



# The communicative benefits of cochlear implantation for children with hearing loss and autism spectrum disorder: A review

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## Abstract

### Introduction

It has been suggested that autism spectrum disorder (ASD) occurs more frequently in children with hearing loss than in the general population. However, little is known about effective intervention for children who have this dual diagnosis. This review of the literature examined the communicative benefits of audiological intervention (in the form of cochlear implantation) for these children.

### Materials and methods

A literature search of 15 databases was conducted, which identified 46 peer-reviewed journal articles that described communication skills in children with a dual diagnosis of hearing loss and ASD. From this set, three articles that reported both pre- and post-cochlear implantation data were selected for review. A search of references cited in these and other papers reporting outcomes for children with hearing loss and additional disabilities resulted in the addition of four more articles to the review set.

### Results

Speech perception and/or language outcomes were available for 14 individual children and two small groups (comprising 4 and 8 participants respectively). Improvements from pre- to post-implant were observed for both groups, but significant for only one. Improvements were also reported for 11 of the 14 individual children, with greater improvements seen in children using some spoken language at the post-implant assessment compared to those using gesture only.

Parent feedback regarding the benefits of cochlear implants was generally positive.

### Conclusion

Children with a dual diagnosis of severe-to-profound hearing loss and ASD appeared to benefit from audiological intervention with cochlear implants, although individual child outcomes were variable and studies did not include “no intervention” control groups. Future research would be strengthened through the use of more effective assessment procedures, and the inclusion of further information on children’s cognitive ability and the nature and severity of their ASD.

### Introduction

Recent reports from the United States, United Kingdom, and Europe indicated that approximately 1% of children in the general population have some form of autism spectrum disorder (ASD)<sup>1,2,3,4</sup>, although rates appear to be increasing<sup>5</sup>. It has been suggested that within the population of children with hearing loss, the incidence of autism is higher than in the general population<sup>6,7</sup>.

The 2009-2010 Annual Survey of Deaf and Hard of Hearing Children and Youth in the United States indicated a rate of 1.7%<sup>8</sup>, as did a study of 475 children who received a cochlear implant at the University of Michigan between 1998 and 2003<sup>9</sup>. Similarly, a population-based study of 451 Australian children with hearing loss found that 2.7% of participants were diagnosed with ASD by 3 years of age<sup>10</sup>.

Diagnosing ASD in children with hearing loss is complicated because both disorders are associated with difficulties in language, communication, and social

interaction<sup>11,12</sup>. As a result, ASD is typically diagnosed later in children with hearing loss than it is in children with typical hearing<sup>12</sup>. In addition, for children who have a dual diagnosis, ASD is generally diagnosed later than hearing loss<sup>13</sup>, which may delay the provision of appropriate intervention services.

For children who have a dual diagnosis of hearing loss and ASD, the pressing clinical question is how to intervene most effectively to ensure optimal outcomes. The paucity of research in this area has been acknowledged in the literature, with regard to educational placement and teacher intervention<sup>7,11,14,15,16,17</sup>, as well as audiological intervention<sup>9,18,19</sup>.

The aim of this review was to synthesise and evaluate published research that has examined the benefits of cochlear implantation for children with hearing loss and ASD, with a view to identifying correlates of successful implantation and avenues for future research. The benefits of hearing aids were not examined due to the lack of relevant published studies.

Consistent with recommendations in the literature, benefits were defined broadly to include objective measures of speech perception and language skills, as well as more subjective measures based on observation and parent report<sup>20,21</sup>. Potential correlates of success were identified from the hearing loss literature<sup>13,22</sup>. They included age at implant, duration of implant use, and cognitive ability.

### The Current Review

Two research questions were addressed.

1. Do children with hearing loss and ASD benefit from cochlear implants? If so, which aspects of performance

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(speech perception, language, communication, or behaviour) are positively affected? Do observed benefits vary as a function of level of cognitive ability or audiological characteristics?

2. What additional research needs to be conducted in order to provide parents with well-informed advice regarding the potential benefits, or otherwise, of cochlear implantation for their children.

### Materials and methods

A literature search was conducted of 15 databases using the terms "Child\*" AND "Hearing" OR "Deaf\*" AND "Autism" OR "ASD" in the title and abstract fields for publications between 1998 and 2013 (inclusive). A total of 2,284 citations was identified and removal of duplicates yielded 571 unique references. The titles and abstracts of citations were examined to determine whether they described the communication skills of children with a dual diagnosis of hearing loss and ASD.

