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A systematic review of cross-linguistic and multilingual speech and language outcomes for children with hearing loss

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The purpose of this study was to systematically review the factors affecting the language, speech intelligibility, speech production, and lexical tone development of children with hearing loss who use spoken languages other than English. Relevant studies of children with hearing loss published between 2000 and 2011 were reviewed with reference to (1) methodologies used, (2) children's outcomes, (3) factors affecting children's outcomes, and (4) publication quality. The review included 117 studies describing 20 languages. Monolingual children were described in 109, and multilingual children were described in 8. Better performance outcomes were frequently associated with earlier age of hearing loss diagnosis, intervention, amplification, and less severe hearing loss – a finding similar to studies of English-speaking children. Studies frequently did not report or include information about participant characteristics, blinding of researchers, and reliability. Cross-linguistic comparison of children's outcomes across studies was not possible due to differences in the outcomes assessed, assessment and analysis methods, and participant characteristics. There is a need for cross-linguistic comparisons of the speech and language outcomes of children with hearing loss, but there is little scope for this using existing published research. Few studies described the outcomes of multilingual children with hearing loss.

Keywords: cross-linguistic; monolingual; multilingual; hearing loss; language skills; sign language

Introduction

Hearing loss is a generic term that describes a deficit in the perception of sound, including the detection, localization, lateralization, and discrimination of sound and speech that may impact on the ability to perceive, understand, and produce spoken language (Northern and Downs 2002; World Health Organization 2007). In 2005, the World Health Organization estimated that 278 million people globally have a hearing loss of 41 dB or greater in their better ear (World Health Organization 2010). The incidence of significant, congenital, bilateral hearing loss is reported to be between 2 and 4 per 1000 live births in developed countries, but at least 6 in 1000 live births in regions such as Africa and the Middle East (e.g. Al Khabori and Khandekar 2004; Attias et al. 2006; Ching et al. 2006; Minja and Machemba 1996; Swanepoel et al. 2009).

People with hearing loss use a wide variety of languages, although the majority of research into the outcomes of children with hearing loss focuses on English-speaking

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children and monolingual children (e.g. CAHE Review Team 2009; Sparreboom et al. 2010; U.S. Preventive Services Task Force 2008). As a population, children with hearing loss are highly heterogeneous not only in terms of their audiological characteristics, but also in their linguistic, cultural, social, cognitive, and developmental profiles (Leigh 2008). The factors that have been identified as influencing children's outcomes vary in both their significance and direction of effect across studies. For example, age of hearing loss identification has been found to effect (e.g. Kennedy et al. 2006) and to not effect (e.g. Nittrouer 2008) children's later language outcomes. Other factors that are commonly examined to explain variance in the speech and language outcomes of English-speaking children with hearing loss include degree of hearing loss, age of amplification fitting, type of amplification, amplification device experience, communication mode, presence of additional needs, cognitive ability, and maternal education (e.g. Beer et al. 2012; Ching et al. 2010; Sininger et al. 2010). Speech intelligibility, the extent to which others can understand children's speech, and speech production, the quality and accuracy of the speech produced by the children, have been investigated in many studies of English-speaking children with hearing loss (e.g. Ertmer 2011). Likewise, the development of expressive and receptive language has been frequently investigated (Kumar 2008). The development of children's ability to perceive and produce meaningful variations in fundamental frequency, or lexical tone, has also been investigated for children with hearing loss who speak tonal languages (Xu et al. 2011). Recent systematic reviews have focused specifically on the impact of universal newborn hearing screening as the key factor influencing outcomes (e.g. U.S. Preventive Services Task Force 2008).

Multilingualism adds further complexity to the investigation of the speech and language outcomes for children with hearing loss. Studies of multilingual people with typical hearing vary in their conceptualizations and definitions of multilingualism. In a review of definitions of multilingualism conducted by Grech and McLeod (2012), six parameters were identified: the number of languages, age and time of acquisition, proficiency, domains, output mode (e.g. oral, written, manual), and match with community language(s). To consider multilingualism in its broadest sense, a person who is multilingual is defined as someone who 'is able to comprehend or produce two or more languages in oral, manual or written form regardless of the level of proficiency or use and the age the languages were learned' (Grech and McLeod 2012, 121). Conversely, a person who is monolingual comprehends and produces only one language.

Hearing loss and multilingualism are both global phenomena (Romaine 2013; World Health Organization 2010). The number of people with hearing loss who are multilingual may be increasing due to globalization and technological and communicative advances. Some reasons for the increased multilingualism that are common to both people with typical hearing and people with hearing loss are increased access to media and internet technology, global literacy, population mobility, and lifelong access to education (Grech and McLeod 2012). Improvements in amplification options (e.g. cochlear implants) and accessibility have greatly enhanced the access to spoken languages for people with hearing loss (Geers 2004). The flow-on effects of these developments include higher levels of linguistic competence, educational attainment, employment, and international travel, all of which may impact in the need for multilingualism (Nardi 2008; Punch et al. 2007; Spencer et al. 2004). The issues faced by children with hearing loss who use more than one spoken language have been documented as early as the 1920s, and there is

ongoing recognition that these children form a complex group that little is known about (Fischgrund 1982; Lerman 1984; Mahon 2009). However, it has also been recognized that with improvements in amplification and habilitation opportunities, multilingualism is increasingly a possibility for young children with hearing loss (e.g. Crowe et al. 2012; McConkey Robbins et al. 2004). The communication of people with hearing loss may be described in terms of the communicate modes that they use, i.e. speech and audition (oral), sign (manual), or both (mixed). Multilingualism in the context of people with hearing loss may also encompass the use of sign language(s), typically involving a sign language and the community spoken language (Grosjean 2010a).

