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Review

Current trends in outcome studies for children with hearing loss and the need to establish a comprehensive framework of measuring outcomes in children with hearing loss in China

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Abstract

Since the 1970s, outcome studies for children with hearing loss expanded from focusing on assessing auditory awareness and speech perception skills to evaluating language and speech development. Since the early 2000s, the multi-center large scale research systematically studied outcomes in the areas of auditory awareness, speech-perception, language development, speech development, educational achievements, cognitive development, and psychosocial development. These studies advocated the establishment of baseline and regular follow-up evaluations with a comprehensive framework centered on language development. Recent research interests also include understanding the vast differences in outcomes for children with hearing loss, understanding the relationships between neurocognitive development and language acquisition in children with hearing loss, and using outcome studies to guide evidence-based clinical practice. After the establishment of standardized Mandarin language assessments, outcomes research in Mainland China has the potential to expand beyond auditory awareness and speech perception studies.

Keywords: Outcome assessment; Language assessment; Mandarin; Standardized assessment; Hearing impaired children

Contents

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1. History

What is the ultimate goal of audiological intervention and aural rehabilitation for children with hearing loss? This question was pondered by great minds like Thomas H. Galladet and Laurent Clerc, who established the American Asylum for the Education of the Deaf and Dumb (now named the American School for the Deaf) in the United States in 1817. The goal of the program was to teach "deaf children" how to communicate, give them an education, and allow them to have a social life through manual communication (i.e. signing). Another group of pioneers include Greene Hubbard, who established the Clarke School (now called the Clarke School for Hearing and Speech) in 1863 and Alexander Graham Bell, who set up the American Association to Promote the Teaching of Speech to the Deaf (now called the Alexander Graham Bell Association for the Deaf and Hard of Hearing) in 1890. These pioneers advocated rehabilitation be centered on oral communication, instead of manual communication rehabilitation, to prepare a child with hearing loss for education and social development primarily through residual hearing, lip reading and tactile cues many years before the advent of hearing technology. Experts from both the manual communication camps and the oral communication camps have debated for over 100 years on the best ultimate communication goal for children with hearing impairment.

2. A framework of outcome assessments

Over the last two decades, universal newborn hearing screenings (UNHS), more sophisticated hearing assessment methods, and advanced hearing technologies such as digital hearing aids and cochlear implants, significantly contributed to the interest in research on outcome studies for children with hearing loss whose intervention and rehabilitation emphasizes developing listening and speaking skills. The ultimate goal for these interventions and rehabilitation procedures is for a child with hearing loss to develop language and speech through listening, receive a mainstream education, acquire social skills with normal hearing children, and potentially have a career among the normal hearing population. As a result, a framework of outcome measurements is needed to study the outcomes in the development of these different areas.

Outcome studies in the areas of language development, education, and psychosocial behavior have existed within the field of deaf education since the 1970s (Davis, 1977; Davis et al., 1981, 1986). A framework of a pediatric outcome evaluation after audiological intervention (e.g. fitting of appropriate hearing technology) and during aural (re)habilitation includes the following components:

a) Assessment of auditory awareness and sound discrimination,
b) Speech recognition (or speech perception) assessment: speech perception is the process by which a perceiver internally generates linguistic structures believed to correspond with those generated by a talker (Boothroyd et al., 1991),
c) Language development assessment,
d) Speech development assessment,
e) Play/cognition skills assessment,
f) Social communication skills assessment,
g) Other related assessments (e.g. sensory processing, sensory integration, sensory-motor, academic skills, and quality of life assessments).

3. Speech perception assessments: initial focus of outcome studies for implanted children

The early outcome studies from the Melbourne clinic for cochlear implantation in children focused on speech perception assessments, especially in the area of open-set speech understanding (Clark et al., 1987; Dawson et al., 1989). These outcome studies influenced early clinical trial study in the United States for the Food and Drug Administration (FDA) (Staller et al., 1991). Speech perception assessments have played an important role in assessing outcomes of hearing aid and cochlear implant use in children with hearing loss (Barker and Tomblin, 2004; Boothroyd et al., 1991; Boothroyd, 2004; Davidson et al., 2011; Geers et al., 2003a,b; Houston et al., 2003; O'Donoghue et al., 1999; Psarros et al., 2002; Seyle and Brown, 2002; Snik et al., 1997; Young et al., 1999).

4. Language development assessments: evaluation of the developmental impact of speech recognition skills

As early as the 1970s, Vandenberg (1972) used language development assessments in outcome studies for children who wore hearing aids. Hasenstab and Tobey (1991) measured language development of children with cochlear implants (CIs). Initial reports of outcomes for CI users (Coerts and Mills, 1995; Dawson et al., 1995) and comparisons of CI and hearing aid use in children (Geers and Moog, 1994) also used language development assessments. The *100th NIH Consensus Development Conference* keynote speech entitled *Cochlear Implants in Adults and Children* recognized the importance of language development assessments for outcome studies in children with CIs (Ruben, 1995). An editorial article in the *International Journal of Pediatric Otorhinolaryngology* boldly stated, “The fundamental goal of implantation in the linguistically developing child should be the linguistic development of that child. The usual measures of sound and speech detection, speech production, etc., are secondary and ultimately unimportant when compared to language function. All studies of the CI in the linguistically developing child must measure and evaluate the child's language” (Ruben, 1995). Research on language development outcomes in children using CIs continued through the 1990s (Blamey et al., 2001; Bollard et al., 1999; Connor et al., 2000; Miyamoto et al., 1999; Moeller, 2000; Tomblin et al., 1999).

