeking profile. We recommend that clinicians utilize sensory screeners in their assessment and intervention to help inform their parent counseling. Additionally, we recommend that clinicians participate in continuing education if they work with children in this population. Continuing education may also improve the rates of diagnosis among children with autism and hearing loss and help to mitigate diagnostic delays that can be brought on by the overlapping of characteristics present in both disorder groups.

The literature concerning the ACI population is sparse. Thus, continued research in this area is of great import. The present results warrant further exploration into the impacts on ACIs of sensory processing and subtypes. Additionally, further investigation of language outcomes in a larger sample of ACIs using language samples would be extremely interesting, particularly if children with various sensory subtypes were available to participate in the study. Another interesting facet of this study may include comparing autistic traits to language outcomes dimensionally.

Conclusion

To date, the current literature states that spoken language development is unrealistic in ACIs, or at least unlikely. Given the immense success of spoken language in cochlear implant users, as well as the reported ability of autistic individuals to develop oral language, this finding was surprising to us. The current study aimed to examine spoken language outcomes in autistic cochlear implant users and why they differ from the ATH and NATH groups using both quantitative and qualitative methods. We found that access to appropriate care and sensory differences have a significant impact on language development outcomes. We also found that ACIs come from an extremely diverse population and that blanket statements should not be used

to define the population, and that spoken language outcomes do seem to be positive in many ACIs. This has implications for future clinicians and researchers who will work with ACIs. Clinicians working with this population would do well to seek additional training and recognize that sensory differences may influence language outcomes and use this information to help them counsel families appropriately. Future researchers would benefit from examining language outcomes in ACIs with a variety of sensory profiles and in a larger group.

References

Alkhamra, R., & Abu-Dahab, S. (2020, September). Sensory processing disorders in children with hearing impairment: Implications for multidisciplinary approach and early intervention. *International Journal of Pediatric Otorhinolaryngology, 136*. https://doi.org/10.1016/j.ijporl.2020.110154

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). https://doi.org/10.1176/appi.books.9780890425596

American Speech-Language-Hearing Association. (2004). *Cochlear implants* [Technical Report]. Available from https://www.asha.org/policy/tr2004-00041/

American Speech-Language-Hearing Association. (2020, June 3). *FDA approves cochlear implantation at 9 months - leader.pubs.asha.org*. ASHA WIRE. https://leader.pubs.asha.org/do/10.1044/leader.NIB3.25062020.11/full/

Baron, S., Blanchard, M., Parodi, M., Rouillon, I., & Loundon, N. (2019). Sequential bilateral cochlear implants in children and adolescents: Outcomes and prognostic factors. *European Annals of Otorhinolaryngology, Head & Neck Diseases, 136*(2), 69–73. https://doi.org/10.1016/j.anorl.2018.09.004

Bebko, J. M., Schroeder, J. H., & Weiss, J. A. (2014). The McGurk effect in children with autism and Asperger syndrome. *Autism Research*, *7*(1), 50–59. https://doi.org/10.1002/aur.1343

Bharadwaj, S. V., Daniel, L. L., & Matzke, P. L. (2009). Sensory-processing disorder in children with cochlear implants. *American Journal of Occupational Therapy, 63*(2), 208–213. https://doi.org/10.5014/ajot.63.2.208

Birmaher., B, Brent, D., Chiapetta, L., Bridge, J., Monga, S., & Baugher, M. (1999). Psychometric properties of the screen for child anxiety related emotional disorders (SCARED): A replication study. *Journal of the American Academy of Child & Adolescent Psychiatry, 38*(10), 1230–1236. https://doi.org/10.1097/00004583-199910000-00011

Black, J., Hickson, L., & Black, B. (2012). Defining and evaluating success in paediatric cochlear implantation – An exploratory study. *International Journal of Pediatric Otorhinolaryngology, 76,* 1317–1326. https://doi.org/10.1016/j.ijporl.2012.05.027

Blanc, F., Blanchet, C., Sicard, M., Merklen, F., Venail, F., & Mondain, M. (2022). Audiological outcomes and associated factors after pediatric cochlear reimplantation. *Journal of Clinical Medicine, 11*(3148), 2–9. https://doi.org/10.3390/jcm11113148

Bono, L. K., Haverkamp, C. R., Lindsey, R. A., Freedman, R. N., McClain, M. B., & Simonsmeier, V. (2022). Assessing interdisciplinary trainees' objective and self-reported knowledge of autism spectrum disorder and confidence in providing services. *Journal of Autism and Developmental Disorders*, *52*(1), 376–391. https://doi.org/10.1007/s10803-021-04948-3

Bottema-Beutel, K., Kapp, S. K., Lester, J. N., Sasson, N. J., & Hand, B. N. (2021). Avoiding ableist language: Suggestions for autism researchers. *Autism in Adulthood*, *3*(1), 18–29. https://doi.org/10.1089/aut.2020.0014

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77–101. https://doi.org/10.1191/1478088706qp063oa

Braun, V., & Clarke, V. (2021). One size fits all? What counts as quality practice in (reflexive) thematic analysis? *Qualitative Research in Psychology, 18*(3), 328–352. https://doi.org/10.1080/14780887.2020.1769238

Brown, T. M., Baas, B. S., Stoeckel, R. E., Belf, L. A., & Poling, G. L. (2021). Assessment of children with hearing loss and co-occurring medical disorders: Challenging cases. *Perspectives of the ASHA Special Interest Groups, 6*(2), 375–383. https://doi.org/10.1044/2021_PERSP-20-00080

Camarata, S. (2013). Pediatric hearing impairment, autism, and Autism Spectrum Disorder: Implications for clinicians. *Perspectives on Hearing and Hearing Disorders in Childhood, 23*(1), 4–12. https://doi.org/10.1044/hhdc23.1.4

Constantino, J. N. (2013). Social Responsiveness Scale. In F. R. Volkmar (Ed.). *Encyclopedia of Autism Spectrum Disorders*. Springer, New York, NY. https://doi.org/10.1007/978-1-4419-1698-3_296

Cooper, C. (2021). *Atypical sensory processing and semantic language in autistic children.* [Master's thesis, Brigham Young University] BYU Scholars Archive. https://scholarsarchive.byu.edu/etd/9352

Crasta, J. E., Salzinger, E., Lin, M.-H., Gavin, W. J., & Davies, P. L. (2020). Sensory processing and attention profiles among children with sensory processing disorders and autism spectrum disorders. *Frontiers in Integrative Neuroscience*, *14*, 22–32. https://doi.org/10.3389/fnint.2020.00022

Cuthbert, B. N., & Insel, T. R. (2010). Toward new approaches to psychotic disorders: The NIMH Research Domain Criteria Project. *Schizophrenia Bulletin, 36*(6), 1061–1062. https://doi.org/10.1093/schbul/sbq108

Demopoulos, C., & Lewine, J. D. (2016). Audiometric profiles in autism spectrum disorders: Does subclinical hearing loss impact communication? *Autism Research*, *9*(1), 107–120. https://doi.org/10.1002/aur.1495

Donaldson, A. I., Heavner, K. S., & Zwolan, T. A. (2004). Measuring progress in children with autism spectrum disorder who have cochlear implants. *Proceedings from the Ninth Symposium on Cochlear Implants in Children, 130*(5), 666–671. https://doi.org/10.1001/archotol.130.5.666

Dunn, W. (1997). The impact of sensory processing abilities on the daily lives of young children and their families: A conceptual model. *Infants and Young Children, 9*(4), 23–35. https://doi.org/10.1097/00001163-199704000-00005

Dworzynski, K., Ronald, A., Bolton, P., & Happé, F. (2012). How different are girls and boys above and below the diagnostic threshold for autism spectrum disorders? *Journal of the American Academy of Child & Adolescent Psychiatry, 51*(8), 788–797. https://doi.org/10.1016/j.jaac.2012.05.018

Dwyer, P. (2022). The neurodiversity approach(es): What are they and what do they mean for researchers. *Human Development, 66*(2), 73–92. https://doi.org/10.1159/000523723

Eshraghi, A. A., Nazarian, R., Telischi, F. F., Martinez, D., Hodges, A., Velandia, S., Cejas-Cruz, I., Balkany, T. J., Lo, K., & Lang, D. (2015). Cochlear implantation in children with autism spectrum disorder. *Otology & Neurotology, 36*(8), e121–NaN. https://doi.org/10.1016/j.ijporl.2021.110876

Faistauer, M., Silva, A. L., Dominguez, D. d. O. R., Bohn, R., Félix, T. M., Costa, S. d. S., & Rosito, L. P. S., (2022). Does universal newborn hearing screening impact the timing of

deafness treatment? *Jornal De Pediatria*, *98*(2), 147–154. https://doi.org/10.1016/j.jped.2021.04.008

Feijóo, S., Foody, M., Pichel, R., Zamora, L., & Rial, A. (2021). Bullying and cyberbullying among students with cochlear implants. *Journal of Deaf Studies and Deaf Education, 26*(1), 130–NaN. https://doi.org/10.1093/deafed/enaa029

Gale, N. K., Heath, G., Cameron, E., Rashid, S., & Redwood, S. (2013). Using the framework method for the analysis of qualitative data in multi-disciplinary health research. *BMC Medical Research Methodology, 13*(117), 1471–2288. https://doi.org/10.1186/1471-2288-13-117

Ghaly, A., Nada, E., Rabie, S., Ragaey, Y., & Elkhatib, H. (2022, July). Assessing post-cochlear implantation anxiety and correlating it to socio-demographic and clinical characteristics. *Middle East Current Psychiatry, 29*, 1–5. https://doi.org/10.1186/s43045-022-00215-y

Glod, M., Riby, D. M., & Rodgers, J. (2019). Short report: Relationships between sensory processing, repetitive behaviors, anxiety, and intolerance of uncertainty in autism spectrum disorder and Williams syndrome. *Autism Research, 12*(5), 759–765. https://doi.org/10.1002/aur.2096

Gundogdu, U., Aksoy, A., & Eroglu, M. (2023). Sensory profiles, behavioral problems, and auditory findings in children with autism spectrum disorder. *International Journal of Developmental Disabilities, 69*(3), 442–451. https://doi.org/10.1080/20473869.2023.2200592

Hale, W. W., 3rd, Crocetti, E., Raaijmakers, Q. A., & Meeus, W. H. (2011). A meta-analysis of the cross-cultural psychometric properties of the Screen for Child Anxiety Related

Emotional Disorders (SCARED). *Journal of child psychology and psychiatry, and allied disciplines*, *52*(1), 80–90. https://doi.org/10.1111/j.1469-7610.2010.02285.x

Hamzah, N. F. A., Umat, C., Harithasan, D., & Goh, B. S. (2021, April). Challenges faced by parents when seeking diagnosis for children with sensorineural hearing loss. *International Journal of Pediatric Otorhinolaryngology*, *143*, Article 110656. https://doi.org/10.1016/j.ijporl.2021.110656

Hansen, S., & Scott, J. (2018). A systematic review of the autism research with children who are deaf or hard of hearing. *Communication Disorders Quarterly, 39*(2), 330–334. https://doi.org/10.1177/1525740117728475

Harmon, T. G. (2020). Everyday communication challenges in aphasia: Descriptions of experiences and coping strategies, *Aphasiology, 34*(10), 1270–1290. https://doi.org/10.1080/02687038.2020.1752906

Hayman, C., & Franck, K. (2005). Cochlear implant candidacy in children with autism. *Seminars in Hearing, 26(*4), 217–225. https:doi.org/10.1016/j.bjorl.2016.10.012

Homans, N. C., & Vroegop, J. L. (2021). Impact of face masks in public spaces during COVID-19 pandemic on daily life communication of cochlear implant users. *Laryngoscope Investigative Otolaryngology, 6*(3), 531–539. https://doi.org/10.1002/lio2.578

Hull, L., Petrides, K. V. & Mandy, W. (2023, January). The female autism phenotype and camouflaging: a narrative review. *Review Journal of Autism and Developmental Disorders 7*, 306–317. https://doi.org/10.1007/s40489-020-00197-9

Jenks, C., Hoff, S., Haney, J., Tournis, E., Thomas, D., & Young, N. (2022). Cochlear implantation can improve auditory skills, language, and social engagement of children

with autism spectrum disorder. *Otology & Neurotology*, *43*(3), 313–319. https://doi.org/10.1097/MAO.0000000000003463

Kotov, R., Krueger, R. F., Watson, D., Achenbach, T. M., Althoff, R. R., Bagby, R. M., Brown, T. A., Carpenter, W. T., Caspi, A., Clark, L. A., Eaton, N. R., Forbes, M. K., Forbush, K. T., Goldberg, D., Hasin, D., Hyman, S. E., Ivanova, M. Y., Lynam, D. R., Markon, K., & Zimmerman, M. (2017). The hierarchical taxonomy of psychopathology (HiTOP): A dimensional alternative to traditional nosologies. *Journal of Abnormal Psychology, 126*(4), 454–477. https://doi.org/10.1037/abn0000258

Lachowska, M., Pastuszka, A., Łukaszewicz-Moszyńska, Z., Mikołajewska, L., & Niemczyk, K. (2018). Cochlear implantation in autistic children with profound sensorineural hearing loss. *Brazilian Journal of Otorhinolaryngology*, *84*(1), 15–19. https://doi.org/10.1016/j.bjorl.2016.10.012

Lester, H. (1987). *Pookins gets her way.* Clarion Books.

Lord, C., Rutter, M., DiLavore, P., Risi, S., Gotham, K., & Bishop, S. (2012). *Autism diagnostic observation schedule–2nd edition (ADOS-2)*. Western Psychological Corporation.

Maenner, M. J., Warren, Z., Williams, A. R., Amoakohene, E., Bakian, A. V., Bilder, D. A., Durkin, M. S., Fitzgerald, R. T., Furnier, S. M., Hughes, M. M., Ladd-Acosta, C. M., McArthur, D., Pas, E. T., Salinas, A., Vehorn, A., Williams, S., Esler, A., Grzybowski, A., Hall-Lande, J., . . . Shaw, K. A. (2023). Prevalence and characteristics of autism spectrum disorder among children aged 8 years - Autism and developmental disabilities monitoring network, 11 sites, united states, 2020. MMWR. Surveillance Summaries, *72*(2), 1–14. https://doi.org/10.15585/mmwr.ss7202a1

Mandell, D. S., Novak, M. M., & Zubritsky, C. D. (2005). Factors associated with age of diagnosis among children with autism spectrum disorders. *Pediatrics, 116*(6), 1480–1486. https://doi.org/10.1542/peds.2005-0185

Marco, E. J., Hinkley, L. B. N., Hill, S. S., & Nagarajan, S. S. (2011). Sensory processing in autism: A review of neurophysiologic findings. *Pediatric Research, 69*(5 Pt 2), 48R–54R. https://doi.org/10.1203/PDR.0b013e3182130c54

Mathew, R., Bryan, J., Chaudhry, D., Chaudhry, A., Kuhn, I., Tysome, J., Donnelly, N., Axon, P., & Bance, M. (2022). Cochlear implantation in children with autism spectrum disorder: A systematic review and pooled analysis. *Otology & Neurotology, 43*(1), e1–e13. https://doi.org/10.1097/MAO.0000000000003353

Mayer, M. (1969). *Frog, where are you?* Dial Books.