Cross-linguistic investigation of the outcomes of monolingual children with hearing loss may also be possible when the outcomes of children using a diverse range of languages are considered. Cross-linguistic study is a means to 'reveal both developmental universals and language-specific developmental patterns in the interaction of form and content' (Slobin 1985, 5). The study of language acquisition utilizing the diversity of linguistic principles that naturally occur in the world's languages allows the opportunity to better understand both language itself, and the process of language acquisition (Goldin-Meadow et al. 2009). Children with hearing loss may experience challenges with typical spoken language acquisition, due to the auditory deprivation in their early linguistic experiences. Thus, the study of speech and language development in children with hearing loss presents a different context for cross-linguistic learning about speech, language, and hearing. In addition to this, an understanding of the speech and language development of children with hearing loss using a wide variety of languages is increasingly important for clinicians and educators as the linguistic diversity of children requiring habilitation grows (Crowe et al. 2012).

The aim of this paper is to provide a systematic review of the literature examining the spoken language, speech intelligibility, speech production, and lexical tone outcomes of monolingual children with hearing loss who use spoken languages other than English, and children with hearing loss who use more than one spoken language. The factors identified by each study as impacting on these children's speech and language outcomes will be examined and the potential for cross-linguistic comparisons identified. Similarities and differences across studies will be documented in terms of the methodologies employed, outcomes, and influences on outcomes. The methodological and reporting quality of each study will also be evaluated.

Methods

Search strategy

The following search terms were used: *children* and *hearing loss* or *deaf* or *hearing disorder* or *hearing impair** for studies published between January 2000 and December 2011. Searches of Cochrane, Medline, Cumulative Index to Nursing and Allied Health Literature (CINAHL), and Education Resources Information Centre (ERIC) databases were conducted.

Inclusion and exclusion criteria

Studies were included if (1) they were published between 2000 and 2011; (2) they addressed spoken language, speech intelligibility, speech production, or lexical tone

in children 18 years of age or younger with permanent hearing loss; (3) they assessed participants' outcomes in a spoken language that was not English; (4) the full text of the study was available in English. Monolingualism and multilingualism involving sign languages were outside the scope of this review, with the focus solely on spoken language development. Studies that reported on children who used sign as part of their communication system were not excluded if the study reported on spoken language, speech, or lexical tone development. Some studies that did not explicitly state the language(s) used by participants or the language of assessments. In these cases this information was obtained by identifying the language in which the assessment was published.

Inter-rater reliability

A speech-language pathologist with expertise in working with children with hearing loss independently reviewed 11% of studies for the following parameters: participant information (age at assessment, language investigated, audiological devices), study information (outcomes investigated, key findings), study design, statement of aims, addressing of sample bias, description of participation rates, description of random selection of participants, use of valid assessment tools, description of researcher blinding, and data reliability procedures. Reliability was established at 89.7% for items describing study content and 93.2% for items describing study quality. Disparities were discussed until consensus was reached. Records were amended accordingly.

Results

A total of 21,098 studies were identified through database searches (Table 1) by the first author, a speech-language pathologist. The first author reviewed the abstracts and excluded studies that clearly did not meet the inclusion criteria or were duplicates, leaving 410 studies. The full texts of these studies were examined and 117 met the inclusion criteria (see supplementary table online for description of each of these studies). The outcome most frequently addressed was language, followed by

Table 1. Number of studies identified through database searches.

Search term combination	Cochrane	Medline	CINAHL	ERIC
Hearing loss + children	112	3143	5575	208
Hearing loss + children + bilingual	1	4	26	3
Hearing loss + children + multilingual	1	0	4	0
Deaf + children	7	1125	4440	792
Deaf + children + bilingual	0	48	408	58
Deaf + children + multilingual	0	0	4	1
Hearing impair + children	0	1284	3803	503
Hearing impair + children + bilingual	0	13	28	10
Hearing impair + children + multilingual	0	0	4	1
Hearing disorder + children	2	18	86	0
Hearing disorder + children + bilingual	0	0	6	0
Hearing disorder + children + multilingual	0	0	2	0

Note: Cochrane = Cochrane library, Medline = Medical literature analysis and retrieval system, CINAHL = Cumulative Index to Nursing and Allied Health Literature, ERIC = Education Resources Information Centre.

speech intelligibility, speech production, and lexical tone (Figure 1). Twenty studies described outcomes in more than one category. Excluded studies totalled 293 and were excluded for the following reasons: 198 did not include relevant outcomes, 51 described sign languages, 9 did not present data, 1 described children with otitis media, and 1 described college-aged participants (Figure 1). In addition, 33 were published in languages other than English: Farsi (8), German (8), Chinese (4), Polish (3), Japanese (2), Turkish (2), Afrikaans (1), Danish (1), Hebrew (1), Italian (1), Portuguese (1), and Spanish (1).