The language assessments that are commonly used in outcome research studies are norm-referenced standardized assessments with a few criterion-referenced assessments. Some examples of these assessments are the MacArthur Communicative Development Inventory (Fenson et al., 2007), Rossetti Infant-Toddler Language Scale (Rossetti, 1990), Reynell Developmental Language Scale (RDLS) (Edwards and Reynell, 1997), Clinical Evaluation of Language Fundamentals (CELF) (Wiig et al., 2003), Preschool Language Scales (PLS) (Zimmerman et al., 2011), Comprehensive Assessment of Spoken Language (CASL) (Carrow-Woolfolk, 1999) the Peabody Picture Vocabulary Test-4 (PPVT) (Dunn and Dunn, 2007), Test of Auditory Comprehension of Language (TACL) (Carrow-Woolfolk, 1998), Test of Reading Comprehension (TORC) (Brown et al., 1995), and Woodcock Reading Mastery Test (Woodcock, 1987; Blamey et al., 2001; Blamey and Sarant, 2002; Bollard et al., 1999; Connor et al., 2000; Davidson et al., 2011; Dettman et al., 2007; Eisenberg and Ying, 2004; Moog and Geers, 2010; Tomblin et al., 2000).

Outcome studies in language development skills also played an important role in researching best practices in hearing impairment identification. Yoshinaga-Itano and others in the 1990s compared the language development of the children with hearing loss who were identified at different ages (White and White, 1987; Stredler-Brown and Yoshinaga-Itano, 1994; Robinshaw, 1995; Apuzzo and Yoshinaga-Itano, 1995; Moeller, 1996; Yoshinaga-Itano et al., 1998). These outcome studies, using language assessments, demonstrated better language development in early identified children with hearing impairment who received early intervention. These studies became strong evidence for the development of new medical policies in newborn hearing screening. In other words, the Universal Newborn Hearing Screening (UNBHS) was birthed as a result of these early outcome studies.

5. A framework of outcome assessments from research to clinical protocol: the role of multicenter large scale outcome studies

In the 2000s, major outcome studies involved multicenter data collected across the United States and Canada that influenced the direction of the research studies in the field and also provided guidance for clinical practice for children with hearing loss. Geers and her colleagues led a multicenter study with University of Texas at Dallas, University of Texas Southwestern Medical Center, Indiana State University, University of Colorado at Boulder, Washington University Medical Center, and Moog Center for Deaf Education. These data examined different factors that influenced outcomes in children with cochlear implants (Table 1). Categories of influencing factors included: (1) educational environment, (2) family variables, (3) the child's medical history, and (4) his/her history of interventions and rehabilitation. Outcome measures spanned multiple domains including auditory awareness, speech perception, language development and speech intelligibility, speech fluency, phonological processing and reading skills, working memory span, psychosocial adjustment, and family adjustment (Davidson et al., 2011; Geers, 2003; Geers and Brenner, 2003; Geers and Hayes, 2011; Geers et al., 2003a,b; Geers and Sedey, 2011; Moog and Geers, 2003; Moog et al., 2011; Nicholas and Geers, 2003; Pisoni and Cleary, 2003; Pisoni et al., 2011; Tobey et al., 2003; Tobey et al., 2011; Tye-Murray, 2003; Uchanski and Geers, 2003).

The Childhood Development after Cochlear Implantation (CDaCI) studies, included six collaborating centers: Johns Hopkins University, House Ear Institute, University of Texas at Dallas, University of Miami, University of North Carolina, and University of Michigan. These multicenter national cohort studies included CI children and normal hearing (NH) peers. The objective of these studies was to compare children who had undergone cochlear implantation with similarly aged hearing peers across multiple domains, including auditory performance (i.e. auditory awareness skills and speech perception skills), oral language development, psychosocial and behavioral functioning, and quality of life (Barker et al., 2009; Cruz et al., 2013; Cruz et al., 2012; Eisenberg et al., 2006; Fink et al., 2007; Lin et al., 2008; Markman et al., 2011; Niparko et al., 2010; Quittner et al., 2010; Wang et al., 2008).