McFayden, T. C., Culbertson, S., DeRamus, M., Kramer, C., Roush, J., & Mankowski, J. (2023). Assessing autism in deaf/hard-of-hearing youths: Interdisciplinary teams, COVID considerations, and future directions. *Perspectives on Psychological Science, 18*(6), 1492–1507. https://doi.org/10.1177/17456916231178711

Meinzen-Derr, J., Wiley, S., Bishop, S., Manning-Courtney, P., Choo, D. I., & Murray, D. (2014). Autism spectrum disorders in 24 children who are deaf or hard of hearing. *International Journal of Pediatric Otorhinolaryngology, 78*(1), 112–118. https://doi.org/10.1016/j.ijporl.2013.10.065

Meinzen-Derr, J., Wiley S, Choo D, Creighton J. (2004). *Auditory Skills Checklist (ASC)*. Cincinnati Children's Hospital Medical Center.

Mikic, B., Jotic, A., Miric, D., Nikolic, M., Jankovic, N., & Arsovic, N. (2016). Receptive speech in early implanted children later diagnosed with autism. *European Annals of*

Otorhinolaryngology, *Head and Neck Diseases*, *133*(Supplement 1), S36–S39. https://doi.org/10.1016/j.anorl.2016.01.012

Miller, J., & Iglesias, A. (2020). Systematic Analysis of Language Transcripts (SALT), Version 20 [Computer Software]. SALT Software, LLC.

Miller, J., & Nockerts, A. (2024). Systematic Analysis of Language Transcripts (SALT), Version 24 [Computer Software]. SALT Software, LLC.

Nicks, S., Johnson, A., Traxler, B., Bush, M., Brame, L., Hamilton, T., & Hartwell, M. (2022). The use of person-centered language in medical research articles focusing on hearing loss or deafness. *Ear and Hearing, 43*(3),703–711, https://doi.org/10.1097/AUD.0000000000001168

Paul, R., Norbury, C., & Gosse, C. (2018). *Language disorders from infancy through adolescence: Listening, speaking, reading, writing, and communicating.* Elsevier.

Pence Turnbull, K., & Justice, L. (2017). *Language development from theory to practice.* Pearson.

Peña, E. D., Gutiérrez-Clellen, V. F., Iglesias, A., Goldstein, B. A., & Bedore, L. M. (2018). *BESA: Inventory to Assess Language Knowledge (ITALK).* Agile Testing Solutions.

Phillips, H., Wright, B., Allgar, V., McConachie, H., Sweetman, J., Hargate, R., Hodkinson, R., Bland, M., George, H., Hughes, A., Hayward, E., De Las Heras, V., & Le Couteur, A. (2022). Adapting and validating the autism diagnostic observation schedule version 2 for use with deaf children and young people. *Journal of Autism & Developmental Disorders, 52*(2)*,* 553–568. https://doi.org/10.1007/s10803-021-04931-y

Rice, M. L., Smolik, F., Perpich, D., Thompson, T., Rytting, N., & Blossom, M. (2010). Mean length of utterance levels in 6-month intervals for children 3 to 9 years with and without

language impairments. *Journal of Speech, Language, and Hearing Research, 53*(2), 333–349. https://doi.org/10.1044/1092-4388(2009/08-0183)

Russell, G., Steer, C., & Golding, J. (2011). Social and demographic factors that influence the diagnosis of autistic spectrum disorders. *Social Psychiatry and Psychiatric Epidemiology*, *46*(12), 1283–1293. https://doi.org/10.1007/s00127-010-0294-z

Sharma, A., Dorman, M., & Spahr A. (2002a). Rapid development of cortical auditory evoked potentials after early cochlear implantation. *Neuro Report*, *13*(10), 1365–1368. https://doi.org/10.1097/00001756-200207190-00030

Sharma, A., Dorman, M., & Spahr, A. (2002b). A sensitive period for the development of the central auditory system in children with cochlear implants: Implications for age of implantation. *Ear and Hearing*, *23*(6), 532–539. https://doi.org/10.1097/00003446-200212000-00004

Simpson, K., Adams, D., Alston-Knox, C., Heussler, H., & Keen, D. (2019). Exploring the sensory profiles of children on the autism spectrum using the Short Sensory Profile-2 (SSP-2). *Journal of Autism and Developmental Disorders. 49*, 2069–2079. https://doi.org/10.1007/s10803-019-03889-2

Sönnichsen, R., Tó, G., Hohmann, V., Hochmuth, S., & Radeloff, A. (2022, November 27). Challenging times for cochlear implant users – Effect of facemasks on audiovisual speech understanding during the COVID-19 pandemic. *Trends in Hearing*, *26.* https://doi.org/10.1177/23312165221134378

Stern, J. A., Gadgil, M. S., Blakeley-Smith, A., Reaven, J. A., & Hepburn, S. L. (2014). Psychometric properties of the SCARED in youth with autism spectrum

disorder. *Research in Autism Spectrum Disorders*, *8*(9), 1225–1234. https://doi.org/10.1016/j.rasd.2014.06.008

Stevenson, R. A., Segers, M., Ferber, S., Barense, M. D., & Wallace, M. T. (2014, May 20). The impact of multisensory integration deficits on speech perception in children with autism spectrum disorders. *Frontiers in Psychology*, *5,* 379. https://doi.org/10.3389/fpsyg.2014.00379

Szarkowski, A., Mood, D., Shield, A., Wiley, S., & Yoshinaga-Itano, C. (2014). A summary of current understanding regarding children with autism spectrum disorder who are deaf or hard of hearing. *Seminars in Speech and Language 35*(4), 241–259. https://doi.org/ 10.1055/s-0034-1389097

Szymanski, C. A., Brice, P. J., Lam, K. H., & Hotto, S. A. (2012). Deaf children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *42*(10), 2027–2037. https://doi.org/10.1007/s10803-012-1452-9

Tager-Flusberg, H., & Kasari, C. (2013). Minimally verbal school-aged children with autism spectrum disorder: The neglected end of the spectrum. *Autism Research*, *6*(6), 468–478. https://doi.org/10.1002/aur.1329

Tavares, F. d. S., Azevedo, Y. J., Fernandes, L. d. M. M., Takeuti, A., Pereira, L. V., Ledesma, A. L. L., & Bahmad, F. (2021). Cochlear implant in patients with autistic spectrum disorder – A systematic review. *Brazilian Journal of Otorhinolaryngology, 87*(5), 601–619. https://doi.org/10.1016/j.bjorl.2020.11.020

Tomchek, S., Huebner, R., & Dunn, W. (2014). Patterns of sensory processing in children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 8*(9), 1214–1224. https://doi.org/10.1016/j.rasd.2014.06.006

van Steensel, F., & Heeman, E. (2017). Anxiety levels in children with autism spectrum disorder: A meta-analysis. *Journal of Child & Family Studies, 26*(7), 1753–1767. https://doi.org/10.1007/s10826-017-0687-7

Wiley, S., Gustafson, S., & Rozniak, J. (2014). Needs of parents of children who are deaf/hard of hearing with autism spectrum disorder. *The Journal of Deaf Studies and Deaf Education, 19*(1), 40–49. https://doi.org/10.1093/deafed/ent044

Yoshinaga-Itano, C., Sedey, A. L., Coulter, D. K., & Mehl, A. L. (1998). Language of early and later identified children with hearing loss. *Pediatrics*, *102*(5), 1161–1171. https://doi.org/10.1542/peds.102.5.1161

Zaidman-Zait, A., & Curle, D. (2018). Complexity: An interpretative phenomenological analysis of the experiences of mothers of deaf children with cochlear implants and autism. *Journal of Health Psychology, 23*(9), 1173–1184. https://doi.org/10.1177/1359105316646171

Zeng, F. G. (2004). Trends in cochlear implants. *Trends in Amplification*, *8*(1), 1–34. https://doi.org/10.1177/108471380400800102

Tables

Table 1

Clinician Demographics

	SLP	AuD	MD
n	4	8	1
Avg # of Years of Experience	20.75	11.38	8
SD Years of Experience	9.98	7.86	0
# of ACIs Currently Treating	**1:** 0 ACIs, **2:** 1-2 ACIs, **1:** 5-10 ACIs	**3:** 1-2 ACIs, **3:** 3-4 ACIs, **2:** 5-10 ACIs	**1:** 5-10 ACIs
Avg % of Low SES Status on Caseload	22%	36%	30%
SD % of Low SES Status on Caseload	8.6%	20%	0%
Confidence Level in ACI Treatment	1 Extremely Confident, 1 Very Confident, 1 Moderately Confident, 1 No Response	1 Extremely Confident, 6 Moderately Confident, 1 No Response	1 Extremely Confident,

Table 2

Description of Questions (by Question Number) – Caregiver Survey

Category	Scored	Descriptive
Expressive Language	Language 2-5, Language 7	Language 1
Receptive Language	Language 6, Language 8	--
Autistic Traits	SRS	--
Anxiety	SCARED	--
Sensory	SSP	SSP 5-6
CI Usage	--	Demographics 10-12

Table 3

Description of Questions (by Question Number) – Caregiver Interview

Category	Scored	Descriptive
Expressive Language	2, 4, 5, 8	--
Receptive Language	--	Auditory Skills Checklist
Anxiety	2	--
Autistic Traits	2, 6	--
Sensory	2, 6, 9	--
CI Usage	2, 3, 4, 5, 10	--

Table 4

Comparison of Sensory Scores Between Groups: Means (M) and Standard Deviations (σ)

	ACI-M(σ)	NACI-M(σ)	ATH-M(σ)	NATH-M(σ)
SSP Total	119.83(30.255)	167.25(5.377)	100.25(18.751)	144.69(32.694)

Table 5

Comparison of Total Language Scores Between Groups

	ACI-M(σ)	NACI-M(σ)	ATH-M(σ)	NATH-M(σ)
Language Total	29(7.043)	34.25(1.5)	17(9.201)	27.11(10.02)

Table 6

Comparison of Language Scores Between Cochlear Implant Users

	ACI-*M*(σ)	NACI-*M*(σ)	*p*	*U*
Language Total	29.00(7.04)	34.25(1.5)	0.26	18.0
Expressive Language	18.83(2.71)	17.75(2.06)	0.476	8.5
Receptive Language	7.67(2.81)	9.00(0.82)	0.476	8.5

Table 7

SSP Categories by Group

Group	*n*	Definite Difference	Probable Difference	Typical
ACI	6	67%	17%	17%
NACI	4	0%	0%	100%
ATH	4	100%	0%	0%
NATH	35	29%	29%	41%

Table 8

ACI Participant Summary

Measure	Owen	Claire	Nora	Sophie	Luke
Sex	M	F	F	F	M
Age	04:02	14:03	03:06	05:06	07:08
CI Implant Age	21 mos	84 mos	9 mos	11 mos	9 mos
CI Experience	39 mos	100 mos	30 mos	57 mos	82 mos
Daily Wear Time	13 hrs	15 hrs	10 hrs	10 hrs	16 hrs
Primary Comm. Method	Oral	Oral	Oral	Total Comm	Oral
Aud. Skills Checklist	68	66	58	2	65
SRS	54	94	--	92	106
SSP	170	144	100	104	94
SCARED	7	32	22	10	54
Language Total	31	29	24	17	29
% Intelligible Utterances	60%	95.1%	--	--	100%
% Intelligible Words	88.4%	99.2%	--	--	100%
MLU in Words	4.95	6.53	--	--	4.83
MLU in Morphemes	5.45	7.31	--	--	5.17
% Utterances with Verbs	77.3%	87.3%	--	--	83.3%
Number of Total Words	109	359	--	--	29
Number of Different Words (NDW)	46	157	--	--	20
Mean Moving-Average NDW	43	62	--	--	20
% Utterances with Errors	38.8%	1.6%	--	--	0%
Length of Sample	10:26	03:28	--	--	01:11