Forty-six journal articles met this criterion and their abstracts were read to ascertain whether the articles contained data relevant to the review questions. Articles were included if they reported both pre- and post-implant data. Three articles were identified and included in the review set. References cited in these and other papers reporting outcomes for children with hearing loss and additional disabilities were then checked, and a further four articles were added to the review set.

### Results

Table 1 presents a summary of the results obtained in seven published studies that reported pre- and post-cochlear implantation assessment results for children with severe-to-profound hearing loss and ASD. Other disabilities were also present in some but not all participants, a variable that was not addressed systematically in any of the studies. Results are described in terms of auditory/speech

perception, language, communication, and other outcomes. Because many participants were unable to complete formal testing<sup>9,21</sup>, missing data complicate interpretation of the findings. The studies were conducted in a variety of languages including English<sup>9,19,21,25</sup>, German<sup>23</sup>, Italian<sup>20</sup>, and Persian<sup>24</sup>.

In five studies, results for individual participants could be identified<sup>9,19,20,21,22</sup>, whereas two studies reported only group data<sup>24,25</sup>. For the 14 participants whose individual data were identifiable, average age at implant was 4;5 (years;months) with a range from 2 to 9 years. Average duration of implant use at post-CI assessment was 2.75 years (range 0.5 to 7).

### Auditory/speech perception and language outcomes

All studies contained information regarding children's speech perception and/or language skills. In the group of 14 children with individual results, only three (21.4%) showed no improvement in speech perception or language skills from pre- to post-implant assessments. These children, participants HA-10, WA-5, and DO-6, all received their implants at a relatively young age compared to the cohort as a whole (between 2;0 and 3;10). In addition, participant HA-10 was diagnosed with severe psychomotor retardation, which probably contributed to her poor results (see Table 1).

The remaining 11 participants (78.6%) all showed some improvement from pre- to post-implantation in speech perception (n = 4), language (n = 1), or both (n = 6). In the five cases where improvement was seen only in speech perception or language, participants' skills in the other area were not assessed.

The remaining two studies presented findings for small groups of participants. Daneshi and Hassanzadeh reported that overall, four children showed improvement on the Persian Auditory Perception Test,

although the result was not quite significant ( $p < .068$ )<sup>24</sup>. Cruz, Vicaria, Wang, Niparko, and Quittner presented data from eight participants, who showed significant improvement from pre- to post-implantation on the expressive and receptive components of the Reynell Developmental Language Scales (see Table 1)<sup>25</sup>.

### Communication outcomes

Communication outcomes were not available in the two studies that reported only group results<sup>24,25</sup>, and could not be interpreted unambiguously for participants WA-5 and WA-11 due to lack of detail<sup>21</sup>. Of the remaining 12 participants with identifiable individual assessment results, pre-implant communication outcomes were not available for nine of them<sup>9,23</sup>.

To examine post-implant communicative strategies children were classified according to whether they used gesture only, with no sign or spoken language (n = 4 participants: HA-10, DO-6, BE-14, BE-15), a combination of sign, gesture and/or word approximations (n = 5 participants: DO-1, DO-3, DO-4, DO-5, DO-7), or a combination of spoken words with sign and/or gesture (n = 3 participants: HA-3, DO-2, RO-1).

There was an association between communication mode used post-implant and extent of improvement in speech perception and/or language outcomes from pre- to post-implant: Children who used gesture only tended to show the least improvement whereas those who used some spoken words showed the greatest improvement.

Although no obvious pattern emerged with respect to age at cochlear implantation, the three children who communicated using some spoken language had an average of 5 years' experience with their devices (range 3 to 7), whereas the four children who communicated using only gesture had an average of just 2.25 years' experience (range 2 to 3; see Table 1).