Initially, the CDaCI team expanded the Central Institute for the Deaf hierarchy (Geers, 1994) and the Indiana University School of Medicine protocol (Kirk, 2000) to establish test batteries to measure speech perception skills (Eisenberg et al., 2006). Based on the child's age and functional hearing abilities, they developed two batteries. The preschool battery included the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS) (Zimmerman-Phillips et al., 2000), Meaningful Auditory Integration Scale (MAIS) (Robbins et al., 1991), Early Speech Perception Test (ESP)-low verbal (Moog and Geers, 1990), Early Speech Perception Test (ESP)-standard version (Moog and Geers, 1990), Pediatric Speech Intelligibility Test (PSI) (Jerger and Jerger, 1984), Multisyllabic Lexical Neighborhood Test (MLNT) (Kirk et al., 1995a,b), and Lexical Neighborhood Test (LNT) (Kirk, 1998). The school battery added the Phonetically Balanced Word List-Kindergarten (PBK) (Haskins, 1949) and Hearing in Noise Test – Children (HINT-C) (Gelnett et al., 1995). They demonstrated a speech recognition hierarchy using these test batteries (Eisenberg et al., 2006). A summary score, the speech recognition index in quiet (SRI-Q), can be used to track

Table 1
Different factors that influenc outcomes in children with cochlear implants.

Outcome measures		Speech perception	Speech production	Spoken language	Total language	Reading
Factors	Child and Family		Female	Female	Female	Female
					Later onset of deafness	Later onset of deafness
		Higher IQ	Higher IQ	Higher IQ	Higher IQ	
		Smaller family	Smaller family	Smaller family	Smaller family	
			Higher SES	Higher SES	Higher SES	Higher SES
	Implant characteristics	SPEAK processor	SPEAK processor	SPEAK processor	SPEAK processor	SPEAK processor
		More electrodes	More electrodes	More electrodes	More electrodes	
		Larger dynamic range	Larger dynamic range	Larger dynamic range	Larger dynamic range	Larger dynamic range
		Loudness growth	Loudness growth	Loudness growth	Loudness growth	
	Education setting	Oral classroom	Oral classroom	Oral classroom	Oral classroom	
			Mainstream class			Mainstream class

Adapted from Ear and Hearing 2003; 24; 121S–125S.

emergent auditory awareness and speech perception skills during the first two years after cochlear implantation in children (Wang et al., 2008).

The multicenter CDaCI studies established a new standard for comprehensive outcome studies. They established multi-dimensional standardized baseline assessments included demographics, hearing and medical history, communication history, cognitive tests, an audiological exam, speech recognition, language tests, psychosocial assessment including parent-child videotapes and quality of life measured by parent report. Their multi-dimensional baseline assessments demonstrated the importance of a comprehensive framework approach in outcome studies for evaluating children with a hearing loss. The specific standardized baseline assessments included the following:

a) Auditory awareness assessment (i.e. IT-MAIS, MAIS)
b) Speech perception assessment (i.e. ESP, PSI, MLNT, LNT, PBK, HINT-C)
c) Language development assessment (i.e. RDLS, MBCDI, OWLS, PLS)
d) Speech development assessment (i.e. Brown)
e) Play/cognition skills assessment (i.e. The Symbolic Play Test)
f) Social communication skills assessment (i.e. The Social Skills Rating System)
g) Quality of life

These large-scale longitudinal studies conducted a full battery of follow-up assessments at six-month intervals. The CDaCI team stated that, “Overall, the CDaCI project is unified by a focus on early language learning” (Fink et al., 2007). Thus, these studies investigated the multiple parameters of early developmental learning, particularly as it relates to spoken language in children with hearing loss receiving advanced hearing technologies. They promoted a comprehensive framework of outcome assessments that emphasized tracking spoken language development and early developmental learning that is related to spoken language development. The CDaCI team believed this comprehensive framework baseline and regular follow-up assessment design offered “the best prospect for quality outcomes data on which parental decisions should be based” (Fink et al., 2007).

Based on the outcome research studies, clinical practice protocols in major cochlear implant centers in the United States (e.g., University of Texas at Dallas Callier Center for Communication Disorders, Hearts for Hearing Center, and Boys Town National Research Hospital) all include an initial baseline evaluation comprised of a battery of assessments performed immediately after audiological intervention (i.e., hearing aids, cochlear implants, and FM systems). The baseline assessment provides audiologists and speech-language pathologists with information to guide them in programming the hearing instrument appropriately and setting up individualized rehabilitation goals to facilitate early language learning.

The follow-up assessments are more frequently administered during the first year after the baseline assessment and then at six-month intervals to track the child's progress in different components. The results guide audiologists in making adjustments to the programs within the hearing instruments and help speech-language pathologists modify language, speech, and social communication goals.

6. A comprehensive framework of outcomes including auditory, language, intervention, speech, social-emotional and quality of life global assessments