Figures

Figure 1

Description of Phases

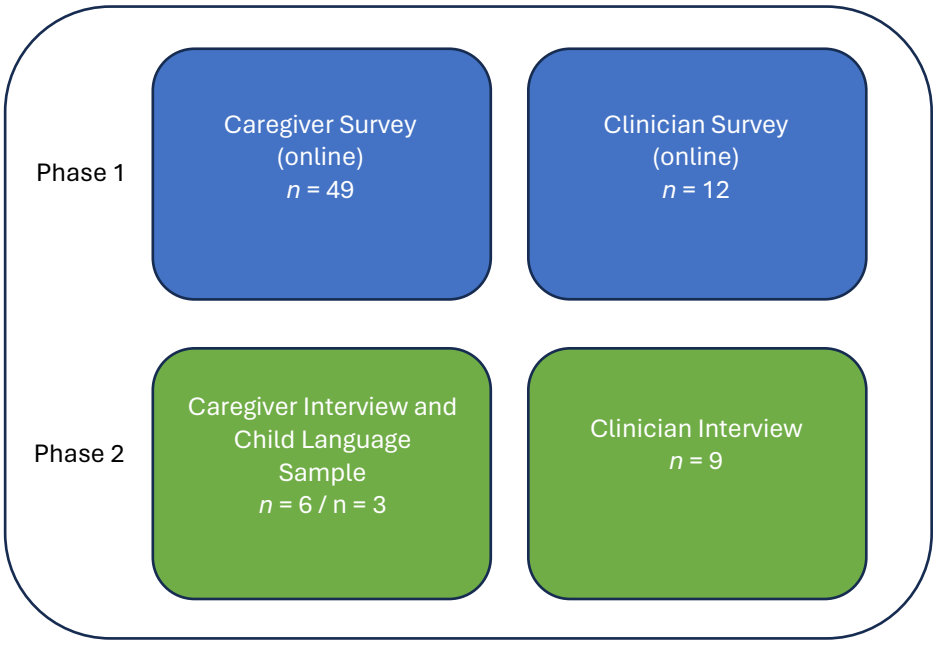


Figure 2

Average Ranking of Challenges as They Impact Treatment, Reported by Clinicians

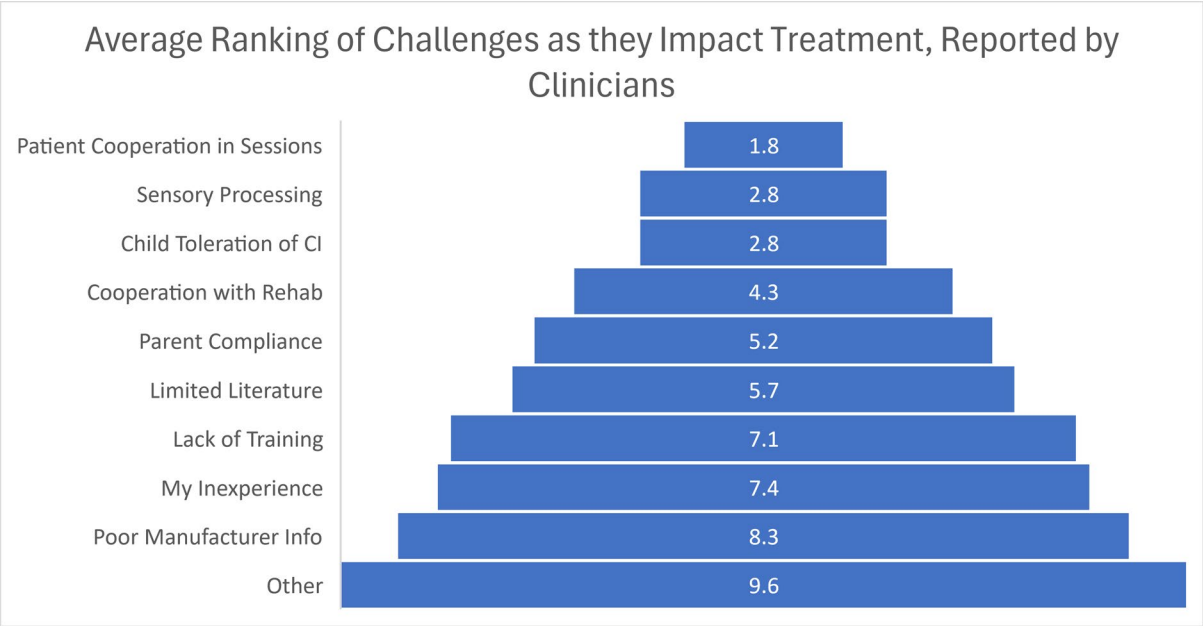


Figure 3

Average Ranking of Challenges as They Impact Spoken Language Development, Reported by Clinicians

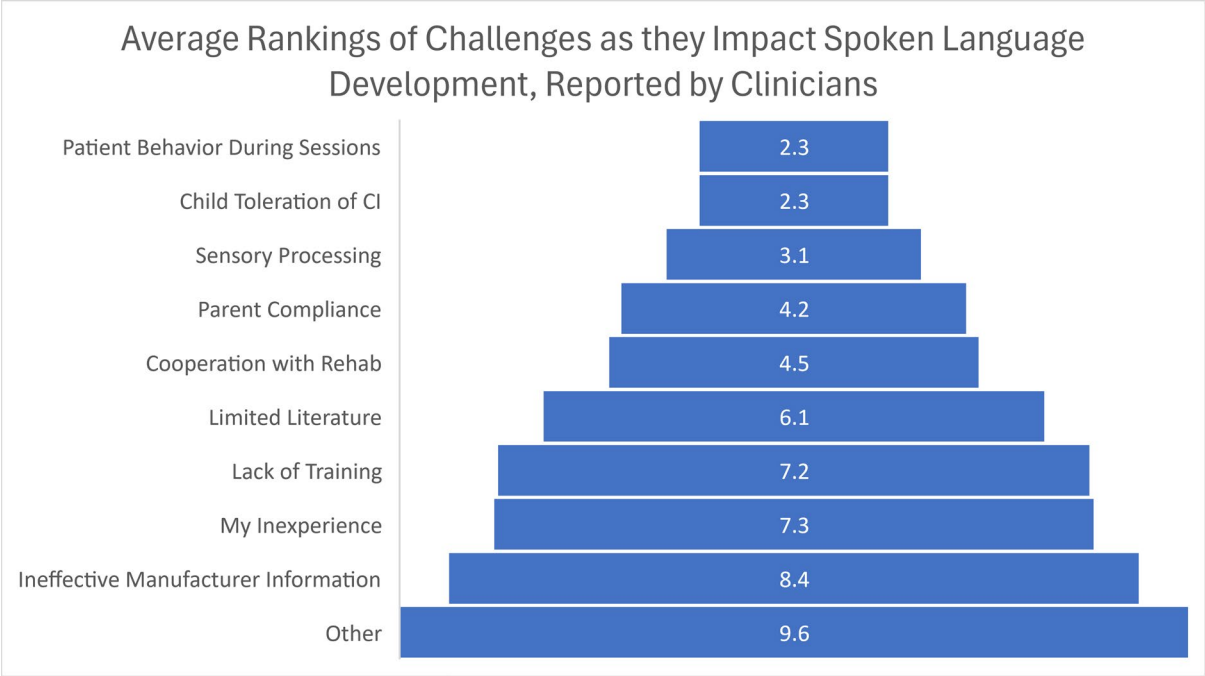


Figure 4

Correlation Between Sensory Profile & Language Measure

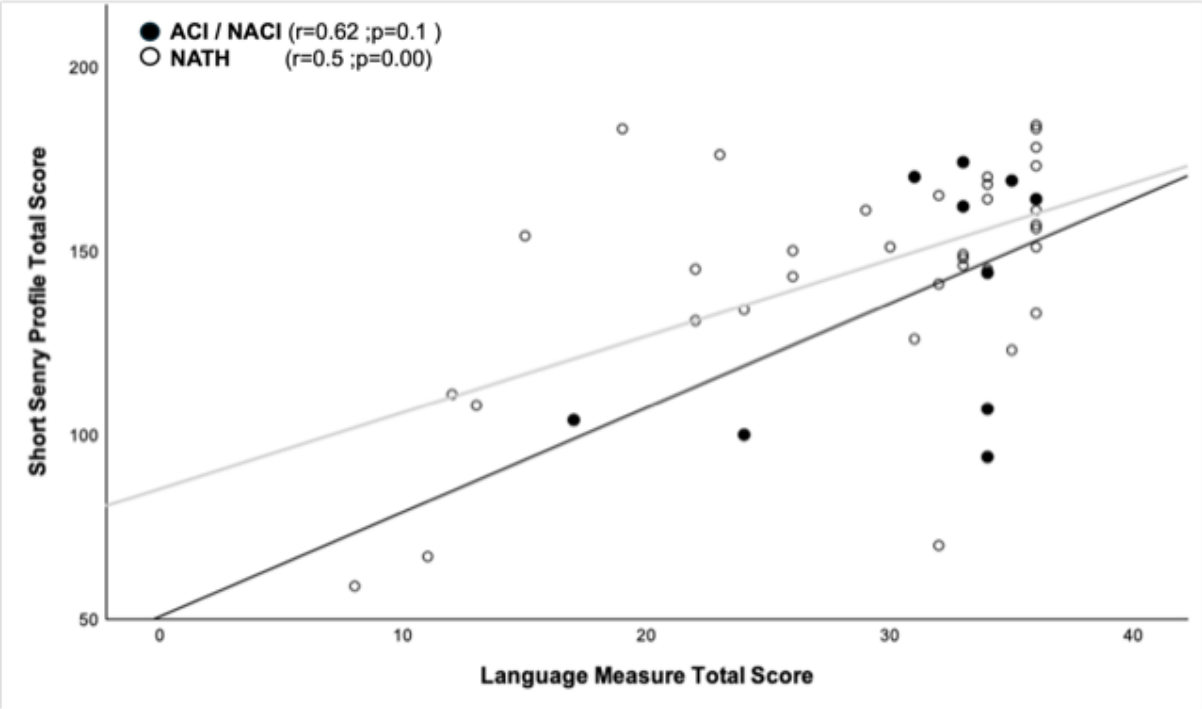
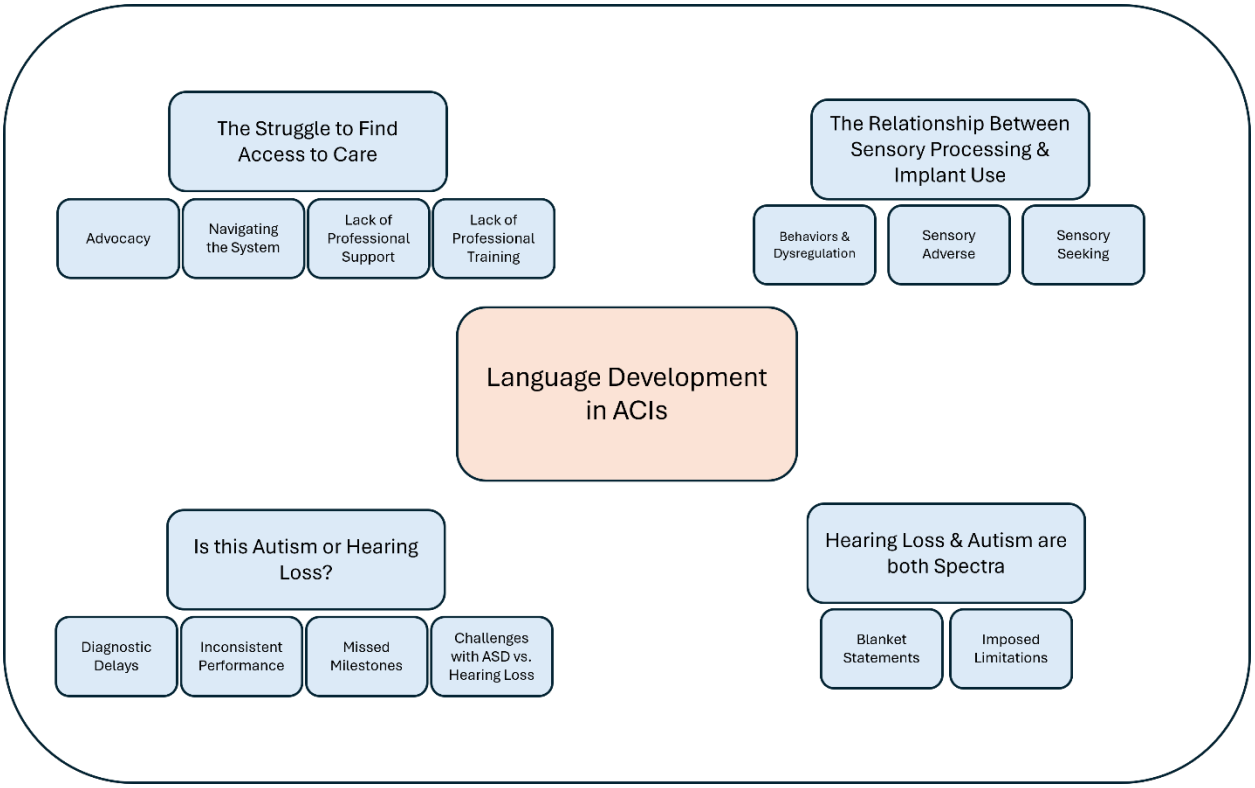


Figure 5

Themes & Subthemes



APPENDIX A

Institutional Review Board Approval Letter

To: Garrett Cardon
Department: BYU – EDUC – Communications Disorders
From: Sandee Aina, MPA, HRPP Associate Director
Wayne Larsen, Macc, IRB Administrator
Bob Ridge, Ph.D., IRB Chair
Date: November 27, 2023
IRB#: IRB2023-315
Title: Caregiver and Clinician Impressions on the Development of Spoken Language in Autistic Cochlear Implantees

Brigham Young University's IRB has approved the research study referenced in the subject heading as expedited level, categories 6 and 7. This study does not require an annual continuing review. Each year, near the anniversary of the approval date, you will receive an email reminding you of your obligations as a researcher. The email will also request the status of the study. You will receive this email each year until you close the study.

The IRB may re-evaluate its continuing review decision for this decision depending on the type of change(s) proposed in an amendment (e.g., protocol change that increases subject risk) or as an outcome of the IRB's review of adverse events or problems.

The study is approved as of 11/27/2023. Please reference your assigned IRB identification number in any correspondence with the IRB.

Continued approval is conditional upon your compliance with the following requirements:

1. A copy of the approved informed consent statement and associated recruiting documents (if applicable) can be accessed in iRIS. No other consent statement should be used. Each research subject must be provided with a copy or a way to access the consent statement.
2. Any modifications to the approved protocol must be submitted, reviewed, and approved by the IRB before modifications are incorporated into the study.
3. All recruiting tools must be submitted and approved by the IRB before use.
4. All data and the investigator's copies of the signed consent forms must be retained for at least three years following the termination of the study.
5. In addition, serious adverse events must be reported to the IRB immediately, with a written report by the PI within 24 hours of the PI's becoming aware of the event. Serious adverse events are (1) the death of a research participant or (2) serious injury to a research participant.
6. All other non-serious unanticipated problems should be reported to the IRB within two weeks of the PI's first awareness of the problem. Prompt reporting is important, as unanticipated problems often require some modification of study procedures, protocols, and/or informed consent processes. Such modifications require the review and approval of the IRB.

Consent Forms

Parental Subject Consent and Permission for a Minor
Title of the Research Study: Caregiver and Clinician Impressions on the Development of Spoken Language in Autistic Cochlear Implantees
Principal Investigator: Garrett Cardon

Introduction

This research study is being conducted by Professor Garrett Cardon and research staff at Brigham Young University to determine the factors associated with successful use of cochlear implants in autistic individuals. You and your child were invited to participate because your child has a confirmed diagnosis of autism and has used a cochlear implant for at least 6 months. They must have no history of epilepsy, head injury, neurological disorders, Fragile X Syndrome, or traumatic brain injuries.

Procedures

If you consent to participate in this study, you will be asked to do the following:

- Participate in an interview with one of our research team members—approximately 30-45 minutes.

In addition, your child may be asked to do one of the following based on age and your/their willingness:

- Your child will be told a story and asked to tell that story back to researchers. (Ages 3:6 – 12:8)
- Tell the researchers about a topic that is of interest to them (Children aged 12:8 and older)

It is anticipated your child will participate for a maximum of 15 minutes. If after 5 minutes your child does not wish to speak to researchers, this portion will be concluded.