Information describing the languages spoken by participants was available in 107 studies (Table 2). The most frequent languages were Cantonese, Mandarin, Dutch, and Hebrew. Ten studies did not describe the language used by participants, but described participant location: Belgium, Denmark, Iran, Israel, Japan, Korea, Scandinavia, the Slovak Republic, and Thailand. One study described participants who were monolingual users of three languages: English, Farsi, and Turkish. Eight studies described children who were 'multilingual' or lived in 'multilingual environments' where one or more of the following languages were used: Albanian, Arabic, Armenian, Berber, Cantonese, English, French, German, Gujarati, Hebrew, Italian, Kurdish, Marathi, Polish, Portuguese, Russian, Serbo-Croatian, Spanish, Turkish, and Yiddish.

Children using cochlear implants were more frequently reported on than children using hearing aids (Table 3). The amplification device used by participants was not specified in five studies. The outcomes most frequently reported for children using cochlear implants were language, followed by speech intelligibility, speech production, and lexical tone (Table 3). The most frequently investigated outcome for children using hearing aids was also language, followed by speech production, speech intelligibility, and lexical tone (Table 3).

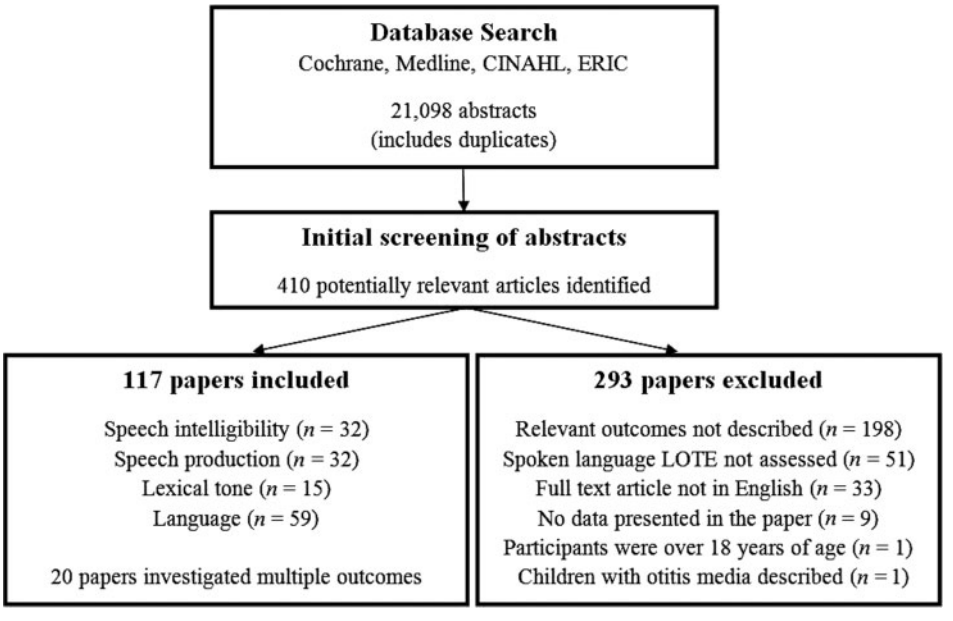

Figure 1. Flowchart of search process.

Table 2. Summary of languages other than English and outcomes investigated.

Language	Reviewed studies ($n=117$)	%	Language ($n=59$)	Speech intelligibility ($n=32$)	Speech production ($n=32$)	Lexical tone ($n=15$)
Cantonese	13	11.0	2	0	2	9
Croatian	2	1.7	0	0	2	0
Danish	1	0.8	1	1	1	0
Dutch	12	10.2	5	6	4	0
Farsi	2	1.7	0	1	0	0
Finnish	4	3.4	2	2	1	0
French	8	6.8	6	4	2	0
German	9	7.6	6	0	3	0
Hebrew	10	8.5	5	1	4	0
Italian	4	3.4	3	1	0	0
Japanese	1	0.8	1	0	0	0
Korean	1	0.8	0	1	0	0
Mandarin	13	11.0	3	5	4	6
Norwegian	2	1.7	1	1	0	0
Serbian	1	0.8	1	0	0	0
Slovene	2	1.7	0	0	2	0
Spanish	3	2.5	2	1	1	0
Swedish	7	5.9	7	0	1	0
Swiss-German	1	0.8	0	0	1	0
Turkish	4	3.4	1	1	1	0
Multilingual	8	6.7	6	1	1	0
Not specified	10	8.5	6	6	2	0

Note: Total exceeds 117. One study investigated multiple languages and 20 studies investigated multiple outcomes.

Quality of studies

Analysis of study design revealed the majority of studies employed non-randomized control trial designs (49). Typically, children with hearing loss were compared to children with normal hearing. One study used a randomized control trial design. Twenty-nine studies used a cross-sectional design and 26 used a cohort design (22 prospective, 5 retrospective). Eleven studies were case reports: single cases (2), case series (4), and case-controls (5).

The majority of studies stated their aims clearly (116); however, participation rates were described in few studies (15) and sample bias was rarely addressed (3). Random selection of participants was not reported in any study and random allocation of participants was described in one study. In all studies the data collection

Table 3. Summary of devices and outcomes investigated.