A review of the CDaCI studies reveals their research emphases started from auditory focused assessments and moved to language-centered comprehensive outcome measurements and intervention. In 2006, the CDaCI research team reported speech recognition skills at 1-year post cochlear implantation (Eisenberg et al., 2006). In 2007, they published their design and baseline characteristics describing the framework of assessments and emphasizing the importance of developmental language assessments (Fink et al., 2007). In 2008, they reported a thorough longitudinal study on speech recognition skills. They established a hierarchical order of the most commonly used speech recognition assessments, which was adopted by major CI centers as a guide for choosing the most appropriate speech perception tests (Wang et al., 2008). The CDaCI research team also demonstrated that language skills were more positively associated with parental perception of development than with a selected measure of closed-set speech perception (Lin et al., 2008). In 2009, the team explored factors influencing language development in these children with CIs and demonstrated language development was influenced by attention (Barker et al., 2009). In 2010, the CDaCI research team reported that earlier age at implantation, greater residual hearing prior to CI, higher ratings of parent-child interactions, and higher SES were associated with greater rates of communication growth in comprehending and expressing language (Niparko et al., 2010). In 2011 (Markman et al.), they further examined a list of factors influencing language development after CI and demonstrated different domains of language (e.g., phonetics, vocabulary, grammar, and pragmatics) were affected differently by these factors. Together with what has been shown by Quittner et al. (2010), namely that delayed exposure caused disruptions in the social/affective process of parentally guided language learning, Markman et al. (2011) proposed an epigenetic model hypothesis for language development in children with hearing impairment. In 2012, they studied language and behavior outcomes for a specific population among children with CI – children with developmental disabilities (Cruz et al., 2012). In 2013, Tobey et al. followed up with the CI children in the CDaCI research to study their language skills when they were already school-aged. Higher standard language scores in expressive vocabulary, expressive syntax, and pragmatic judgments were associated with children implanted before 2.5 years. At the same time, in both early and later implanted groups, large variability in language outcomes was observed, with some children performing more than two standard deviations below the standardization group mean, while some children scored at or above the mean. Cruz and colleagues (Cruz et al., 2013) examined parent-child interactions for subjects in the CDaCI research study. They found that higher-level facilitative language techniques predicted growth in expressive language and total number of different words used by the parents predicted growth in receptive language during the first 3 years post implantation. These findings provide guidance for language interventions for children with hearing impairment. Deficits in joint engagement skills in deaf children were associated with poor oral language development. Therefore, they proposed "interventions that promote a richer language environment and foster positive, dyadic interactions may be helpful in increasing the use of symbols in play and thus, indirectly affect the growth of oral language." (Cejas et al., 2014).

Other studies conducted in the large multicenter research echo the findings in the CDaCI studies. In 2010, Moog and Geers studied the relationship between early education placement and later language outcomes from thirty-nine listening and spoken language (LSL) programs located in 20 different states across the United States. They concluded that the possibility for CI children to reach normal language levels by kindergarten increased significantly if implantation and early parent-infant intervention started by age one and was followed up at age two with an intensive toddler class specifically designed for developing spoken language. Other studies in the large multicenter research documented positive psychosocial outcomes of children who received CIs (Nicholas and Geers, 2003; Moog et al., 2011). Wiefferink et al. (2012), examined the relationship between language development and social functioning and emotion regulation in children with CIs. Their results indicated CI children have less adequate emotion-regulation strategies and less social competence than normal hearing peers. Better language skills in CI children were associated with higher social competence and fewer observed behavior issues. The authors advocated for longitudinal studies to further explore the development of emotion regulation and social functioning in children using CIs. Clark et al. (2012) assessed CI children's health-related quality of life (HRQL) and development through parental reports. They concluded the validity of parental global assessments, such as the HRQL measurement, is supported by speech perception and language development outcome measures. Recently, Meserole et al. (2014) reported that higher family stress was negatively associated with parent-reported health-related quality of life (HRQOL) outcomes for families with CI children and advocated for the potential need for family therapy.

Outcome studies were also conducted for speech sound development for children with CIs. Tobey et al. reported lower overall accuracy of consonant production of children who received CIs before 5 years of age than hearing children with similar hearing ages (Tobey et al., 2003). Connor et al. (2006) compared overall accuracy of consonant production between early and later implanted children. They found children who were implanted before 2.5 years of age exhibited early bursts of growth in consonant-production accuracy and had overall better speech production compared with same age peers who were implanted at later ages. Other studies Schorr et al. (2008) and Tomblin et al. (2008) also demonstrated implanted children may show improvement in speech production accuracy over time. Schorr et al. showed children with CIs did not reach typical levels of speech production even after more than 5 years of CI experience, while Tomblin et al. (2008) showed the average standard scores for a speech production assessment for children with at least 5 years of CI experience was 93.54 (SD = 18.94), a score which was not significantly different from the hearing children. The discrepancy across findings may be confounded by the wide range of age of implantation. Besides speech sound accuracy, sound production inventories and error patterns were also examined in other studies. Warner-Czyz and Davis (2008) reported consonant and vowel inventories and error patterns during the single-word period in four normal hearing and four CI children. Both groups improved over a period of 6 months and showed similar patterns for segmental accuracy. For the children with CIs, stops and nasals were used more frequently than fricatives. In the area of place of articulation, labials and coronals were used more frequently than dorsals. Recent studies of speech production with early implanted children revealed children implanted before 30 months had similar patterns of acquisition in their consonant repertoires and standardized speech sound assessment scores when compared to their normal hearing age matched children (Spencer and Guo, 2013). Children with early implantation and early auditory-verbal language therapy met developmental expectations by 3- and 4- years of age for consonant cluster production (Fulcher et al., 2014).