All research activities will take place at the John Taylor Building on the BYU campus in the principal investigator's laboratory (in person) or in your home via Zoom (remote). We anticipate that your research appointment will last approximately 1 hour for the parent and child portions combined. If you come to our laboratory, you and your child will be given as much time as you need to familiarize yourselves with the building, room, and personnel involved in the study, ask questions, as well as breaks during the research activities.

Risks/Discomforts

There are no known significant risks involved in this research study, but there is always a possibility a small, unknown risk may exist to this or any test (i.e., discomfort related to questions or activities). However, we believe that we have taken reasonable precautions to ensure your safety. None of the questions we will ask or procedures related to the study are overtly distressing or meant to cause

discomfort or offense. If you have any questions about your safety in this experiment, please feel free to discuss them with us at any time. There is a risk that people outside of the research team will see your research information. We will do all that we can to protect your information.

Benefits

There will be no direct benefits to you. However, this study is designed for the researcher to learn more about the development of spoken language in autistic cochlear implant users. This study is not designed to treat any illness or to improve your health. We will not release any clinically un-interpretable results.

Confidentiality

Brigham Young University and the research team have rules to protect information about you. Federal and state laws including the Health Insurance Portability and Accountability Act (HIPAA) also protect your privacy. This part of the consent form tells you what information about you may be collected in this study and who might see or use it. We cannot do this study without your permission to see, use and give out your information. You do not have to give us this permission. If you do not, then you may not join this study.

We will see, use, and disclose your information only as described in this form. We will do everything we can to keep your records a secret. It cannot be guaranteed.

The use and disclosure of your information has no time limit. Data will always be stored on password protected computers, in filing cabinets in locked offices on the BYU campus, external hard drive, and/or with a secure cloud storage service (Box). Following data collection, research information will be stored in one or more of the above repositories indefinitely to ensure the potential of future analysis, if needed. You can cancel your permission to use and disclose your information at any time by writing to the study's Primary Investigator, at the name and address listed below. If you do cancel your permission to use and disclose your information, your part in this study will end and no further information about you will be collected. Your cancellation would not affect information already collected in this study.

Garret Cardon

Brigham Young University

Department of Communication Disorders

1190 N 900 E 130 TLRB

Provo, UT 84604

Both the research records that identify you and the consent form signed by you may be looked at by others who have a legal right to see that information. The participant's name will immediately be replaced with an identifying code in order to protect your confidentiality. Other identifying information will only be used to make calculations (such as chronological age) or contact you, if you provide permission (see below), but will never be used in any publication, presentation, or other form of communication with anyone other than you.

Federal offices such as the Food and Drug Administration (FDA) that protect research subjects like you. People at the Brigham Young University Institutional Review Board (BYUIRB), the study investigator and the rest of the study team.

Information about you that will be seen, collected, used, and disclosed in this study:

- Name and Demographic Information (age, sex, ethnicity, address, phone number, etc.)
- Research Visit and Research Test records
- Diagnoses that have been given to you or your close family members, such as anxiety, Autism Spectrum Disorder (ASD), or Attention Deficit Hyperactivity Disorder (ADHD)

What happens to Data that is collected in this study?

The scientists on the research team work to discover new information about autism. The data collected from you during this study is important to this study and to future research. If you join this study:

- Both the investigators and any sponsor of this research may study your data
- Any product or idea created by the researchers working on this study will not belong to you.
- There is no plan for you to receive any financial benefit from the creation, use or sale of such a product or idea.

Data Sharing

We will keep the information we collect about you during this research study for analysis and for potential use in future research projects. If the study data contain information that directly identifies subjects: Your name and other information that can directly identify you will be stored securely and separately from the rest of the research information we collect from you.

De-identified data from this study may be shared with the research community, with journals in which study results are published, and with databases and data repositories used for research. We will remove or code any personal information that could directly identify you before the study data are shared. Despite these measures, we cannot guarantee anonymity of your personal data.

Compensation

You will receive $20/hour (or any portion thereof) in the form of Visa gift cards for your participation in this study. There will be no monetary cost to you for participating in this study.

Participation

Participation in this research study is voluntary. You have the right to withdraw at any time or refuse to participate entirely without any risk to you whatsoever.

Questions about the Research

If you have questions, concerns, or complaints, you can contact the Principal Investigator,

Garrett Cardon, 303-241-6666, garrett.cardon@byu.edu or study coordinator, Courtney Marks, cblack98@byu.edu

Questions about Your Rights as Research Participants

If you have questions regarding your rights as a research participant contact Human Research Protections Program by phone at (801) 422-1461; or by email: BYU.HRPP@byu.edu.

Participation

Participation in this research study is voluntary. You are free to decline to have your child participate in this research study. You may withdraw you child's participation at any point without affecting his/her study benefits**.**

Please Indicate the Statement that Reflects Your Consent:

_____ I give consent to both my own and my child's participation in the research procedures mentioned above.

_____ I consent to my own participation but NOT to my child's.

Child's Name:_____

Parent Name:_____ Signature:_____ _____Date:

PERMISSION TO CONTACT FOR FUTURE RESEARCH STUDIES: Sometimes after a research project is finished, there are new questions that researchers need to ask and new research studies that need to be done. We would like your permission to contact you for participation in future studies that you may qualify for. We will not contact you unless you give us your permission.

_____ I agree to be contacted for future research studies that I/my children might be eligible for.

_____ I do not wish to be contacted in the future for any additional research studies.

If you agree to be contacted, please list an address, phone number, and email address where you can be reached:

Phone:_____

Email:_____

Clinician Participant Consent

Title of the Research Study: Caregiver and Clinician Impressions on the Development of Spoken Language in Autistic Cochlear Implantees
Principal Investigator: Garrett Cardon

Introduction
This research study is being conducted by Professor Garrett Cardon and research staff at Brigham Young University to determine the factors associated with successful use of cochlear implants in autistic individuals. You were invited to participate because you have experience working clinically with autistic cochlear implantees in one capacity, or another (e.g., audiologist, speech-language pathologist, occupational therapist, medical doctor, etc.)

Procedures
If you consent for them to participate in this study, you will be asked to do the following:

- Participate in an interview with one of our research team members—approximately 30-45 minutes.

All research activities will take place at the John Taylor Building on the BYU campus in the principal investigator's laboratory (in person) or in your home via Zoom (remote).

Risks/Discomforts
There are no known significant risks involved in this research study, but there is always a possibility a small, unknown risk may exist to this or any test (i.e., discomfort related to questions or activities). However, we believe that we have taken reasonable precautions to ensure your safety. None of the questions we will ask, or procedures related to the study are overtly distressing or meant to cause discomfort or offense. If you have any questions about your safety in this experiment, please feel free to discuss them with us at any time. There is a risk that people outside of the research team will see your research information. We will do all that we can to protect your information.

Benefits
There will be no direct benefits to you. However, this study is designed for the researcher to learn more about oral language development in autistic cochlear implant users. This study is not designed to treat any illness or to improve your health. We will not release any clinically un-interpretable results.

Confidentiality
Brigham Young University and the research team have rules to protect information about you. Federal and state laws including the Health Insurance Portability and Accountability Act (HIPAA) also protect your privacy. This part of the consent form tells you what information about you may be collected in this study and who might see or use it. We cannot do this study

without your permission to see, use and give out your information. You do not have to give us this permission. If you do not, then you may not join this study.
We will see, use, and disclose your information only as described in this form. We will do everything we can to keep your records a secret. It cannot be guaranteed.

The use and disclosure of your information has no time limit. Data will always be stored on password protected computers, in filing cabinets in locked offices on the BYU campus, and/or with a secure cloud storage service (Box). You can cancel your permission to use and disclose your information at any time by writing to the study's Primary Investigator, at the name and address listed below. If you do cancel your permission to use and disclose your information, your part in this study will end and no further information about you will be collected. Your cancellation would not affect information already collected in this study.

Garret Cardon
Brigham Young University
Department of Communication Disorders
1190 N 900 E 130 TLRB
Provo, UT 84604

Both the research records that identify you and the consent form signed by you may be looked at by others who have a legal right to see that information. The participant's name will immediately be replaced with an identifying code in order to protect your confidentiality. Other identifying information will only be used to make calculations (such as chronological age) or contact you, if you provide permission (see below), but will never be used in any publication, presentation, or other form of communication with anyone other than you.
Federal offices such as the Food and Drug Administration (FDA) that protect research subjects like you.
People at the Brigham Young University Institutional Review Board (BYUIRB), the study investigator and the rest of the study team.

Information about you that will be seen, collected, used, and disclosed in this study:

- Name and Demographic Information (age, sex, ethnicity, address, phone number, etc.)
- Research Visit and Research Test records

What happens to Data that is collected in this study?

The scientists on the research team work to discover new information about autism. The data collected from you during this study is important to this study and to future research. If you join this study:

- Both the investigators and any sponsor of this research may study your data
- Any product or idea created by the researchers working on this study will not belong to you.

- There is no plan for you to receive any financial benefit from the creation, use or sale of such a product or idea.

Data Sharing

We will keep the information we collect about you during this research study for analysis and for potential use in future research projects. If the study data contain information that directly identifies subjects: Your name and other information that can directly identify you will be stored securely and separately from the rest of the research information we collect from you.

De-identified data from this study may be shared with the research community, with journals in which study results are published, and with databases and data repositories used for research. We will remove or code any personal information that could directly identify you before the study data are shared. Despite these measures, we cannot guarantee anonymity of your personal data.

Compensation

You will receive $20/hour (or any portion thereof) in the form of Visa gift cards for your participation in this study. There will be no monetary cost to you for participating in this study.

Participation

Participation in this research study is voluntary. You have the right to withdraw at any time or refuse to participate entirely without any risk to you whatsoever.

Questions about the Research

If you have questions, concerns, or complaints, you can contact the Principal Investigator, Garrett Cardon, 303-241-6666, garrett.cardon@byu.edu or study coordinator, Courtney Marks, cblack98@byu.edu

Questions about Your Rights as Research Participants

If you have questions regarding your rights as a research participant contact Human Research Protections Program by phone at (801) 422-1461; or by email: BYU.HRPP@byu.edu.

Name: _____ Signature: _____

Date: _____

PERMISSION TO CONTACT FOR FUTURE RESEARCH STUDIES: Sometimes after a research project is finished, there are new questions that researchers need to ask and new research studies that need to be done. We would like your permission to contact you for participation in future studies that you may qualify for. We will not contact you unless you give us your permission.

_____ I agree to be contacted for future research studies that I/my children might be eligible for.

_____ I do not wish to be contacted in the future for any additional research studies.
If you agree to be contacted, please list an address, phone number, and email address where you can be reached:

Phone:_____

Email:_____

APPENDIX B

Instruments

Caregiver Survey

Demographics

1. What is your child's exact age in years and months?
2. What is your child's sex assigned at birth?
3. What is your child's race?
4. In which state do you currently reside?
5. What is the primary language spoken at home?
6. What is your child's ASD diagnosis status?
    a. Confirmed ASD (e.g. testing by a licensed professional has confirmed a diagnosis)
    b. Suspected ASD (e.g. you believe your child may be autistic but have not had confirmatory testing)
    c. My child is non-autistic/I do not suspect my child is autistic
7. What is your child's ADHD diagnosis status?
    a. Confirmed ADHD (e.g. testing by a licensed professional has confirmed a diagnosis)
    b. Suspected ADHD (e.g. you believe your child may be autistic but have not had confirmatory testing)
    c. My child does not have ADHD/I do not suspect my child of having ADHD.
8. What is your child's hearing loss diagnosis status?
    a. Confirmed hearing loss (e.g. testing by a licensed professional has confirmed a diagnosis)
    b. Suspected hearing loss (e.g. you believe your child may have hearing loss but have not had confirmatory testing)
    c. My child does not have hearing loss/I do not suspect my child has hearing loss.
9. Please describe your child's hearing loss (e.g. severity and treatment received).

10. At what age did your child receive their cochlear implant in months, if applicable?
11. At the time of this study, how many months of experience/use does your child have with their cochlear implant? (i.e. how long since your child received their implant?)
12. How many hours a day on average does your child use their cochlear implant?

Language Measure

1. What is your child's preferred/main method of communication?
    a. Oral (Spoken words or word approximations)
    b. Sign (American Sign Language)
    c. Total communication (Equal use of oral & sign)
    d. Vocalizations (sounds that are not words, e.g., grunting, vowel sounds, humming, yelling)
    e. Gestures or Body Movements (pointing, grabbing your hand to bring you to a desired object, throwing their body away from undesired objects)
    f. AAC Device or Speech Generating Device
    g. My child does not use any of the above (Please describe your child's communication)

Questions 2-6 Taken & Adapted from the BESA ITALK

2. How many spoken words does your child use from words learned at home (e.g., home words such as toys or words from daily routines, or school words such as science or math terms)
    a. A few words
    b. A limited range of words
    c. Some words
    d. Many words
    e. Extensive vocabulary
    f. Don't know
3. How often can you understand what your child says orally?

   a. Never

   b. Rarely

   c. Sometimes

   d. Very Often

   e. Always

   f. Don't know

4. How long are your child's spoken sentences?

   a. 0-1 words

   b. 1-2 words

   c. 2-3 words

   d. 3-4 words

   e. 4-5 words

   f. 5 or more words

   g. Don't know

5. How often does your child produce well-formed spoken sentences when conversing or telling stories?

   a. Never

   b. Rarely

   c. Sometimes

   d. Very Often

   e. Always

   f. Don't know

6. How often does your child understand what other people say to them orally? (For example, you may notice that your child often ask for repetitions or only follow parts of directions).

   a. Never

b. Rarely

c. Sometimes

d. Very Often

e. Always

f. Don't know

Questions 7-8 taken from Eshraghi et al., 2015

7. How would you rate your child's speech expression?
    a. No vocalization
    b. Some vocalization (consonants, vowels, nasal sounds)
    c. Words only
    d. Simple phrases and commands (where is X, let's go, etc.)
    e. Able to produce sentences.
8. How would you rate your child's speech perception? (Speech perception is your child's ability to recognize that speech is happening around them and has meaning).
    a. No awareness of environment
    b. Awareness, detection, or localization of sound (localization = they know which direction sounds come from)
    c. Identification/recognition of words
    d. Identification/recognition of simple phrases (2 words) and commands
    e. Understands conversations

Cognition Measure

1. What services if your child receiving, if any?
    a. Speech Language Pathologist services
    b. Audiological services
    c. Psychological services

d. Physical or Occupational Therapy

e. Counseling services

f. School services as determined by an IFSP, IEP, or 504

g. Other (Please Indicate)

h. None

2. Please indicate your child's current educational performance in the following areas:

    a. Reading

    b. Writing

    c. Language

    d. Math

    e. Overall, how is your child doing in school?

        i. Each area rated with the following:

            1. Far below age level

            2. Somewhat below age level

            3. Age level

            4. Somewhat above age level

            5. Far above age level

3. Please indicate any of the following diagnoses that your child has received.

    a. Anxiety disorders (such as separation anxiety, obsessive compulsive disorder, panic disorder, or selective mutism)

    b. Behavior disorders (such as conduct disorder or oppositional defiant disorder)

    c. Learning disorders (such as intellectual disability, dyslexia, or dyscalculia)

    d. Other: please indicate (i.e. concussion, seizure, diabetes, genetic disorders, asthma, digestive disorder, cancer, sensory processing disorder, etc.)