Device	Language ($n=71$)	Speech intelligibility ($n=39$)	Speech production ($n=39$)	Lexical tone ($n=16$)
Cochlear implant ($n=94$)	41	31	27	14
Hearing aid ($n=41$)	27	7	11	2
Not specified ($n=5$)	3	1	1	0

Note: Totals exceed 117. Twenty-three studies investigated children using cochlear implants and hearing aids and 20 studies investigated multiple outcomes.

tools and methods, and data analysis methods were acceptable for the purpose for which they were employed. Blinding of researchers to children's characteristics was rarely reported (4). The reliability of data collection, transcription, and/or analysis was reported in 40 studies.

Outcomes of monolingual children using languages other than English

Language

Discussion of language here is confined to the structure and content of spoken language communication, specifically semantics (e.g. vocabulary), morphology (e.g. word endings), and syntax (e.g. word order). Development of spoken language is now a realistic goal for the majority of children with hearing loss (Moog and Stein 2008). Spoken language outcomes in monolingual children were investigated in 15 languages across 46 studies: Cantonese, Danish, Dutch, Finnish, French, German, Hebrew, Italian, Japanese, Mandarin, Norwegian, Serbian, Spanish, Swedish, and Turkish (Table 2). The language investigated was not specified in six studies, one each from Denmark, Israel, Japan, Korea, Scandinavia, and Thailand.

Methodological factors. Children's language performance was measured in many different ways including through analysis of language samples, checklists, standardized assessments, and custom-designed tools. Language samples were elicited from children during play (e.g. Ouellet et al. 2001) and using prepared pictures and questions (e.g. Most 2003), as well as through wordless books (Tur-Kaspa and Dromi 2001). Language samples were often analyzed for parameters such as mean length of utterance and lexical diversity with the assistance of computer technology (e.g. Ouellet et al. 2001). Checklists of vocabulary, language skills, and ratings of language ability were conducted across languages, with the MacArthur-Bates Communicative Development Inventory (CDI) frequently used (e.g. Korver et al. 2010). Many studies reported using published and standardized language assessments (e.g. Artieres et al. 2009). Use of language-specific versions of the Peabody Picture Vocabulary Test was reported for Cantonese, French, Italian, Mandarin, Spanish, and Swedish.

Vocabulary outcomes. The development of children's ability to understand and use words (i.e. vocabulary) was investigated in a number of studies. Vocabulary development for children with hearing loss was within the expected range for children with typical hearing in some studies (e.g. Duchesne et al. 2009), but not others (e.g. Ostojic et al. 2011; Rinaldi and Caselli 2009). Longitudinal measures of vocabulary showed improvement over time (Ouellet et al., 2001). A study of German children's vocabulary acquisition found that those with milder hearing losses improved more quickly over time than peers with more severe hearing losses (Kiese-Himmel and Reeh 2006). Japanese-speaking children with hearing loss had more difficulty with abstract vocabulary than concrete vocabulary (Kunisue et al. 2007). Working memory (Hansson et al. 2004) and larger vocabularies (Sahlén and Hansson 2006) were related to better novel word learning for Swedish-speaking children with hearing loss. Studies also investigated the effect of interventions on vocabulary outcomes (e.g. Fung et al. 2005).

Morphosyntactic outcomes. A broad range of skills related to the formation and order of words, morphosyntax, were investigated (e.g. pronoun production, comprehension of syntax, grammatical errors). Children with hearing loss performed within the range expected for typically hearing children in French (Duchesne et al. 2009) and Mandarin (Wu et al. 2011), but also had lower mean length of utterance in French (Ouellet et al. 2001), poorer grammatical development in Italian (Rinaldi and Caselli 2009), and more grammatical errors in Hebrew (Tur-Kaspa and Dromi 2001) and German (Szagun 2004). The general language abilities of children with hearing loss were also found to be poorer than hearing peers in studies of Arabic (Most 2006) and Swedish (Borg et al. 2002). Over half of the adolescents with mild or moderate hearing loss in Delage and Tuller's (2007) study were identified as having a language disorder in French and Keilmann et al. (2011) found that the receptive German language skills of children with hearing loss in their study were poorer for children with specific language impairment. Children also showed difficulty with morphosyntactic transformations in Hebrew (Friedmann and Szterman 2006, 2011).

Factors affecting language outcomes. Relationships between different aspects of age and language outcomes were investigated in many studies. Earlier age of hearing loss diagnosis was associated with better language outcomes in some areas, such as Mandarin vocabulary size and receptive language (Lin et al. 2011) and Swedish narrative skills (Reuterskiold et al. 2010), but not others such as Dutch vocabulary and morphosyntax (Korver et al. 2010) and Mandarin expressive language (Lin et al. 2011). Earlier age of hearing aid fitting positively affected syntactic skills in Hebrew (Friedmann and Szterman 2006, 2011). Earlier age of implantation was related to better language outcomes in a number of studies (e.g. Wang et al. 2007), but did not ensure language outcomes on par with hearing peers (e.g. Duchesne et al. 2009). Greater duration of cochlear implant use was associated with better language outcomes in some studies (e.g. Wie 2010). Age of intervention was positively associated with language outcomes in some studies (Bubbico et al. 2007), but not others (Lertsukprasert et al. 2010).