7. Moving from short-term to mid-term outcome studies and expanding to long-term outcome studies

Ear and Hearing 2003 Special Edition on Cochlear Implants focused on outcomes for prelingually deaf children who received CIs before five years of age and after three years of device use with rehabilitation. In 2010, *Ear and Hearing* had another special edition on cochlear implants. The 2010 supplement focused on long-term outcome studies after over ten years of device use and rehabilitation for the same group of children who participated in the studies in the 2003 special edition. Both compilations of studies demonstrated that both mid-term and long-term speech, language, and reading outcomes were better for children whose educational environment emphasized auditory-oral communication versus total communication (Moog and Geers, 2003). In the 2003 studies, children who were implanted at 2, 3, or 4 years of age did not perform differently in their speech, language, and reading outcomes by 8 years of age. The investigators concluded that "any advantage associated with implanting children at age 2 years instead of 4 years may no longer be apparent by 8 years of age". This was also consistent with findings of Dunn et al. (2014) that the positive effects of age of implantation in preschool became not significant for both receptive and expressive language at ages 7–10 years. However, when these same children were re-examined for long-term outcome studies, it was demonstrated the children implanted at the youngest ages (2 years in this sample) were more likely to achieve age-appropriate language and reading skills in high school compared to children implanted at later ages (4–5 years) (Geers and Sedey, 2011; Geers and Hayes, 2011; Geers and Nicholas, 2013). From elementary school to high school, significantly improved performances in speech perception and speech intelligibility were observed (Davidson et al., 2011; Tobey et al., 2011). About 70% of the high school children had standard language scores within normal limits; about 50% of them had similar reading skills to their normal hearing age peers, and an even fewer percentage of them had comparable writing skills to their normal hearing age peers (Geers and Sedey, 2011; Geers and Hayes, 2011). Variation in speech production, language, reading, and writing skills were observed in these CI children even after they reached high school age.

8. Expanding from severe-profound hearing loss to mild-severe hearing loss

Systematic multicenter longitudinal outcome studies focusing on children with severe to profound hearing loss provided valuable clinical guidance. The National Institutes of Health funded a five year study from 2008 to 2013 to examine longitudinal outcomes. To this end, a large scale longitudinal outcome study of 400 children with a mild to severe hearing loss and 150 normal hearing children was conducted at the University of Iowa, Boys Town National Research Hospital, and the University of North Carolina. Outcomes in speech perception, language development, speech production, education achievement, psychosocial behavioral development, and quality of life/family life were reported in the *Ear and Hearing 2015 Special Edition on Outcomes of Children with Hearing Loss (OCHL)* in relationship to background characteristics of children with mild to severe hearing losses and their families, intervention services factors, hearing aid types and fitting factors. The following conclusions were summarized in the Epilogue of the special issue (Moeller et al., 2015a, 2015b): 1. Children with mild-to-severe hearing loss are at risk for language development delay, and the risk increases with the severity of unaided hearing levels (Tomblin et al., 2015); 2. Provision of well-fit hearing aids reduces risk and provides some degree of protection against language delay. Greater aided audibility is associated with better outcomes in preschool (McCreery et al., 2015a); 3. More than half of children's hearing aids were not fit optimally, which negatively impacted aided audibility (McCreery et al., 2015a); 4. Early hearing aid provision results in better early language outcomes, but later-fit children demonstrated accelerated growth patterns once aided (Tomblin et al., 2015); 5. Consistent hearing aid use provides some protection against language delay and supports auditory development (Tomblin et al., 2015); 6. Qualitative dimensions of caregiver input influence child language outcomes (Ambrose et al., 2015); 7. Both receptive language abilities and aided audibility influenced children's functional auditory and speech recognition skills (McCreery et al., 2015b); 8. A child who is hard of hearing appears to be at particular risk for delays in structural aspects (i.e., form) of language (Tomblin et al., 2015); 9. Sole reliance on norm-referenced scores may overestimate the outcomes of a child who is hard of hearing (Tomblin et al., 2015); 10. Aided audibility, hearing aid use, and characteristics of the language environment interact to moderate the influence of hearing loss on children's outcomes (Ambrose et al., 2015, McCreery et al., 2015a, 2015b; Tomblin et al., 2015; Walker et al., 2015).

9. From excluding children with disabilities to special studies focusing on children with disabilities

Several reports showed up to 30–40% of children with hearing impairments have at least one additional disability (Van Naarden and Decouflé, 1999; Picard, 2004; Gallaudet Research Institute, 2011). Recently, many studies indicated consistent improvement in speech perception, language comprehension, and language production for implanted children with developmental disabilities (Cruz et al., 2012; Youm et al., 2012). Wiley et al. (2012) also reported daily functional ability outcomes in children with CIs and associated disabilities. They concluded that these CI children made progress in functional skill development and receptive language skills appeared to play a key role in the social functioning in these children. Another study (Birman et al., 2012) reported additional disabilities significantly influenced the outcomes of CIs in speech perception and language development during the first year. Steven et al. (2011) demonstrated cognitive status was the most important prognostic indicator regarding outcomes. Some researchers reported positive outcomes in either speech perception skills and/or language development skills for populations with a specific disability, such as cerebral palsy (Steven et al., 2011; Lamônica et al., 2014), Usher syndrome type 1 (Henricson et al., 2012), congenital cytomegalovirus infection accompanied by psycho-neurological disorders (Yamazaki et al., 2012), CHARGE syndrome (Birman et al., 2015), and autism spectrum disorders (Meinzen-Derr et al., 2014; Eshraghi et al., 2015). Further research is required to help guide expected outcomes of implantation of this diverse population.