4. Please indicate your child's demonstration of the following cognitive skills compared to other kids your child's age.
    a. Attention (focuses on tasks for an age-appropriate length of time)
    b. Memory (retention and recall)
    c. Processing speed (responds to new information without unusual delay)
    d. Decision making (weighs choices and selects logically)

The Short Sensory Profile (SSP)

Screening for Child Anxiety Related Disorders (SCARED)

Caregiver Interview

The Auditory Skills Checklist (Meinzen-Derr et al., 2004)

Interview Questions

1. Please tell us a little about your child (likes/dislikes/personality).
2. Please tell us a little about your child's and family's journey with autism and hearing loss.
3. Can you describe what your biggest challenges related to hearing healthcare have been?
4. What did you expect the outcome of cochlear implantation would be before surgery?
5. What does a successful outcome of cochlear implantation look like to you?
6. Were you aware or suspicious that your child may have autism before CI implantation?
7. What factors do you believe contributed most to successful cochlear implantation?
8. What factors do you believe had an impact on your child's ability to acquire spoken language?
9. How much of a impact on spoken language development/CI success in ACIs do you believe sensory differences have?
10. What benefits does your child receive from their implant?

Clinician Survey

1. Please indicate your profession.
    a. Audiologist
    b. Speech-Language Pathologist
    c. Physician (ENT, CI Surgeon)
    d. Occupational Therapist
    e. Teacher
    f. Other
2. Please indicate your years of experience.
3. Where do you currently practice?
4. In what clinical settings do you currently practice?
    a. School
    b. Hospital
    c. Private Clinic
    d. Other (Describe)
5. What best describes the setting in which you work?
    a. Rural
    b. Suburban
    c. Urban
6. Please estimate the percentage of clients you treat with the following SES status (sum of the 3 must equal 100)
    a. Low
    b. Average
    c. High
7. How many ACIs have you treated throughout your career?

a. Clinicians were asked to indicate how many within:
    i. 0-3 Years
    ii. 4-7 Years
    iii. Within the last 10 years
    iv. More than 10 years ago
        1. None
        2. 1-5
        3. 5-10
        4. 10-15
        5. 15-20
        6. 20+

8. How many ACIs are you currently treating?
    a. None
    b. 1-2
    c. 3-4
    d. 5-10
    e. 11-15
    f. 16-20
    g. 20+

9. How confident are you in your ability to provide high quality care for ACIs and their families?
    a. Not confident at all
    b. Slightly confident
    c. Moderately confident
    d. Very confident
    e. Extremely confident

10. Please rank the following challenges as they impact your ability to treat ACIs (1 being the most challenging and 10 being the least challenging)
    a. Patient behavior/Cooperation during treatment
    b. Sensory Processing difficulties
    c. My relative inexperience in working with ACIs
    d. Lack of training or preparation from coursework or residency
    e. Limited resources/literature about ACIs
    f. Parent/family compliance
    g. Child toleration of CI on a daily basis
    h. Cooperation with CI rehab efforts
    i. Ineffective CI manufacturer resources for ACIs (e.g. software, mapping methods, etc.)
    j. Other (fill in the blank)
11. Please rank the following challenges as they impact the ability of the ACIs you've worked with to develop spoken language. (1 being the most challenging and 10 being the least challenging)
    a. Patient behavior/Cooperation during treatment
    b. Sensory Processing difficulties
    c. My relative inexperience in working with ACIs
    d. Lack of training or preparation from coursework or residency
    e. Limited resources/literature about ACIs
    f. Parent/family compliance
    g. Child toleration of CI on a daily basis
    h. Cooperation with CI rehab efforts
    i. Ineffective CI manufacturer resources for ACIs (e.g. software, mapping methods, etc.)
    j. Other (fill in the blank)
12. When working with ACIs, how often do you participate in interdisciplinary collaboration?

a. Never

b. Sometimes

c. About half the time

d. Most of the time

e. Always

13. What professionals do you collaborate with, if any, when working with ACIs?

a. None

b. Speech-Language Pathologist

c. Audiologist

d. Hearing Instrument Specialist

e. ENT

f. Pediatrician

g. Genetics Counselor

h. Social Worker

i. Deaf Educator

j. Occupational Therapist

k. Psychologist

l. Classroom Teacher

m. Neurologist

n. Psychiatrist

o. Other (with box)

14. Do you specifically counsel all parents to be aware of signs of autism soon after their child receives their cochlear implant?

a. Yes

b. No

c. NA (I am not an audiologist or CI surgeon)

Clinician Interview

1. Tell us about your experience working with ACIs.
2. Tell me more about your confidence as a clinician during treatment for ACIs.
3. Tell me more about some of your biggest challenges when working with this population, what does that look like during a session or appointment?
4. How often are you aware of an ASD diagnosis or notice indicators of ASD before implantation? What about after implantation?
5. What have you found to be most effective when working with/treating an ACI? Are there differences between working with ACIs and other CI recipients?
    a. Possible Probes: Specific protocols, treatment resources, evaluation methods
6. What do you define as a successful outcome for an ACI? For a NACI?
7. In your experience are ACI's likely to develop spoken language, why or why not?
8. How much of an impact on spoken language development/CI success in ACIs do sensory differences have? Explain.
    a. Possible Probes: Sensory differences like the ones seen commonly in autism – hypersensitivity, hyposensitivity, sensory seeking.

APPENDIX C

Annotated Bibliography

Baron, S., Blanchard, M., Parodi, M., Rouillon, I., & Loundon, N. (2019). Sequential bilateral cochlear implants in children and adolescents: Outcomes and prognostic factors. *European Annals of Otorhinolaryngology, Head & Neck Diseases, 136*(2), 69–73. https://doi.org/10.1016/j.anorl.2018.09.004

Introduction: Bilateral Cis have a variety of benefits. Some include understanding speech in noise, sound localization, and enhanced quality of life. Factors that will affect the outcome of a bilateral implantation include hearing loss etiology and duration, age at implantation for both implants, and the interval between the implantations.

Method: This study included 109 children who received a sequential cochlear implant, meaning they had one previously and received another one. They were evaluated both before their second implantation and at 3-, 12-, and 24-months post-op on their speech intelligibility and speech perception.

Results: Analysis of the data showed that sequential contralateral Cis provided significant benefits in terms of speech intelligibility and perception in silence as of three months after the second implantation. An additional CI could help accelerate language development in children who were not performing as well with one CI.

Relevance: The development of spoken language by both autistic and non-autistic cochlear implantees is paramount to this study. This article provides a potential solution for CI patients who are not successfully developing spoken language or who seek further benefit from their implant.

Black, J., Hickson, L., & Black, B. (2012). Defining and evaluating success in paediatric cochlear implantation – an exploratory study. *International Journal of Pediatric Otorhinolaryngology, 76,* 1317–1326. https://doi.org/10.1016/j.ijporl.2012.05.027

Introduction: Success in cochlear implantation is defined in this article as developing age-equivalent speech and language, as well as the family of the patient being happy with the services they received as well as with the speech outcome. Failure is when there is no observable benefit from the CI and the family is not happy with the process or management.

Method: This was done using a group of 25 profoundly deaf children with a unilateral CI. Each case was evaluated for things that may have a detrimental effect on the success of the implantation. Speech, language, vocabulary, and auditory skills were all assessed using standardized assessments.

Results: Some of the greatest prognostic factors for success includes family influence, compliance with habilitation, or the presence of auditory neuropathy or other syndromes.

Relevance: One of the aims of this thesis is to determine definitions of CI success for both caregivers and clinicians and this article begins to explore definitions of success for pediatric Cis.

Blanc, F., Blanchet, C., Sicard, M., Merklen, F., Venail, F., & Mondain, M. (2022). Audiological outcomes and associated factors after pediatric cochlear reimplantation. *Journal of Clinical Medicine, 11*(3148), 2–9. https://doi.org/10.3390/jcm11113148

Introduction: Cochlear Implants are typically considered reliable, but in 1.3–11.2% of recipients, reimplantation is required due to medical complications or device

malfunctions. Recently some have been reimplanted due to the availability of updated technology. This study aimed to identify the factors that influenced the recovery of speech perception following cochlear implantation in children.

Method: This was a retrospective study of 67 CI reimplantation outcomes over 30 years (1989–2019) The participants included 31 boys & 35 girls who underwent reimplantation within 6 years of being age 15. Success of reimplantation was assessed using criteria based on better/stable audiological outcomes, recovery from infection without recurrence, and the recovery of non-auditive symptoms.

Results: The factor with the biggest effect was participating in a speech rehabilitation program. Aural rehab was more indicatory of successful reimplantation than age/sex/etiology of deafness/indication for reimplantation. Reimplantation was associated with enhanced audiological performance. This suggests that Cis can be replaced without concern for speech or language complications, so long as appropriate aural rehab is done.

Relevance: This study focused on outcomes of pediatric Cis following reimplantation. This study may include children who have been reimplanted.

Bottema-Beutel, K., Kapp, S. K., Lester, J. N., Sasson, N. J., & Hand, B. N. (2021). Avoiding ableist language: Suggestions for autism researchers. *Autism in Adulthood*, *3*(1), 18–29. https://doi.org/10.1089/aut.2020.0014

Introduction: Ableism is the belief that disabled people are inferior to non-disabled people and is a discriminatory mindset. Autism is fundamental to how autistic people see and experience the world and for many of them, identity first language is preferred.

Results: We can avoid ableism in autism research by following a few principles. When screening has been done to rule out autism, that person should be called "non-autistic". Neurotypical should be reserved for extensive testing that has ruled out other types of neurodivergence such as ADHD, anxiety, depression, or other. The words "comparison group" or "non-autistic" group is preferred to "control group". Researchers of this study also recommend describing the characteristics of participants rather than labeling them as "high" or "low functioning".

Relevance: This study is focused on autistic people, and it will be important for me to know how to avoid ableism and ableist language in my reporting.

Brown, T. M., Baas, B. S., Stoeckel, R. E., Belf, L. A., & Poling, G. L. (2021). Assessment of children with hearing loss and co-occurring medical disorders: Challenging cases. *Perspectives of the ASHA Special Interest Groups*, *6*(2), 375–383. https://doi.org/10.1044/2021_PERSP-20-00080

Introduction: This study recognizes the importance of and difficulty of assessing individuals with hearing loss and other co-occurring pathologies, such as autism. They also emphasize the importance of interdisciplinary collaboration.

Method: This article goes through various cases to give an idea about how important interdisciplinary work is for assessing children with hearing loss who have an additional disability.

Results: Gallaudet estimates that as many as 1/59 children with hearing loss also have autism. There is still a consensus lacking in the literature, however some research suggests that up 3.5% of autistic individuals have hearing loss. Due to the similarity of symptoms experienced between individuals with hearing loss and autistic individuals,

diagnosis can be delayed or missed. The literature suggests that in autism, sensorineural hearing loss is the most common. The prevalence of autism in children with hearing loss is higher than in children with typical hearing, and children with profound hearing loss in both ears have a higher incidence of autism than any other category of hearing loss.

Relevance: This study will focus on clinician confidence and interdisciplinary collaboration. It is also relevant to this study to know that profound hearing loss is more closely linked with autism and that the prevalence of autism among the hearing loss population and vice versa is decently high.

Camarata, S. (2013). Pediatric hearing impairment, autism, and autism spectrum disorder: Implications for clinicians. *Perspectives on Hearing and Hearing Disorders in Childhood, 23*(1), 4–12. https://doi.org/10.1044/hhdc23.1.4

Introduction: Separating features of pediatric hearing loss and ASD are difficult because autism assessments are based largely on communication skills and social development: which are both influenced by hearing loss. The ADOS-2 (Lord et al., 2012) is not validated for children with hearing loss and should not be used as a standardized measure, although it can be used as a qualitative measure in conjunction with other tests.

Results: This study emphasizes that the best diagnosis of autism comes from clinical judgment based on case history and a child's development. During the ADOS clinicians must focus on assessing social interaction while ensuring that what they're observing is not attributable to a language disorder. It's also important to recognize that comorbidity of hearing loss and ASD can happen, but when the ADOS is used without considering the impact of hearing loss children can be misdiagnosed.

Relevance: For the ACIs participating in this study, it will be important to recognize that the ADOS has not been validated for them which further complicates intervention and diagnosis. This may also affect how many participants this study is able to recruit.

Campbell, J., Cardon, G., & Sharma, A. (2011). Clinical application of the P1 cortical auditory evoked potential biomarker in children with sensorineural hearing loss and auditory neuropathy spectrum disorder. *Seminars in Hearing, 32*(2), 147–155. https://doi.org10.1055/s-0031-1277236

Introduction: The P1 of the cortical auditory evoked potential (CAEP) shows age related decreases in latency and these latencies can help us visualize the development of the auditory pathway. For children with hearing loss, appropriate amplification at an early age can drive auditory development and auditory skill development so that they reach typical hearing levels.

Method: Researchers used synthesized speech /ba/ as stimulus and CAEPs were collected using EEG recording. Subjects were seated on a caregiver's lap or on their own. Participating subjects included one child with hearing aids, one child with a CI, and one child with auditory neuropathy, all were males. Their eyeblinks were tracked and those sweeps were rejected, the child with the CI had additional electrodes to minimize artifact from interference.

Results: The P1 CAEP is useful for assessing whether or not early treatment is effective and to examine clinical management of patients with pediatric hearing loss.

Relevance: The P1 waveform can help us see how the auditory pathway is benefited from amplification as well as can help us know if the amplification a child is

receiving is enough to drive typical auditory development. Other measures, such as behavioral and development testing, can be used as a compliment to audiological testing to help guide intervention.