Severity of hearing loss was related to language outcomes in a number of studies that investigated language outcomes. Children with more severe hearing losses performed more poorly than children with less severe losses in some studies (e.g. Borg et al. 2002). Exclusive use of oral communication mode was associated with better spoken language outcomes in some studies (e.g. Rinaldi and Caselli 2009), but had no impact in others (e.g. Jiménez et al. 2009). Sex (Borg et al. 2002) and etiology of hearing loss (Yoshida et al. 2009) did not affect language outcomes. All studies of children with additional needs reported improved communication and language following cochlear implantation (e.g. Berrettini et al. 2008).

Speech intelligibility

Speech intelligibility is a measure of the degree to which the speech produced by an individual can be understood. Developing intelligible speech by children with significant hearing loss is a principle goal of habilitation (Baudonck, Dhooge, and Van Lierde 2010). Speech intelligibility in monolingual children was investigated in 12 languages across 24 studies: Danish, Dutch, Farsi/Persian, Finnish, French, Hebrew, Italian, Korean, Mandarin, Norwegian, Spanish, and Turkish (Table 2).

The language investigated was not specified in six studies, two from Iran and one each from Belgium, Korea, Scandinavia, and the Slovak Republic.

Methodological factors. Measurement of speech intelligibility varied along the following dimensions: the method of assessment, the length of utterance evaluated, the evaluator of speech intelligibility, the number of evaluators, and timing of assessments. Many studies did not specify these parameters. The majority of studies assessed speech intelligibility using a rating scale specifying how intelligible the children were perceived to be (Daneshi et al. 2011). Ratings from speech samples were based on single words and sentences (e.g. Baudonck et al. 2011), stories (e.g. Baudonck, Dhooge, and Van Lierde 2010), and spontaneous communication (e.g. Wang et al. 2007). Ratings were made from listening to audio (e.g. Van Lierde et al. 2005) and video recordings (e.g. Huttunen 2008) of the children speaking, as well as live speech (e.g. Phillips et al. 2009). Ratings were completed by caregivers (e.g. Huang et al. 2005), speech-language pathologists (e.g. Bakhshaee et al. 2007), audiologists (e.g. Scherf et al. 2009), and unfamiliar listeners (e.g. Girgin 2008). Ratings for each child may have been completed by one (e.g. Jiménez et al. 2009) or more individuals (e.g. Girgin 2008). Speech intelligibility was also determined through agreement of listeners' transcriptions of children's productions compared to targets (e.g. Huttunen 2008).

Speech intelligibility outcomes. The development of intelligible speech was reported in the majority of studies, particularly following cochlear implantation (e.g. Bakhshaee et al. 2007). Large variations of the rate at which intelligible speech developed were also reported (De Raeve 2010). Smaller and slower changes in speech intelligibility were reported for children with additional needs (Dammeyer 2008). Phillips et al. (2009) investigated the development of speech intelligibility in native speakers of English, Turkish, and Farsi following cochlear implantation in a cross-linguistic paradigm. Overall, children's speech intelligibility increased over time and no differences were found between speakers of different languages.

Factors affecting speech intelligibility. The majority of studies reported increased speech intelligibility over time. Better speech intelligibility was related to age of diagnosed (e.g. Philips et al. 2009), pre-operative hearing thresholds (e.g. Artieres et al. 2009), age of implantation (e.g. De Raeve 2010), duration of implant use (e.g. Huang et al. 2005), exclusive use of oral communication (e.g. Jiménez et al. 2009), and placement in a mainstream education setting (e.g. Most 2007). In direct comparisons, children with cochlear implants had better speech intelligibility than children using hearing aids (e.g. Van Lierde et al. 2005). Hearing loss etiology did not affect speech intelligibility (e.g. Daneshi et al. 2011); however, children with additional needs showed slow improvement in speech intelligibility (e.g. Dammeyer 2008).

Speech production

Speech production describes an individuals motor-planning, phonological representations, and articulation. Huttunen (2001, 79) described the acquisition of phonology as being of the 'utmost importance for a child when he/she is learning to decode and convey meanings using spoken language.' Speech production in monolingual

children was investigated in 14 languages across 29 studies: Cantonese, Croatian, Danish, Dutch, Finnish, French, German, Hebrew, Mandarin, Slovene, Spanish, Swedish, Swiss-German, and Turkish (Table 2). The language investigated was not specified in two studies one each from Denmark and the Slovak Republic.

Methodological factors. Children's speech was elicited and measured in a number of different ways. Pictures were commonly used to elicit spontaneous or imitated speech samples (e.g. Hansson et al. 2007) and objects were sometimes used to elicit speech from young children (e.g. Peng, Tomblin, et al. 2004). Stimuli were drawn from published word lists (e.g. Baudonck et al. 2011), published assessments (e.g. Law and So 2006), and study-specific word lists (e.g. Boes 2004). Spontaneous speech was elicited in some studies (e.g. Moore et al. 2006). Speech samples were analyzed using phonological process analyses (e.g. Baudonck et al. 2010), rating scales (e.g. Profant et al. 2008), and objective instrumentation (e.g. Liker et al. 2007). Transcription and analysis reliability were reported in some studies.