10. Growing interest in psycholinguistic and cognitive processing studies to investigate possible reasons for the vast differences in outcomes

Overall findings from large studies revealed early implanted children do not achieve language skills comparable to their hearing peers on average and there is a great variability in performance on language assessments (Niparko et al., 2010; Tobey et al., 2013; Geers and Nicholas, 2013). Ching et al. (2013) reported performances averaged one SD below the normative mean in a comprehensive outcome measurement (i.e., auditory function, receptive and expressive language and speech production skills) even after adjusting for additional disabilities. Fitzpatrick et al. (2011) also reported more variable language outcomes in children with a 70–90 dB range of hearing losses using hearing aids compared with their normal hearing peers.

Development of multimodal processing during the first months of infancy is important for early language development (Fagan and Pisoni, 2009). Recent theoretical work in speech perception and language development suggests language processing may partly involve underlying attentional and neurocognitive mechanisms that are part of other cognitive domains (Ullman, 2004; Behme and Deacon, 2008; Conway and Pisoni, 2008).

In the past decade, considerable research interest was shown in understanding the relationships between neurocognitive development and language acquisition in children with CIs. Houston et al. (2012) reported "early auditory deprivation may have a modality-specific effect on processing (i.e., affecting the general cognitive processing of auditory input only) or have a modality-general effect on processing (i.e., affecting the cognitive processing of both auditory and visual input)". A study of executive functions (e.g., working memory) in children with CIs demonstrated poor working memory was associated with poor comprehensive standard language scores and poor speech perception in noise scores but did not have the same association or effect with simple receptive vocabulary scores or speech perception in quiet scores (Beer et al., 2011). In CI children, larger mismatch negativity (MMN) generated in the frontal cortex was reported to be positively correlated with working memory and phoneme discrimination skills, while a stronger activation of the temporal cortex negatively corresponded with phoneme discrimination skills (Ortmann et al., 2013), indicating different auditory speech processing strategies for CI children with good or poor speech perception skills. In most domains of executive function, children with CIs were two to five times at greater risk of clinically significant deficits compared with hearing peers (Kronenberger et al., 2014a,b). Recently, further research in executive function reported, "Verbal working memory and fluency-speed (processing speed during cognitive operations requiring effortful controlled attention) were more strongly associated with speech-language outcomes in the CI users than in the normal-hearing peers. Spatial working memory and inhibition-concentration correlated positively with language skills in normal-hearing peers but not in CI users." (Castellanos et al., 2015). Sequential learning abilities (Conway et al., 2014) and temporal-sequential integration skills measured by the ability to use sentence context to perceive speech (Conway et al., 2014) also were reported to be poorer in CI children compared with their hearing peers. Conway et al. (2011) looked at motor sequencing skills and language development in children with CIs. Their study indicated auditory deprivation before cochlear implantation influenced motor sequencing skills. Motor sequencing disturbance was significantly correlated with language development in children with CIs. Bharadwaj et al. (2012) documented differences in multisensory processing development in children with CIs. They found children with profound hearing losses performed significantly below average performance on two temporal processing tasks when compared to a normative group. These recent findings demonstrated the complexities in the relationships between underlying neurocognitive processing and auditory, language, and speech outcomes for children with hearing impairment. Researchers from Indiana University described this complexity as "The ear is connected to the brain" (Houston et al., 2012).

11. Outcome studies guide research and clinical decisions

11.1. Outcome studies provide guidance in audiological interventions

A series of evidence-based systematic review articles discussed the best practices of pediatric hearing aid fitting (McCreery et al., 2015a, 2015b). These comprehensive reviews compared different types of amplitude compression, discussed the efficacy of digital noise reduction and directional microphones, and compared frequency lowering processing with conventional processing amplification. They measured the outcomes of audibility, speech perception skills, language and speech development, and self or parent-report in pediatric hearing aid users.

Outcome assessments also were used in studies exploring best practices in age of implantation. By the year 2000, the FDA lowered the eligibility age for pediatric cochlear implantation to 12 months. Researchers continued to explore potential outcome benefits of cochlear implantation in children younger than 12 months. Waltzman and Roland (2005) reported good short-term outcomes (6 months after aural rehabilitation) with auditory awareness and speech perception assessments for children who received CIs during the first 12 months. Dettman et al. (2007) demonstrated better developmental language skills in children who received CIs during the first 12 months (19 subjects) compared to those who received CIs between 12 and 24 months (87 subjects). Miyamoto and colleagues (Miyamoto et al., 2008) did not report a significant difference in the developmental language skills in similar groups. Miyamoto et al. interpreted their results with caution because they had a very small number of subjects within their pool of cochlear implantation before 12 months (8 subjects) and the subject variables were not well controlled. Holt and Svirky (2011) tracked speech perception and language development of children who received cochlear implants before 12 months and those who received cochlear implants after 12 months for at least two years after implantation. Their study indicated speech perception failed to demonstrate a significant benefit in earlier implantation, but the earlier implanted group developed language at a faster rate in both receptive and expressive areas.