Daneshi, A, & Hassanzadeh, S. (2007). Cochlear implantation in prelingually deaf persons with additional disability. *Journal of Laryngology and Otology*, *121*(7), 635–638. https://doi.org/10.1017/S0022215107005051

Introduction: The frequency of additional disabilities in addition to hearing loss is 3x as great in the Deaf/hard of hearing population than in the typical hearing population. The three most common disabilities reported in addition to hearing loss are learning disabilities, intellectual disabilities, and emotional/behavioral disabilities. Prelingual deaf patients with additional disabilities does not mean that Cis are contraindicated, but they may not be appropriate for all patients.

Method: This was a retrospective study on 398 prelingually deaf CI patients to identify frequency of additional disabilities. All patients participated in a screening and, if indicated, a comprehensive psychological assessment. Researchers tested auditory perception and compared their auditory perception results from pre-op to the time of the assessment.

Results: Researchers found that 60 of the 398 children with hearing loss had an additional disability (15%) Other authors have found as much as 30% - this study used younger children and that may have influenced their results. This study also didn't include children with 'severe mental retardation." Autism was the additional disability in four children (6.66%). Following implantation auditory perception improved in all groups but improved the least in congenitally deaf-blind and autism. D/Blind and patients with

ASD had limited development of auditory perception after CI: these patients require unique rehabilitation for the maximum outcome.

Relevance: This study is focused on children with Cis who have an additional disability, namely autism. It is important to understand prevalence of additional disabilities in the target population.

Demopoulos, C., & Lewine, J. D. (2016). Audiometric profiles in autism spectrum disorders: Does subclinical hearing loss impact communication? *Autism Research*, *9*(1), 107–120. https://doi.org/10.1002/aur.1495

Introduction: Rates of HL in people with ASD are higher than the reported general population, however, it should be noted that hearing loss does not cause ASD. Children with hearing loss and autistic children both show similar deficits such as in emotion recognition. There may be a sensory developmental component to skill acquisition in the social cognitive domain.

Method: Seventy-six children/adolescents were used in this study, 60 with autism and 16 without. All participants underwent comprehensive audiological screening. The ADI-R (Lord et al., 1994) is an extensive diagnostic interview that was used, as well as the ADOS, to determine autism diagnoses. Language was evaluated using the Clinical Evaluation of Language Fundamentals, fourth edition (CELF-4; Semel et al., 2003) and intelligence and vocal affect recognition were assessed using other measures. For tests of hearing, PTA and UCL were assessed along with tympanometry and ABR.

Results: Abnormal audiological findings were higher in the ASD group than in the control group (55 versus 6%). ASD had higher rates of peripheral audiological abnormality. Children with later onsets of hearing loss have better outcomes in emotion

recognition. HL can contribute to/exacerbate ASD symptomology. Frequent ear infections are also common in ASD, as well as abnormal tympanometry.

Relevance: Understanding the audiological profiles of autistic children as well as their strengths and weaknesses is paramount to understanding why spoken language development may be more difficult for them.

Donaldson, A. I., Heavner, K. S., & Zwolan, T. A. (2004). Measuring progress in children with autism spectrum disorder who have cochlear implants. *Proceedings from the Ninth Symposium on Cochlear Implants in Children, 130*(5), 666–671. https://doi.org/10.1001/archotol.130.5.666

Introduction: This study aimed to quantify successes, gains, and difficulties experienced by cochlear implantees with ASD. There has been an increase in the number of implantees with autism, possibly due to more cases of autism in the United States or lower ages of implantation.

Method: This study retrospectively looked at the speech and language scores of seven children who received a cochlear implant at the University of Michigan. Each of them had been diagnosed with autism or pervasive developmental disorder (five and two respectively) either before or after they received their implant. A survey was also given to parents of these children about their child's preferred mode of communication, behaviors, relationships with family members, and their views of success.

Results: While the ACIs did benefit from their cochlear implant, only one of them relies on spoken language as their primary form of communication. More common benefits included increased vocalization, eye contact, use of sign language, reactions to sound, and responses to requests. Most parents did indicate that they would recommend

cochlear implantation to other families with an ACI child. These researchers indicated that oral communication may not be a realistic goal for ACIs.

Relevance: This study supports one of our research questions about the development of oral communication in ACIs by providing evidence that they do not develop spoken language very commonly. Our question is why that happens and if we can find anything to support what may cause that to be the case.

Eshraghi, A. A., Nazarian, R., Telischi, F. F., Martinez, D., Hodges, A., Velandia, S., Cejas-Cruz, I., Balkany, T. J., Lo, K., & Lang, D. (2015). Cochlear implantation in children with autism spectrum disorder. *Otology & Neurotology, 36*(8), e121-NaN. https://doi.org/10.1016/j.ijporl.2021.110876

Introduction: This article also examined the outcomes of cochlear implantation in children with autism. Autistic children's difficulties with communication and sensory processing add a new layer of complexity to adapting to and functionally using a cochlear implant to learn oral language. This study assessed receptive and expressive language in ACIs.

Method: This was another retrospective study that compared preimplantation receptive and expressive language abilities to post implantation receptive and expressive language abilities. Participants included 15 children who were implanted at the University of Miami and were diagnosed with autism or pervasive developmental disorder not otherwise specified. Speech perception and expression was evaluated using the Early Speech Perception test, the Multisyllabic Lexical Neighborhood Test, or the Phonetically Balanced Kindergarten Test. A parental survey was also done with parents to evaluate how they felt the CI was benefiting their child and focused on communication

skills, behavior, and interactions with others. A control group of 15 CI patients with no other co-occurring disabilities was also used.

Results: The top three reported improvements after implantation included name recognition, improved response to verbal requests, and enhanced enjoyment of music. ACIs did not develop oral communication to the same extent as the NACI control group, but families are still happy with the outcome of implantation although clinicians should be sure to counsel them about having realistic expectations for outcomes after the implantation.

Relevance: Understanding typical outcomes for ACI patients is critical to this study, particularly how those outcomes relate to speech and language acquisition. This study also supports that ACIs do not develop oral communication to the same extent as their NACI peers and also emphasizes the importance of parental counseling by clinicians about expectations after implantation.

Gilley, P. M., Sharma, A., Dorman, M., Finley, C. C., Panch, A. S., & Martin, K. (2006). Minimization of cochlear implant stimulus artifact in cortical auditory evoked potentials. *Clinical Neurophysiology: Official Journal of the International Federation of Clinical Neurophysiology, 117*(8), 1772–1782. https://doi.org/10.1016/j.clinph.2006.04.018

Introduction: This study compared ways to diminish CI artifact during CAEP recordings. In infants the P1 latency can be 300–400ms but in adults can be as short as 50ms. CIs create electrical artifact on the scalp that can interfere with CAEP recordings, this artifact can be influenced by the type of CI, mode of stimulation, and surgical placement of remote return electrode. It can hide the biological response or be confused for the biological response - neither of which are good things!

Method: This study described two methods for removing artifact. The first was called Post-Processing Analysis for Removal. This method can help diminish artifact based on statistics, although it may not separate the biological artifact from the statistical response. The other method is called Independent Component Analysis (ICA). This is a generative model that maximizes information from statistics. It decorrelates data using PCA model and then does advanced statistics to remove artifact. This is a plausible technique for identifying and removing artifact from EEG before averaging. Something else that could be done which would reduce having to take out artifact post scan could be Optimized Differential Reference (ODR) or searching for a different electrode placement on the head that would have null artifact.

Results: ICA is a viable tool for minimizing artifact, but the difficulty is the correct identification of artifacts from the analysis. P1 latencies from ODR were similar when compared to the ICA analysis, but ODR could have more problems for some reasons. For example, if more electrodes were used it could be hard to find additional places on the head to place them while still minimizing artifact.

Relevance: In our EEG study it will be important to minimize artifact and to have it taken out of our scan results so that we can reliably see the maturation of the auditory cortex.

Jenks, C., Hoff, S., Haney, J., Tournis, E., Thomas, D., & Young, N. (2022). Cochlear implantation can improve auditory skills, language, and social engagement of children with autism spectrum disorder. *Otology & Neurotology*, *43*(3), 313–319. https://doi.org/10.1097/MAO.0000000000003463

Introduction: The goal of this study was to understand the range of outcomes that occur after cochlea implantation in autistic children. They hoped to be able to make the case for more support for these children and emphasize the benefits that come from implantation in these dually diagnosed children. This study was the largest series with the longest follow up completed to date.

Method: This was a retrospective clinical review and parental survey. There were 30 ACI participants and the mean length of follow up was 10:05 years. Average age of implantation was 03:06 and average age of ASD diagnosis was 05:01, of which 73% were diagnosed after receiving their cochlear implant. This study did not have cognitive measures or ASD severity measures.

Results: Thirty-three percent of all patients developed measurable open set speech perception and of the consistent device users, 45%. Children who developed spoken language included 31% who used spoken language exclusively, 14% who used a mix of sign and speech, sign alone was used for another 14%. Some additionally used AAC and 28% had no mode of communication. Parent report showed that social engagement improved after receiving their cochlear implant. Communication and attention were the areas most impacted by the CI as indicated by parents. 73% percent of the children were consistent users, 17% inconsistent, and 10% non-users. Reaffirm that 25-30% of autistic children do not develop spoken language. It should be noted that they found that of their children who used spoken language or signed language that with the exception of four children, none of the rest had age-appropriate communication and were limited.

Relevance: This study is essential to my research as it focuses on spoken language development in ACIs – something that is not widely studied. This study also gave

interesting insight into consistency of CI usage, oral language versus signed language versus total communication, and explore a range of outcomes following CI implantation in children with ASD.

Lachowska, M., Pastuszka, A., Łukaszewicz-Moszyńska, Z., Mikołajewska, L., & Niemczyk, K. (2018). Cochlear implantation in autistic children with profound sensorineural hearing loss. *Brazilian Journal of Otorhinolaryngology*, *84*(1), 15–19. https://doi.org/10.1016/j.bjorl.2016.10.012

Introduction: The goal of this study was to examine the benefits of cochlear implantation in Deaf autistic children with ASD as their only additional disability. Limited research exists that is specific to implanted autistic children. Data from evoked potentials suggests that there is abnormal cognitive processing of auditory information despite there being typical processing of sensory perception.

Method: Researchers analyzed six prelingually deaf children with ASD (all males). Children were all extensively evaluated using a careful multidisciplinary approach to determine candidacy for the study. None had ASD diagnosis at time of implantation, although for some it was suspected but not confirmed. One specific child was highly suspected of having an additional ASD diagnosis. Each child had no post-surgery complications, regular follow up, and multidisciplinary rehabilitation.

Results: All children had delayed receptive and expressive language and none of the children were using gestures to communicate. Three would nod or shake head for yes/no. The oldest child would use a few short-spoken words, but none used verbalizations to truly communicate. Two responded to their name and spoken requests as well as the Ling 6 sound test. Most children had reduced anxiety while wearing their

processor and all families reported positive changes in their child's ability to interact with family members. The researchers concluded that children with ASD do not perform as well after implantation as implanted children without other disabilities and said, "The usual goal of spoken language after cochlear implantation may be unrealistic." They also emphasized the importance of caution during mapping due to autistic children's sensitivity to sound. A jarring quote from the study was: "Based on our experience and those from other studies, receiving an implant by autistic children in most cases does not lead to the development of speech and language even after many years." Other more common benefits from CI implantation in this population are typically: response to name, response to environmental sounds, reduced anxiety and better personal interaction with family.

Relevance: This study is vital– I want to know why spoken language is so difficult for ACIs to develop, even after successful implantation and appropriate aural rehabilitation. Why is it that even after many years, these children do not seem to develop spoken language at a rate that resembles NACIs, or even the rate of language acquisition in typical hearing autistic children?

McRackan, T. R., Hand, B. N., Chidarala, S., & Dubno, J. R. (2022). Understanding patient expectations before implantation using the cochlear implant quality of life-expectations instrument. *JAMA Otolaryngology-Head & Neck Surgery, 148*(9), 870–878. https://doi.org/ 10.1001/jamaoto.2022.2292

Introduction: Realistic patient/family expectations are the most important non audiological factor to successful implantation. Prior qualitative interviews with adult CI implantees suggested that most patients didn't feel like they had a clear understanding of

what post implantation would be like or what a realistic outcome would be following their surgery.

Method: Converted the Cochlear Implant Quality of Life (CIQOL) profile into statements that reflected a future outcome. There were 21 participants recruited from patients undergoing CI candidacy evaluations at the researcher's CI center and experienced psychometricians evaluated the results and their psychometric properties.

Results: Two participants recommended clarifying the instructions, but no other changes were suggested by participants, so the results supported the use of CIQOL-Expectations instrument for research and clinical use

Relevance: I will be doing qualitative research with caregivers of cochlear implantees, and part of this study is about learning definitions of success and what expectations were for the CI and implantation process.

Meinzen-Derr, J., Wiley, S., Bishop, S., Manning-Courtney, P., Choo, D. I., & Murray, D. (2014). Autism spectrum disorders in 24 children who are deaf or hard of hearing. *International Journal of Pediatric Otorhinolaryngology, 78*(1), 112–118. https://doi.org/10.1016/j.ijporl.2013.10.065

Introduction: Autism diagnoses are challenging at young ages because instead of looking for noticeable features, typically you must look for absent skills such as eye contact. It's much easier for parents to notice problems with hearing or sight due to missing developmental milestones. The literature on the dual diagnosis of ASD and hearing loss is severely lacking, although existing data supports profound HL in children with ASD as the most common degree of loss.

Method: Participants included 24 ACIs. Each child was diagnosed using the ADOS and researchers collected information about communication modality, language, and cognitive abilities. As this was a descriptive study, descriptive statistics were used that included means and ranges as well as frequencies with percentages. Sum tests and Chi-Square tests were also used.

Results: Of their 24 subjects, 14 had a CI, with 3 no longer using it, and ages ranged from 14-91 months at age of implantation. Two of these children used oral communication primarily and ASD was diagnosed in 22 children before the implant. For the children who used oral communication, they also supplemented with ASL and other visual strategies. It's difficult to know if speech and language difficulty in the other children was due to the autism, the hearing loss, or a combination of the two.

Relevance: It was interesting to read a study where some ACIs did develop oral communication, this shows that it is possible! Their study raises more questions about why some develop language but still not at the same rate as NACIs or even other autistic children.