Consonant production outcomes. Consonants are speech sounds that are produced by creating an obstruction to the airflow of the vocal tract. Many studies compared the consonant acquisition and errors of children with hearing loss to hearing peers. Investigation of different parameters across different languages showed children with hearing loss performed better than (e.g. Moore et al. 2006), similarly to (e.g. Adi-Bensaid and Ben-David 2010), or worse (e.g. Peng, Weiss, et al. 2004) than typically hearing peers. Canonical babbling in German showed accelerated development following cochlear implantation (Schramm et al. 2009). Patterns of consonant acquisition following cochlear implantation differed in French (Bouchard et al. 2007) and German (Seifert et al. 2002). Initial consonant acquisition was poorer for children using cochlear implants than hearing peers in Mandarin (Peng, Weiss, et al. 2004). The acquisition of consonant clusters (two or more consecutive consonants within the same segment of the syllable) in Hebrew was similar for children with typical hearing and children with cochlear implants (Adi-Bensaid and Ben-David 2010). Productions of 's' and 'sh' were found to overlap (Liker et al. 2007) and affricate duration was longer (Mildner and Liker 2008) for Croatian children with hearing loss compared to those with typical hearing. Dutch-speaking children with bilateral cochlear implants had more accurate consonant articulation than children with unilateral cochlear implants or hearing aids (Baudonck et al. 2011). An investigation in Swedish found that children with mild-moderate hearing loss had better consonant accuracy than children with specific language impairment (Hansson et al. 2007).

A case study examined cross-lingual speech production, comparing a monolingual Spanish-speaking child with a cochlear implant with four English-speaking peers also using cochlear implants (Moore et al. 2006). The Spanish-speaking child significantly outperformed the English-speakers at the post-implant assessment. The authors suggested this may have been due to Spanish syllables being more consistently consonant-vowel in structure compared to English syllables, a structure more aligned with babbling.

Vowel production outcomes. Vowels are speech sounds produced through shaping of the vocal tract without obstructing the airflow. Vowel production was investigated in Croatian (Liker et al. 2007), German (Neumeyer et al. 2010), and Slovene (Ozbic and

Kogovsek 2010). All three studies found differences in the vowel articulation (vowel space) of children with hearing loss compared to children with typical hearing. Barry, Blamey, and Fletcher (2006) also reported children's vowel inventories in Cantonese steadily increased following cochlear implantation, although variations existed in individual rates of acquisition.

Factors affecting speech production outcomes. A variety of factors contributed to explaining difference in speech production outcomes. Younger age of hearing loss identification (e.g. Profant et al. 2008) and implantation (e.g. Lin and Peng 2003) were associated with better speech production. Younger age of implant was not always associated with improvements in speech production (e.g. Bouchard et al. 2007) and definitions of early versus late implantation varied across studies. Degree of hearing loss (Huttunen 2001) and type of amplification (Law and So 2006) explained differences in outcomes in some studies. Use of an oral communication mode (Bouchard et al. 2007) and being female (Percy-Smith et al. 2010) were also associated with better outcomes.

Lexical tone

In over half the languages of the world it is possible to alter a word's meaning by altering the fundamental frequency (or pitch) at which syllables are spoken (Crystal 1997). For example, Cantonese has six contrastive lexical tones and three stopped tones (So 2007), whereas Mandarin has four contrastive lexical tones (Hua 2007). Lexical tone in monolingual speakers of languages other than English was investigated in two languages across 15 studies: Cantonese and Mandarin (Table 2).

Methodological factors. Children's mastery of lexical tone was measured using a number of different methods. Studies employed meaningful stimuli (e.g. Ciocca et al. 2002), nonsense stimuli (e.g. Barry, Blamey, and Martin 2002), or both (e.g. Wong and Wong 2004). Picture identification and/or naming (e.g. Peng, Tomblin, et al. 2004), and discrimination tasks (e.g. Wong and Wong 2004) were used. Young children were assessed using an adapted play audiometry paradigm (e.g. Barry et al. 2002). Scoring of children's performance was generally based on whether the children's responses matched the target item (e.g. Han et al. 2007); however, Huang et al. (2005) required naive listeners to transcribe children's speech and the number of tones listeners transcribed correctly was analyzed.

Tone perception outcomes. Tone perception is the ability of an individual to perceive and identify variations in pitch that are linguistically significant. Studies investigating lexical tone perception were identified for Cantonese, but not Mandarin. Cochlear implant users were able to discriminate tones satisfactorily, but less accurately than children with typical hearing (e.g. Lee et al. 2002). The most salient tone features were average pitch height and pitch direction (Barry, Blamey, and Martin 2002); however, differences in error patterns (Lee et al. 2002) and discrimination accuracy (Wong and Wong 2004) across tones were reported.

Tone production outcomes. Tone production is the ability of an individual to produce variations in pitch that are linguistically significant. In Mandarin, children with typical hearing had better tone production than children using cochlear implants

(e.g. Huang et al. 2005), although large individual differences in performance were recorded in the cochlear implant group (e.g. Han et al. 2007). Tone production in Mandarin-speaking cochlear implant users was described as flat by Xu et al. (2004) and production of tones 1 (high level) and 4 (falling) were better than for tones 2 (rising) and 3 (low dipping). In the Xu et al. (2004) study, the pattern of tone acquisition was the same as for typically hearing children. Lee, van Hasselt, and Tong (2008) found that in Cantonese, the accuracy of tone production for children using cochlear implants increased over time.