Outcome studies were used in providing clinical guidance in unilateral versus bilateral cochlear implantation (Lammers et al., 2014). Consistent evidence indicated better outcomes of bilateral implantation for sound localization (Beijen et al., 2007; Grieco-Calub et al., 2008; Lovett et al., 2010; Murphy et al., 2011; Grieco-Calub and Litovsky, 2012). Consistent evidence also indicated no benefit for speech perception outcomes in quiet even after two years' experience with a second implant (Litovsky et al., 2006; Grieco-Calub et al., 2009; Sparreboom et al., 2011; Nittrouer et al., 2013). Advantages in speech perception outcomes in noise were shown for bilaterally implanted children over unilaterally implanted children or over bimodally fitted children only when the noise was presented from the side of the first or only CI (Lovett et al., 2010; Mok et al., 2010; Sparreboom et al., 2011). Both receptive language and expressive language outcomes were compared between 25 bilaterally implanted children with a carefully matched (e.g., age of implantation, no other disabilities, monolingual environment) control group of 25 unilaterally implanted children. Significantly higher standard scores in both receptive and expressive language assessments were shown for the group with bilateral implantation. In addition, a shorter interval between both implantations was related to higher standard language scores (Boons et al., 2012). Baudonck et al. (2011) found bilaterally implanted children had fewer distortions in speech articulation than the children using only one implant.

Several outcome studies in language development skills supported early intervention/rehabilitation services that facilitated parental engagement in the child's habilitation (Boons et al., 2012; DesJardin and Eisenberg, 2007; Moog and Geers, 2010; Yanbay et al., 2014; Zaidman-Zait and Young, 2008).

11.2. Outcome studies can provide guidance in treatment of specific auditory disorders

Our current understanding of auditory neuropathy spectrum disorder (ANSD) and its management is still at an early stage. Outcome studies recently have played an important role in exploring the best clinical practices for ANSD. Roush and colleagues (Roush et al., 2011) systematically reviewed studies related to audiological management of children with ANSD. Eighteen studies were carefully chosen and examined for methodological quality. Most of the studies reviewed in their report, along with a study by Breneman et al. (2012), reported improved auditory outcomes measured by speech perception tests for children with ANSD who received CIs. Better speech perception outcomes were demonstrated for bilaterally implanted children with ANSD compared with unilaterally implanted children with ANSD (Dean et al., 2013). Rance and Barker (2009) reported comparable developmental language skills in children with auditory neuropathy regardless if they used hearing aids or cochlear implants, suggesting that children with ANSD should not automatically be considered CI candidates. They also demonstrated that there was no significant differences between the implanted auditory neuropathy group and the group with sensorineural hearing loss. A multicenter study (Berlin et al., 2010) indicated good speech perception skills with hearing aids in quiet have not led to age appropriate language skills in the majority of the patients. More recent systematic reviews (Roush et al., 2011; Harrison et al., 2015) advocated that “further research is needed to address other functional aspects including speech, language, learning, social/emotional development, and psycho-educational performance”.

11.3. Objective and functional assessments in outcome studies

Cortical auditory evoked potentials (CAEPs) are important components of outcome assessments. P1, an important aspect of CAEP, played an important role in providing evidence for the existence of a sensitive period for the development of the central auditory system neural plasticity (Sharma et al., 2002). Central auditory maturation measured by P1 latencies and the development of early language communication (assessed by canonical vocalizations) followed a similar developmental trajectory in children implanted early (Sharma et al., 2004). These studies provided evidence for positive outcomes associated with early implantation. Several outcome studies for children with ANSD used both the P1 latency measures and an auditory awareness assessment (i.e. IT-MAIS) (Sharma et al., 2011; Cardon and Sharma, 2013). The results indicated P1 latencies appeared to be a good predictor of behavioral outcomes measured by the IT-MAIS in ANSD patients. Cardon and Sharma (2013) also reported “cochlear implants seem to yield more favorable results in a greater number of children than hearing aids". A large number of research articles have shown behavioral auditory thresholds cannot predict behavioral outcomes in children with ANSD (Rance et al., 2002; Sharma et al., 2011; Cardon and Sharma, 2013) and proposed the possibility of utilizing CAEPs in obtaining auditory thresholds in children with ANSD (Sharma and Cardon, 2015). Collectively, the authors suggested P1 CAEP might provide an useful clinical tool for guiding intervention choices and assessing their efficacy in children with ANSD.