Mikic, B., Jotic, A., Miric, D., Nikolic, M., Jankovic, N., & Arsovic, N. (2016). Receptive speech in early implanted children later diagnosed with autism. *European Annals of Otorhinolaryngology*, *Head and Neck Diseases*, *133*(Supplement 1), S36–S39. https://doi.org/10.1016/j.anorl.2016.01.012

Introduction: The aim of this study was to assess the development of auditory perception and speech intelligibility in implanted children who were diagnosed with ASD compared to typical developing children. They reported that 1/68 children in the US in 2014 have ASD. Current methods of ASD screening may have difficulties establishing a diagnosis

where other developmental delays co-occur, such as hearing loss – particularly because the ADOS is not validated for D/HH children. CI implantation outcomes in deaf children later diagnosed with autism are unclear and difficult to predict The aim of the study was to follow the development (auditory perception and speech production) of implanted children with ASD and compare it to implanted neurotypical children. It should be noted that this study is about receptive language, not expressive.

Method: Fourteen children with CIs from Serbia implanted between 12–18 months with congenital bilateral profound sensorineural type HL and limited acquired language. Four were later diagnosed with ASD (three male and one female) and 10 typically developing. All underwent intensive speech & hearing therapy. Their auditory comprehension using CAP (Categories of Auditory Perception) & SIR (speech intelligibility rating). Testing completed starting at age 2 up until age 6 annually. Children with ASD developed slower, by age 6 depending on the child, they were able to identify environmental sounds or discriminate speech sounds. Speech intelligibility was rated at the highest on the SIR a two with little to no progress up to age 6 despite intense speech-language therapy (average score for neurotypical children age 6 is 4.6)

Results: Children with ASD, auditory processing developed more slowly but it was very child dependent. At best rated SIR as category two, with very little or no progress up to age 6 despite intensive therapy, there was a significant statistical difference between both groups on both CAP & SIR scores. This study also emphasized that hearing loss can further delay diagnosis of ASD and that early diagnosis of ASD and HL are vital to providing appropriate services to these children. After implantation autistic children showed improvements in behavior, vocalization, eye contact, reaction to

sound, although they had limited auditory perceptual development and speech intelligibility compared to other implanted children with other diagnoses. Auditory performance developed at varying degrees, and none developed intelligible expressive speech. Expectations for implanted children should be strongly modified and the main goal should be to set a foundation for communication and eventually support sound and speech development. When combined with hearing loss, children with ASD showed less progress after implantation than other children who had hearing loss and a disability other than ASD. They also said that parents who are undergoing implantation for their child should be counseled about a possible future ASD diagnosis.

Relevance: This is another study that did not find CIs to be helpful for autistic children in developing spoken language and found other benefits to be more common. This study also mentions the importance of parent counseling and part of my study will include a qualitative caregiver interview about their experience throughout the implantation process and about caring for their autistic child with hearing loss.

Muncy, M. P., Yoho, S. E., & McClain, M. B. (2019). Confidence of school-based speech-language pathologists and school psychologists in assessing students with hearing loss and other co-occurring disabilities. *Language, Speech & Hearing Services in Schools, 50*(2)*,* 224–236. https://doi.org/10.1044/2018_LSHSS-18-0091

Introduction: This article assessed the confidence of SLPs and SPs (school psychologist) in working with children with HL and other co-occurring disabilities. This is important because approximately 38.9% of children with HL have other disabilities or disorders. The first question they were asked was about confidence in assessing children with multiple disabilities, including HL, the second was about their experiences and attitudes

about interdisciplinary collaboration, and the third was asking what additional training would help them.

Method: Fifty-nine questions on the survey, completed through REDCap. 204 surveys were analyzed. Quantitative analysis used to evaluate all questions except one of them. They used 2-way analysis of variance (ANOVA). There was one open ended question: thematic analysis used for analysis.

Results: Researchers determined that SLPs & SPs have not received sufficient training or experience with children with HL + something else. This can affect the quality of service these children receive. Low confidence across several skill set types that are needed for proper assessment and treatment of children with HL + something else. Barriers prevent collaboration with professionals with knowledge about HL – specifically the participating clinicians felt that they need more access to AuDs.

Relevance: This study gave great insight into the confidence and experience of school based SLPs as they work with dually diagnosed populations, in this case hearing loss with another comorbid diagnosis. This study indicates a lack of training and experience providing services to children with hearing loss in addition to another disability.

Samson, F., Mottron, L., Jemel, B., Belin, P., & Ciocca, V. (2006). Can spectro-temporal complexity explain the autistic pattern of performance on auditory tasks? *Journal of Autism & Developmental Disorders, 36*(1), 65–76. https://doi.org/ 10.1007/s10803-005-0043-4

Introduction: This paper aimed to see if the complexity hypothesis of Bertone et al (2005) can be applied to auditory stimuli. Auditory complexity has two levels, spectral

and temporal. Spectral means that it contains energy at one frequency (pure tone) versus energy at several frequencies (harmonic series), and temporally complex means that it has amplitude changes between onset and offset or have sequences of sounds delimited by an onset and offset. Vocal sounds are especially complex because they include harmonics, formants, rapid sequences of speech sounds.

Method: Simplest behavioral assessment of cortical auditory processing is represented by categorization, labeling, and discrimination tasks. MEG (Better spatial) EEG and ERP were all used to look at the N1 primary auditory sensory response in adults. MMN/MMF (mismatch negativity and mismatch magnetic field) can be done using auditory and speech stimuli and don't require active participation. This can be done on young children and autistic people who don't have expressive language.

Results: Researchers found decreased perfusion at rest in associative auditory cortex, superior temporal gyrus, multimodal superior temporal gyrus in autistic children, although they recognize that it's hard to make a true comparison there because language abilities of controls were likely higher than the autistic group. Ceponiene et al., (2003) found intact MMN for language delayed autistic children pure tone stimuli. The problem with brain imaging studies on autistic people with language processing difficulties is that it's hard to know what the root cause of the difficulty is: the autism, less experience, or just impaired language development overall.

Relevance: This paper looks at another reason why language processing may be difficult for autistic individual specifically. Auditory processing is already difficult and then if you add a cochlear implant into the mix, it becomes even harder.

Sharma, A., Campbell, J., & Cardon, G. (2015). Developmental and cross-modal plasticity in deafness: Evidence from the P1 and N1 event related potentials in cochlear implanted children. *International Journal of Psychophysiology, 95*(2), 135–144. https://doi.org/10.1016/j.ijpsycho.2014.04.007

Introduction: CAEPs are biomarkers that help us to evaluate the maturation of the auditory cortex using EEG. Auditory cortex maturation is vital to effective speech and oral language acquisition. In typically developing humans, the P1 happens 300ms after the stimulation and then at age 2 it goes down rapidly to 100ms. From there a gradual decrease to 50–70ms in adults takes place. Children with CIs who were implanted early had more successful CI outcomes as a result of better auditory cortex maturation.

Results: CAEPs with P1 and N1 support the idea of sensitive periods for auditory cortex maturation. Studies on both humans and animals who received CIs also support this sensitive period. These may be caused by enhanced synaptogenesis that turns into synaptic refinement after age 3.5–4. After the sensitive periods end, cross modal reorganization can happen where other senses take over auditory cortex space because it's not being used, and this can cause CI surgeries to be unsuccessful.

Relevance: Auditory cortex maturation is vital for speech and oral communication. Part of this study includes looking at the maturation of the auditory cortex in autistic brains and seeing if the cause of impaired language acquisition for ACIs is due to a problem with auditory cortex maturation. Perhaps the CI is not working in autistic brains to help the auditory cortex mature the way that it does in non-autistic brains.

Sharma, A., Dorman, M., & Spahr., A. (2002). Rapid development of cortical auditory evoked potentials after early cochlear implantation. *Neuro Report*, *13*(10), 1365–1368. https://doi.org/ 10.1097/00001756-200207190-00030

Introduction: This study aimed to test the hypothesis that early implantation in a highly plastic system helps the effects of acoustic deprivation to be overcome very quickly.

Method: Consent was obtained from parents and then researchers used CAEPs to measure the auditory cortex maturation of cochlear implantees using EEG.

Results: In early implanted congenitally deaf children, the CAEP changes dramatically in the first 6–8 months after they receive their implant. Many children reach age appropriate CAEP levels within 8 months and it was interesting to see that the rate of development was faster for these implanted children than in typical hearing controls.

Relevance: Non-autistic children who receive a cochlear implant catch up with their typical hearing peers, as far as auditory cortex maturation, after 8 months of using the implant in many cases. This indicates that the cochlear implant is doing its job and that continued hearing loss or the auditory cortex not having developed enough are not contributing factors to spoken language delays.

Szymanski, C. A., Brice, P. J., Lam, K. H., & Hotto, S. A. (2012). Deaf children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *42*(10), 2027–2037. https://doi.org/10.1007/s10803-012-1452-9

Introduction: There are higher rates of autism among those with hearing loss than among typical hearing individuals and the prevalence of profound hearing loss in autism is ten times higher than in the public. Autism is often overlooked in children with hearing loss and intervention research is only just now beginning to emerge for children who are both

Deaf and have ASD. Unfortunately, educational services and resources are severely lacking for Deaf autistic children and even cochlear implants were discouraged for Deaf children with multiple disabilities until 1992.

Method: This study used the Gallaudet 2009–2010 Annual Survey which surveyed 37,828 Deaf and Hard of Hearing children.

Results: Of those surveyed, 12, 595 (39.5%) reported having an additional disability and 611 (1.9%) reported having a diagnosis of both hearing loss and autism. Overall, the prevalence of autism in Deaf/HH children in the school was reported as 1/53. Interestingly, for the Deaf-ASD children there were higher occurrences of pregnancy related deafness which suggests that neurological risk factors may better explain the co-occurrence rather than sensory impairment from autism. Diagnostic overshadowing can happen in this population where hearing loss shields autism due to their similar characteristics.

Relevance: Understanding more about the Deaf/Autistic population is vital to this study as it focuses on ACIs. One of our ideas for why spoken language acquisition is more difficult in this population is because of sensory processing difficulty, so it's interesting to see that the cause of deafness in many of these autistic children is pregnancy related.

Tavares, F. D. S., Azevedo, Y. J., Fernandes, L. D. M. M., Takeuti, A., Pereira, L. V., Ledesma, A. L. L., & Bahmad, F. (2021). Cochlear implant in patients with autistic spectrum disorder - A systematic review. *Brazilian Journal of Otorhinolaryngology*, *87*(5), 601–619. https://doi.org/10.1016/j.bjorl.2020.11.020

Introduction: This article sought to find evidence in the literature that CIs are beneficial for autistic people with hearing loss and was a great resource for finding additional CI/autism related articles.

Method: This study was a systematic review of literature. Studies that met the eligibility criteria were included in a qualitative synthesis.

Results: Researchers found 284 articles in eight databases and 100 in the gray literature. They read 12 full articles and chose seven to be included in the qualitative analysis. Combined there were 66 ACIs included in the study. Not all individuals in their studies developed communication. More common benefits included interacting with family, better eye contact, and the ability to identify sounds. The ACIs did not have as significant gains as the NACI participants throughout the studies. They also found that diagnosing either autism or hearing loss often causes a delay in diagnosing the other.

Relevance: This was a great resource for additional CI/autism articles. They also found that ACIs did not develop oral communication as well as NACIs and talked about diagnostic overshadowing being a problem for this population.

Wiley, S., Gustafson, S., & Rozniak, J. (2014). Needs of parents of children who are deaf/hard of hearing with autism spectrum disorder. *The Journal of Deaf Studies and Deaf Education, 19*(1), 40–49. https://doi.org/10.1093/deafed/ent044

Introduction: The goal of this study was to identify the needs of children who are D/HH with ASD from the family's perspective. These families found a lack of professionals who understand ASD and Deafness and have turned to the internet and technology to find their own answers and connect with other families. The prevalence of ASD among children with who are D/HH has been estimated to be between 1–4% but that is probably

not accurate because of small samples and old data. The ADOS not validated on children who are D/HH and existing evidence for intervention is based on case reports of single children.

Method: Four parents of three children participated in a group discussion about diagnostic process, impact of dual diagnosis on communication, and helpful resources. This study used a focus group methodology with structured, open-ended questions. Participants were identified through a pediatric urban tertiary care center. Each participant had to have received a D/HH and ASD diagnosis via interdisciplinary assessment. The focus group questions were generated based on a background literature review and a trained facilitator from a state educational agency focused on autism and other low incidence disabilities was used. Responses were audiotaped and transcribed.

Results: Lack of parental involvement in evaluation process and all families noted that children were asked to do things they knew they couldn't do. Non-Effective reinforcements were provided for the children without parental input and evaluation including an interpreter, but the child had no concept of how to use an interpreter. Many felt that the evaluator was insufficiently trained and inexperienced in assessment for the dual diagnosis of D/HH of ASD. One child was diagnosed by a grad student with supervision through a mirror, but the parent felt that their child was a guinea pig. Some parents felt anger about the diagnostic process. Other things they reported were that schools were the primary resource for information and support, the Internet was a consistent resource, they wanted to prioritize functional and social skills over academic skills, and that social skills groups were helpful, but access was limited.

*Relevance***:** This study used both a survey and interview method which my study will also be using. It also made think about a trained facilitator and wonder if it will be important for me to specifically train or if that won't be as important because all my interviews will be individual.

Zaidman-Zait, A., & Curle, D. (2018). Complexity: An interpretative phenomenological analysis of the experiences of mothers of deaf children with cochlear implants and autism. *Journal of Health Psychology, 23*(9), 1173–1184. https://doi.org/10.1177/1359105316646171

Introduction: Understanding parent experience is vital because a lot of aural rehabilitation after implant relies on parents' involvement. This study also recognizes the stress that having a child diagnosed with hearing loss or autism can incur and the dual stress that having a child diagnosed with both would experience.

Method: Participants were recruited through online websites and groups as well as through professional organizations. Nine ACI mothers participated, all of whom had a male ACI child. Demographically, 2/9 used "oral communication" 1/9 used "total communication" 2/9 used signs, gestures, and vocalizations, and 4/9 used early communication behaviors. Telephone interviews were conducted with each of the mothers, and they were asked questions about their child's CI use, parenting their child, social support, family relationships, domestic workload and more. Results were analyzed using the IPA 5 Step Process.

Results: Three themes were recognized: complexity, family and personal sacrifices, and parent-professional partnerships. Learning about ASD and HL together was difficult, there were limited resources. The mothers also expressed difficulty with communicating with their child and many observed their children closely to try and make

sense of their behaviors. This was made further complex due to the mothers having to juggle multiple roles as well as make decisions. There are also negative impacts on family that requires sacrifices such as moving to a more urban area where their child would have access to better care or professionals. They also feel that their social relationships have suffered. Some mothers, however, also felt that their child's disabilities had strengthened their family bonds. Mothers also expressed frustration with professional relationships.