Factors influencing performance. Difficulties in lexical tone for children using cochlear implants has been related to the paucity of pitch information which is able to be transmitted by the cochlear implant (Xu et al. 2004). The cochlear implant coding strategy was not found to affect children's performance (e.g. Han et al. 2009). Earlier implantation and longer duration of implant use were associated with better tone perception and production in some studies (e.g. Han et al. 2007). Hearing thresholds of less than 90dB were associated with better tone perception (Lee et al. 2008); however, differences between cochlear implant and hearing aid users were not always found (e.g. Lee et al. 2010). Correlations were also found between tone perception and tone production skills (e.g. Xu et al. 2011).

Outcomes of multilingual children

Eight studies addressing the outcomes of children from multilingual environments were identified during the literature search (Table 2). Two investigated children who used hearing aids, four investigated children who used cochlear implants, and two investigated children with either device. Children were from environments where two or more of the following languages were used: Albanian, Arabic, Armenian, Berber, Cantonese, English, French, German, Gujarati, Hebrew, Italian, Kurdish, Marathi, Polish, Portuguese, Russian, Serbo-Croatian, Spanish, Turkish, and Yiddish.

Language development

Six studies investigated the language development of children who had experienced language acquisition in multilingual environments. Two studies described children's outcomes in the dominant community language only (i.e. the most frequently used language in that geographic location): English and German. Four studies described children's outcomes in the dominant community language (English or German) as well as the language used in the children's home environment (i.e. their home language). Home languages investigated were: Albanian, Arabic, Armenian, Berber, Cantonese, English, French, German, Gujarati, Hebrew, Italian, Kurdish, Marathi, Polish, Portuguese, Russian, Serbo-Croatian, Spanish, Turkish, and Yiddish.

Outcomes in dominant community languages. Overall, children with hearing loss from bilingual homes could develop language skills in their dominant community language. Development of dominant community language skills within the range expected for monolingual children was demonstrated in three studies (McConkey Robbins et al. 2004; Thomas et al. 2008; Waltzman et al. 2003). Contrary to these findings, two studies reported that children from bilingual homes performed worse than their monolingual peers in two German studies (Kiese-Himmel 2008;

Teschendorf et al. 2011). Difficulty with development of dominant community language skills was reported in a case study by Murphy and Dodd (2010), who described the English language profile of a bilingual (English-Vietnamese) child with hearing loss who also showed characteristics of specific language impairment.

Outcomes in home languages. Children developed skills in both their home language and the dominant community language in all studies. The Student Oral Language Observation Matrix (SOLOM; Montebello Unified School District Instructional Division 1978) was used to measure home language skills in all four studies (McConkey Robbins et al. 2004; Teschendorf et al. 2011; Thomas et al. 2008; Waltzman et al. 2003); however, the SOLOM is not norm referenced so it was not possible to objectively compare the children's home language development to that of their monolingual peers. Thomas et al. (2008) completed the SOLOM and the MacArthur CDI in both of the children's languages. These children showed greater development in the community language than their home language. Both McConkey Robbins et al. (2004) and Waltzman et al. (2003) reported that over half of their participants were educated in bilingual settings, potentially making their experiences of bilingualism different from Teschendorf et al. (2011) whose participants were all educated in the dominant community language, and Thomas et al. (2008) whose participants primarily received education and intervention in the dominant community language.

Speech intelligibility

Speech intelligibility was investigated by Lejeune and Demanez (2006) for French-speaking children in Belgium. Participants' caregivers reported use of French, Arabic, Turkish, or a sign language at home. Children's speech intelligibility was rated annually, showing improvement over the five years of the investigation. After five years, children with cochlear implants had significantly more intelligible speech than children using hearing aids.

Speech production

Guiberson (2005) presented a case study of the speech development of a young Spanish-English bilingual girl with a bilateral, severe-to-profound, post-meningitic hearing loss who used a cochlear implant. Her Spanish speech production was not assessed prior to implantation due to her limited vocabulary, and was not assessed post-implant. Her English speech production was assessed post-implant (chronological age: 101 months; hearing age: 39 months) with her speech determined to be equivalent to that of a 49-month-old hearing child.

Methodological factors

Assessing the language development of multilingual children with hearing loss is complex. First, standardized language assessments and norm referenced language measures are not available in many languages. Second, even when these assessments did exist, a competent speaker of the target language may not be available to administer the assessment (Thomas et al. 2008). Third, it is difficult to find assessments that measure the same parameters in the same way across languages,

especially for studies where participant age range was large or a number of different languages were used. Assessment of home language skills in the studies examined was through report rather than assessment. SOLOM scoring was completed by the examiner who assessed dominant community language skills in two studies (McConkey Robbins et al. 2004; Waltzman et al. 2003) and by children's caregivers in two studies (Teschendorf et al. 2011; Thomas et al. 2008). In addition to this, caregivers in the Thomas et al. (2008) study completed the SOLOM and the MacArthur CDI in both of the child's languages, allowing direct comparison of children's skills in each of their languages.

Discussion

Factors affecting the outcomes of children with hearing loss were identified across 20 languages other than English, and the methodological construct and reporting of these studies were examined. Cross-linguistic comparison of children's outcomes from the published literature identified is problematic and few studies were identified that reported on the outcomes of multilingual children with hearing loss.