11.4. Trends in outcome studies: a model for best practices for management and outcome assessments research in Mainland China

In the past 10 years within the field of pediatric audiology, outcome researchers in China have made significant progress in developing outcome assessment tools and utilizing outcome studies to provide clinical guidance.

In the area of auditory awareness and speech perception assessments, many assessments were adapted for Mainland children from well-established Western assessments. In 2009, Zheng et al. (2009a) reported the normative data for an auditory awareness assessment, Mandarin Infant-Toddler Meaningful Auditory Integration Scale (MIT-MAIS). From 2004 to now, different groups of researchers developed a series of speech perception assessments. They included the Mandarin monosyllabic lexical neighborhood test (Mandarin LNT) (Yang et al., 2004), the Mandarin Hearing in Noise Test (MHINT-C) (Wong et al., 2005), the Mandarin Early Speech Perception Test (MESP) (Zheng et al., 2009b), the Mandarin Pediatric Speech Intelligibility (MPSI) (Zheng et al., 2009c), and the Mandarin Bamford-Kowal-Bench (BKB)-like sentences (Xi et al., 2012).

In the area of language assessment, in addition to the adaptation of English assessments, efforts were made to develop indigenous Mandarin language assessments for Mainland China. Li and colleagues proposed a simplified short form (SSF) of the Mandarin Communicative Development Inventory to be used as a measurement of vocabulary growth following early intervention with children who have hearing impairment (Li et al., 2014a,b). Considering the challenges of culturally irrelevant items for Mainland children in adapted vocabulary tests (Tardif et al., 2008), Lu et al. (2013) reported the development and standardization of the Mandarin Expressive and Receptive Vocabulary Test (MERVT). MERVT was normed on 245 normal-hearing children ranging in age between 1; 6 to 3; 11 in Beijing, China. MERVT scores strongly correlated with an intelligence test for Mandarin speaking children with CIs (Lu et al., 2013). Cultural and grammatical content irrelevancy advocated the development of a comprehensive indigenous language assessment. Liu and colleagues reported the development and standardization of the Diagnostic Receptive and Expressive Assessment of Mandarin (DREAM) (Liu et al., 2015). DREAM was normed on 969 normal-hearing children between the ages of 2; 5 to 7; 11 years in multiple sites located in Northern and Southern regions of Mainland China (Liu et al., 2015).

In 2011 and 2012, the newly developed Mandarin auditory awareness assessments and Mandarin speech perception assessments were used to evaluate early prelingual auditory development (EPLAD) and early speech perception outcomes over the first year of use of hearing aids or cochlear implants (Zheng et al., 2011, 2012). Another group of researchers reported speech perception skills for CI children in the northern part of China who were not exposed to southern dialects, but only Mandarin (Chen et al., 2015). Their results showed similar pre-lingual auditory skills measured by Mandarin speech perception assessments, regardless of exposure to a different dialect before implantation. Better Mandarin speech perception outcomes were documented for children with early CIs. Other factors, such as having undergone a hearing aid trial (HAT) before implantation, maternal educational level (MEL), and having undergone universal newborn hearing screening (UNHS) before implantation had indirect effects on speech perception outcomes via their effects on age at implantation (Chen et al., 2015). Speech perception assessments used in a 7-year-longitudinal study for children with CIs revealed age of implantation and lexical neighborhood difficulty level influenced speech perception outcomes (Liu et al., 2015). Using the speech perception assessments, Liu et al. (2014) demonstrated that approximately half of the children with ANSD who received cochlear implants showed improved open-set speech recognition. Li and colleagues documented better vocabulary growth rate in Mandarin for children implanted before 3 years old when using the simplified short form (SSF) of the Mandarin Communicative Development Inventory (Li et al., 2014a,b).

Mandarin is a tonal language. Early research has reported tone perception deficits for children with CIs who speak tonal languages (e.g., Wei et al., 2000; Ciocca et al., 2002; Lee et al., 2002; Wong and Wong, 2004). Tone recognition tests for Mandarin were described by Zhou and Xu (2008) and Yuen et al. (2009) for the study of tone development in children with CIs. Tone identification benefits were reported for both hearing aid and CI users who speak Mandarin (Li et al., 2014a,b), though poorer tone perception in implanted children compared to normal hearing peers was found even 3 years after the implantation (Chen et al., 2014). Other groups of researchers outside Mainland China also investigated language skills (Lin et al., 2011; Wu et al., 2011), reading (Wu et al., 2015a,b), written language skills (Wu et al., 2015a,b), behavior and attention outcome (Chao et al., 2015) in Mandarin speaking CI children in regions other than Mainland China.

This review of the most recent literature suggests that auditory awareness, speech perception, language and speech development, neurocognitive development, and social skill assessments are crucial parts of the comprehensive framework of outcome measures for children with hearing impairments. With the availability of Mandarin auditory awareness tests, speech perception tests, standardized vocabulary tests and standardized comprehensive language assessments normed in Mainland China, researchers in Mainland China will be able to design studies that investigate comprehensive outcomes and provide accurate evidence to guide best intervention and rehabilitation clinical practices for Chinese children with hearing impairments.

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Further reading

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