Relevance: Part of this study is aimed at understanding parent perspectives on their child's language development for ACIs. This is the only other study I've seen that specifically looks at parents and how they handle or have reacted to having a child that is dually diagnosed and how it has impacted their life. While this study doesn't aim to look at reasons why spoken language may be difficult to develop or evaluate in this population, it does serve as a great foundation for questions that we could ask in an interview and help us understand more of their perspective.

Zheng Y. N., Waite, M., Ekberg, K., & Hickson, L. (2022) Clinicians' and managers' views and experiences of audiology and speech-language pathology service provision for culturally and linguistically diverse families of young children with hearing loss. *Journal of Speech, Language & Hearing Research, 65*(7), 2691–2708. https://doi.org/10.1044/2022_JSLHR-21-00378

Introduction: This study focused on learning from the experiences of clinicians and managers who work with culturally and linguistically diverse (CALD) individuals whose children have co-occurring hearing loss.

Method: Qualitative descriptive study involving 27 semi structured interviews with AuDs, SLPs, and managers working with CALD families of young children with hearing loss. Interview schedule was developed and then questionnaires used to gather demographic data, then they conducted one-to-one interviews with clinicians and managers over the space of 1 year. Interviews were all conducted in person and ranged from 30–90 minutes. Audio recordings were transcribed verbatim and analyzed using reflexive thematic analysis informed by Braun & Clarke (2006).

Results: The clinicians and managers experienced culture differences with some of the families on their caseloads. Some of their challenges included stigmas about hearing loss, gender roles, and resistance to hearing intervention. Some caregivers had a hard time using the strategies they were taught in their home language.

Relevance: This study used a similar design to what we're considering with our clinician interviews, and it is interesting to read about what has been asked of clinicians in the past when working with the Deaf population. We aren't focusing on CALD populations, but some people consider being autistic to be part of their culture.

APPENDIX D

Supplementary Data

Supplementary Data Table 1

Average Ranking of Challenges as they Impact Treatment, Reported by Clinicians

Challenge	Avg. Rank	SD
Patient Cooperation in Sessions	1.8	1.03
Sensory Processing	2.8	1.14
Child Toleration of CI	2.8	1.93
Cooperation with Rehab	4.3	1.83
Parent Compliance	5.2	2.04
Limited Literature	5.7	1.42
Lack of Training	7.1	2.02
My Inexperience	7.4	1.35
Poor Manufacturer Information	8.3	1.06
Other	9.6	1.26

Supplementary Data Table 2

Average Ranking of Challenges as they Impact Spoken Language Development, Reported by Clinicians

Challenge	Avg. Rank	SD
Patient Cooperation in Sessions	2.3	1.42
Child Toleration of CI	2.3	1.49
Sensory Processing	3.1	1.10
Parent Compliance	4.2	2.10
Cooperation with Rehab	4.5	2.17
Limited Literature	6.1	1.37
Lack of Training	7.2	1.40
My Inexperience	7.3	1.83
Poor Manufacturer Information	8.4	0.97
Other	9.6	1.26

APPENDIX E

Quotes

1. “We’ve found the caregivers and experts to [provide services to our child] but it has been extremely challenging.”
2. “Parents know their child more than any other person and I think that parent’s voices get lost within the system.”
3. “I don’t want to start crying, but it was difficult.”
4. “I essentially convinced [the clinic] to [pay for further education] and I was like, you know, some of these concepts might be helpful for other kids in the program too.”
5. “I’m a mom that knows how to navigate [the system] and knows the language to put up the fight to get the right people at the table?”
6. “What about the other hundred kiddos who don’t have an educated mom? Even my husband says all the time, “if it wasn’t for [parent name], I’d have been twiddling my thumbs four years ago, being like, what the hell do I do?””
7. “there are not speech providers that understand hearing loss and autism and how to bring those two together.”
8. ‘someone has to advocate for these little guys to get the proper services because nobody knows really knows what to do with them.”
9. “It’s a constant work in progress to try and find. I mean, I’ve looked all over. I don’t know if you guys know of a program that specializes in working with kids with both of these things going on, like I don’t. It seems like it’s all kind of one or the other.”
10. “[We’re] talking about a population of autistic users who need more support.”
11. “I think unless you do some extra program, you're not going to get a lot at all.”

12. “We don’t get a lot of instruction in autism in our graduate programs. We don’t get a lot of instruction on hearing loss in our graduate programs.”
13. “If you just do the general grad program, no, ‘d probably say not.”
14. “I definitely was not prepared for these kids who have ASD and CIs.”
15. “When you think about SLP’s and everything they have to learn in a two year program, then you can’t really go very deep because you have to go really broad.”
16. “I don’t know that that’s something really that can be changed other than the professionals taking upon themselves the opportunity to specialize.”
17. “How do we make continuing education really valuable in our specialties? And really accessible, making sure that if you’re gonna be working with children who have autism, you are able to get more training because you’re probably not gonna be able to get it in your AUD program.”
18. “On the job experience is big and I think finding somebody to mentor you that has those skills, and that experience is big too.”
19. “If you’re not good at what you do, that kid’s gonna pay for it.”
20. “Your familiarity with autism and um yeah, your skills, your experience, with that does play a part absolutely. That’s a big part of the kid’s success, which definitely puts a lot of pressure on you.”
21. “His hearing loss is not affecting his academic career, his autism component is.”
22. “[Child name] wears [their] implants, Neptune style. So, clips on the back [because they] have sensory averse behaviors over the ear.”
23. “It’s a work in progress right now, getting them over the ear.”

24. “The audiologist] turned on [their] implants and programmed them and mapped them out and really set [child’s name] up to be successful and not have sensory overload to [their] environment.”
25. “[They’re] definitely sensory aversive when it comes to touch.”
26. “[Child’s name] doesn’t like wearing long sleeves and generally dislikes a lot of clothing.”
27. “[They are] definitely aversive to overwhelming social environments.”
28. “I’ve even asked the school to um I’ve had a letter from an OT to say, you know, if you can incorporate [sensory activities] in the morning and afternoon, that would be great.”
29. “[They’re} a seeker.”
30. “There’s not a whole lot that [they’re] super averse to.”
31. “If it coils off, you know [they try] to put it back on, [they don’t] like it when they’re not on.”
32. “[Child’s name] has a special chair with a TheraBand at the bottom of it where [they’re] kicking it instead of sitting on the carpet kicking [their] peers.”
33. “Doing otoscopy on them is impossible.”
34. “If there was a loud sound or something highly sensory, as far as auditorily speaking, that would put [the patient] kind of into a downward spiral. And we had to start over again.”
35. “Most of the time, I’m convinced that what I’m getting is minimal response. Like that’s not your true threshold, but I don’t know what your true threshold really is.”
36. “They certainly are challenging in terms of testing.”
37. “They’re certainly challenging, but I think it’s rewarding.”

38. “A lot of typical hearing kids that have autism end up signing anyways or using an AAC device anyways, so it’s kind of like why are we going to put them through this surgery if we’re not even going to be able to measure benefit?”
39. “[There’s a] focus on objective testing, but then you run into those sensory difficulties saying how much objective testing can I do or how much will they tolerate.”
40. “I think they’re a population that probably gets written off very quickly.”
41. “We rely so much on speech perception testing, whether it be words, whether it be sentences, whatever that is. And very often, these kids are nonverbal or limited, or echolalic, but not in a functional way, and so it’s like, how do we measure awareness?”
42. “The majority of my kids with autism prefer their off ear processors, umm, and I don’t know if it’s because it’s a little less of a tone hook touching their pinna… but I have 4 kids that just don’t tolerate an on the ear processor and thankfully we have the ability to have an off the ear processor.”
43. “There’s a few hurdles that we that we that we oftentimes run into sensory, I think being a huge one.”
44. “He wears that thing like 12 hours a day. As long as nothing is touching his ear.”
45. “It’s gonna impact wear time. It’s gonna impact compliance of use.”
46. “We take the batteries out of the device. But just put it on the head, right. So, there’s no sound. And sometimes we start there.”
47. “We certainly have kids who are on the spectrum, who have sensory defensiveness that is not related to sound, which is not impacting, you know, their overall development of speech and spoken language. But I would say for sure, if you’re sensory defensive related to auditory input, then that’s going to be a problem.”

48. “There’s got to be something to that. Those sensory seekers are more OK with more sound.”
49. “I think that’s an important thing to like kind of keep in the back pocket too is knowing that there are different styles.”
50. “Along our journey we were lucky enough to have an a strong audiologist team where it was [provider names] and between the two of them they collaborated really, really well.”
51. “The one person…that I had to really fight for was [provider name] and without her, we would not be the parents that we are and [child name] would not be doing as well as [they are] without her expertise.”
52. “She’s very willing to learn and. And she’s super motivated, too. I like her a lot.”
53. “I feel like the professionals that we have worked with have all been wonderful.”
54. “[They have] the hearing loss. [They have] ADHD and DMDD. I kind of felt like any of the concerns that we were having could kind of be explained by some of these other diagnoses.”
55. “[They] would have been diagnosed as autistic like you know a lot earlier had the hearing loss aspect been taken aside.”
56. “When there’s so many overlapping things having to weed out and you know, oftentimes even the professionals didn’t always know, I feel like it was a lot of guess and check.”
57. “[Their] sound booth um testing didn’t match his sedated ABR test.”
58. “[They] would pick up on Ling sounds then [they] wouldn’t pick up on Ling sounds. [They were] babbling the Ling sounds then [they weren’t] babbling the Ling sounds.”
59. “I was becoming more concerned, like, what’s going on here, like [they] used to say, like, at least a few things. And then [they] completely stopped.”

60. “There’s something else going on here that’s complicating this, [they] should be further along than this.”
61. “[The clinicians] never really told us how far behind [they were] or they never told us, you know, they just say “oh [they’re] making progress and that’s what we want to see.”
62. “The slope of the trajectory was never part of the conversation. And they were always very, very vague.”
63. “Ultimately ended up being [the] autism component, umm, and not [the] hearing loss because the hearing loss component was ruled out because we knew [they were] hearing.”
64. “[They] would bang [their] head…[they] would spin in circles…behaviors as far as hitting, kicking, biting, hair pulling, head banging, at that point, [they were] throwing [their] hearing aids and actually right after for about 6 months or so, [they were] throwing [their] implants too.”
65. “[They’re] in this, like, screaming stage…It’s not associated with how his implants are programmed or the environment of it being too loud or not. We’ve done so much… to ensure that it’s not a sound input sensory behavior. It’s an autism attention seeking behavior.”
66. “These two worlds for the most part, don’t really get along very well.”
67. “It wasn’t like she was like, oh, [they’re] autistic. But I’m not going to be flexible at all. I think she just didn’t know what to do.”
68. “Sometimes, you know [they have autism], even before the parent knows which is hard. Because then you feel like you’re keeping a secret from them when you’re not. You just,

it's not your place. It's not, you know, you're there to treat the hearing loss. You're there to support in whatever way you can."

69. "I think that when I already have a diagnosis of autism for a patient, I think it makes it easier."
70. "Children have hearing loss so much earlier oftentimes professionals with good intentions default to this is a behavior indicator that is associated with hearing loss."
71. "[That child] is already carrying that diagnosis. So until they start to see things that don't look like hearing loss then we don't really make any headway in appropriate diagnostics."
72. "Those of us who know what hearing loss looks like, we watch them for a couple of days and we think, "Mmm, there's more going on here.""
73. "Does early newborn hearing screening, early intervention help or hurt [the autism] diagnostic process? I think it depends…generally we're moving to the implantation before we're moving to the diagnosis of autism because of early intervention."
74. "It's hard for us to say, well, I can kind of help you understand why your child won't be speaking, because I know he has autism, but nobody has gone that far to tell you that yet."
75. "I wish I could say that I had ever gotten a child that I had that benefit."
76. "I've had families that did not [have an autism diagnosis], and I had to counsel families saying, "this is not my specialty, but of the behaviors that I'm noticing, I think it would be good to get a developmental evaluation.""

77. “They do always show similar. So I always tell them, like, I need you to have a good hearing test done first, because if you don’t have access to the language, a lot of the behaviors that we’ll see mimic each other.”
78. “Some of these kids are not getting diagnosed, umm, with autism until well after their hearing loss has been diagnosed potentially after they’re implanted, depending on age of implantation.”
79. “As time has passed, we have seen both of those things move younger and younger.”
80. “One of the things that we’ve seen for sure is younger and younger diagnosis of both and earlier intervention for both.”
81. “The real obvious autism signs don’t show up, you know, at two months old or whatever.”
82. “I would say more often than not, we know that they have a hearing loss before we know they have autism.”
83. “I don’t think that there should be a stereotypical autistic person because you know, autism is such a [spectrum].”
84. “He wasn’t what they envisioned as, you know, an autistic kid and so um they just thought, you know, he’s being difficult.”
85. “Autism is so different to begin, like it’s, you know, the characteristics present differently.”
86. “[There’s] implicit bias. I think they really are big on wanting their numbers to look good of success.”
87. “I think because [autism and hearing loss] are a spectrum, [language development] just really depends on the child.”

88. “The biggest thing they all have in common, is that they have nothing in common.”
89. “It’s rage provoking in me that [centers] are denying these kids the opportunity [to have access to sound]. Like why? Even if they don’t have expressive language in the way that they want or define it, why don’t they have the opportunity to have access to sound?”
90. “It would be just more of a cautious prognosis because communication is already compromised as a part of the autism diagnosis.”
91. “Reasonable expectations…is such a stupid phrase.”
92. “We always want to presume confidence.”
93. “Don’t limit yourselves. Don’t predisposition the families.”
94. “I am not oblivious to the fact that there’s plenty that I can still [do]. But I also have intended to learn as much as I can and to reach out to people um who I know have more experience than I do.”
95. “Putting them into one group is quite a challenge.”
96. “We had four therapists and nobody was on the same page.”
97. “Everybody wanted to do their own approach. Everybody had their own expertise.”
98. “[It’s hard] just finding that right therapist that knows how to tackle both components.”
99. “There really aren’t services out there for. Targeted, I would say for kids with hearing loss and autism.”
100. “There was zero parent coaching. There was zero, like, telling me what to do at home. So I’d just be sitting there.”
101. “Especially with kids with autism who really really need and thrive better with that predictability being able to know what’s going on [is huge].”

102. “One of her accommodations is to sit close to the front and I talk to all her teachers at the beginning of the year and her math teacher was like, well, she said she can hear me fine if she sits in the back of the class by the air conditioner, and yet she has a D in math.”
103. “The school is kind of like, well, if she needs something, just let us know. It’s hard for her to…remember what her accommodations are to ask [to use them], and she feels embarrassed.”