Factors that explained variance in studies across all four outcomes examined were age of diagnosis, age of intervention and amplification, and degree of hearing loss. These factors, and the variability of their impact across studies, have also been identified in studies examining the outcomes of monolingual English-speaking children with hearing loss (e.g. CAHE Review Team 2009). Heterogeneity in outcomes is a common finding across studies of children with hearing loss, even with homogenous participant groups (Pisoni et al. 2008). Studies identified many participant characteristics that affected children's performance in the specific outcomes examined, in the specific group of participants examined. Examples include children's education setting, type of audiological amplification, communication mode (use of sign and/or speech), sex, and social well-being. However, the factors identified as influencing children's outcomes were often contradictory between studies, possibly due to differences in the specific outcomes measured and the characteristics of participants in each study. For example, children's use of sign as part of their communication system was associated with better (Rinaldi and Caselli 2009), comparable (Jiménez et al. 2009), and worse (Percy-Smith et al. 2010) language outcomes than use of speech alone.

Overall, the quality of reporting of demographic information was weak. Crucial information about participant characteristics and assessments was often difficult to find, ambiguous, or absent. The language used by participants or the language the assessment was conducted in was not stated in 26.5% of the studies reviewed. Furthermore, the age of participants at the time of assessment was not stated in 40.1% of the studies reviewed. It was also rarely specified whether children were monolingual. Similarly, many studies did not specify participants' audiological characteristics such as type of amplification or hearing thresholds. Participant age was also frequently not clearly reported, which was particularly problematic for studies reporting longitudinal data. Poor collection and reporting of participant characteristics and research methods impacts: accurate replication of research finding, appropriate generalization of research findings, and comparison of factors and outcomes across studies (Hammer 2011). The importance of this knowledge should not be underestimated as the cause of differences in outcomes across languages may be related to participant characteristics, assessment methods, or

possibly to the nature of languages being assessed. Accurate cross-linguistic comparisons of children's outcomes depend on differences in participant characteristics and assessment methods being eliminated as causes of differences in outcomes. It is not possible to do this from the studies identified in this systematic review.

Eight studies compared the performance of children from multilingual environments to that of monolingual peers using the dominant community language. These eight studies represent preliminary work into the outcomes of multilingual children with hearing loss, and acknowledge the need for further research in this area. The factors to consider when examining the outcomes of multilingual children are even more heterogeneous than for their monolingual peers (Grosjean 2010b). Multilingual language acquisition can be affected by the number of languages that children use, the age at which each language is acquired, whether acquisition is simultaneous or sequential, children's relative proficiency in each language, use of speech and writing in each language, the purpose for which each language is used, and the languages used by the people the speaker interacts with (Grech and McLeod 2012). These factors are important to keep in mind when considering multilingual acquisition for children with hearing loss as they can experience disruption in the normal process of language acquisition and their caregivers may be advised to limit children's exposure to the home language (Waltzman et al. 2003).

Limitations

While the search terms and databases employed to identify articles for this systematic review were broad, it is possible that some relevant studies were not identified. A major limitation of this review was the inability of the authors to include studies that were not published in English. While 'English is the *lingua franca* of science' (Meneghini and Packer 2007, 112), there are undoubtedly publications written in other languages describing the speech and language development of children with hearing loss. Thirty-three studies published in languages other than English, but with an English abstract, were identified in the present literature search. It was beyond the resources of the authors to examine the content of these studies so they were unable to be included in this review. It is likely that there are further studies with abstracts written in languages other than English that would have met the criteria for this review had translations been available.

Clinical implications and future research

The identification of cross-linguistic trends in the outcomes of children with hearing loss and the factors influencing outcomes will give insight into similarities and differences across languages and groups of language users. There is a need for more cross-linguistic comparisons of children's speech and language development of children with hearing loss that is currently unmet in the literature. Studies of different languages with comparable methodologies will provide greater understanding of the relationship between linguistic factors and the outcomes of children with hearing loss.

Demand is growing for clinicians and educators to be able to assess children's speech and language development in their home language (Guiberson and Atkins 2012; Williams and McLeod 2012). There is currently little research describing the speech and language outcomes of children with hearing loss who speak the majority

of the world's languages. Accessing speech and language acquisition data regarding monolingual children can be difficult to even for typically developing children in many languages (Cheng 2007), and speech and language assessment tools are unavailable in many languages (McLeod 2012) or not equivalent across languages. Demand for high-quality research in these topic areas is likely to increase as clinicians and educators are required to follow principles of evidence-based practice and to provide evidence-based recommendations to the families of young children from diverse language backgrounds.

Romaine (2013, 445) stated 'Bilingualism and multilingualism are normal, unremarkable necessities of everyday life for the majority of the world's population.' From this basis, multilingual children with hearing loss are under-represented in the literature. There has long been a call for quality research into the speech and language outcomes of multilingualism in children with hearing loss (Fischgrund 1982). This demand is likely to grow as studies demonstrate that children with hearing loss come from multilingual home environments (Crowe et al. 2012; Mahon et al. 2011) and that oral multilingualism is a possibility for children with hearing loss (Waltzman et al. 2003).

Conclusion

Heterogeneous outcomes and factors associated with outcomes were identified in this systematic review. Factors affecting the language, speech intelligibility, speech production, and lexical tone development of children with hearing loss were identified across 20 languages other than English. The most reported factors were age of identification, amplification and intervention, and degree of hearing loss. Cross-linguistic comparison of children's outcomes and factors influencing outcomes using currently published literature is not possible. This is principally due to methodological differences, poor reporting of participant characteristics and differences in assessment methods across studies.

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