**Table 1: Summary of papers reporting speech perception, language, or communication outcomes for children with ASD and hearing loss pre- and post-cochlear implantation (in chronological order).**

Participants with ASD <sup>a</sup>	Implant age (use) <sup>b</sup>	Diagnosis	Cognitive Ability	Auditory/ Speech Perception <sup>c</sup>	Language <sup>c</sup>	Communication <sup>c</sup>	Other changes
Hamzavi et al. (2000; <i>N</i> = 10 Austrian children with hearing loss and additional disabilities) <sup>23</sup>							
HA-3	4;1 (3)	Autism	Mild intellectual disability	+EARS battery (German version)	+Vocabulary (30 words at 3 yrs post-implant) <sup>d</sup>	Responded and produced words meaningfully post-implant.	Reduced severity of autism and anxiety, and improved motor behaviour by 3 mths post-implant.
HA-10	3;10 (2)	Autism	Severe psychomotor retardation	No benefits	No benefits	Minimal speech development post-implant.	Improved phonation and happiness with noise at 1 year post-implant; and a reduction in stereotyped behaviours.
Waltzman et al. (2000; <i>N</i> = 29 American children with profound hearing loss and additional disabilities) <sup>21</sup>							
WA-5	2;0 (≥ 1)	PDD	No data	No benefits	No benefits	Total communication	WA-5 could not complete any assessment tasks pre- or post-implant.
WA-11	2;11 (5)	Autism	No data	+ESP, +GASP-W, +MLNT after 5 yrs.	No data	Sign	WA-11 could not complete NU-CHIPS, GASP-S, PBK, LNT, MLNT, or common phrases sentence test at 5 yrs post-implant.
Donaldson et al. (2004; <i>N</i> = 7 American children with hearing loss and ASD) <sup>9</sup>							
DO-1	9 (1)	PDD	No data	+MAIS, +GASP-W	+CDI, +EVT	Gesture, word approx.	Communication benefited most from implant.
DO-2	4 (5)	PDD (mild)	No data	+GASP-W, +GASP-S	+EVT, +PPVT-III	Spoken words, gesture	Social interaction benefited most from implant.
DO-3	7 (1)	Autism	No data	+MAIS	+EVT	Sign, gesture	Awareness of environment benefited most from implant.
DO-4	3 (2)	Autism	No data	No data	+CDI	Sign, gesture, word approx.	Awareness of environment benefited most from implant.
DO-5	8 (3)	Autism	No data	+MAIS	+CDI	Sign, gesture, word approx.	Communication benefited most from implant.
DO-6	3 (2)	Autism	No data	No benefits	No benefits	Gesture	Increased vocalisation, enjoyment of music.
DO-7	3 (0.5)	Autism	No data	+MAIS	+CDI	Word approx., other	Overall family interaction benefited most from implant.
Daneshi and Hassanzadeh (2007; <i>N</i> = 60 Iranian children with profound hearing loss and additional disabilities, drawn from a population of 398) <sup>24</sup>							
<i>n</i> = 4	5;8 (1) (Mean age for pop <sup>n</sup> )	Autism	No data <sup>e</sup>	+Persian Auditory Perception Test (Improvement not significant <i>p</i> < .068)	No data	No data	Participant sub-groups with other additional disabilities improved significantly from pre- to post-cochlear implant, but three children with congenital blindness did not.

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**Table 1 (continued)**

Berrettini et al. (2008; <i>N</i> = 23 Italian children with profound hearing loss and additional disabilities) <sup>20</sup>							
BE-14	6;0 (3)	ASD	No data	+Geers and Moog category (from 0 to 3) <sup>f</sup>	No data	Gesture (no sign language) = no change pre- to post-cochlear implant	BE-14's parents reported less benefit from cochlear implant than 85.7% of parents whose children had additional disabilities other than ASD.
BE-15	2;6 (2)	ASD	No data	+Geers and Moog category (from 0 to 1) <sup>f</sup>	No data	Gesture (no sign language) = no change pre- to post-cochlear implant	BE-15's parents reported less benefit from cochlear implant than all other parents in the study.
Cruz et al. (2012; <i>N</i> = 31 American children with severe-to-profound hearing loss and additional disabilities) <sup>25</sup>							
<i>n</i> = 8	2;6 (3)  (Mean age for group of 31)	PDD / Autism	No data	No data	+RDLS-exp, +RDLS-rec, (both <i>ps</i> ≤ .01)	No data	Children with PDD/autism achieved lowest language scores pre-implant, and developed more slowly than children without additional disabilities. CBCL externalising behaviours increased in children with PDD/autism post-implant ( <i>p</i> < .01).
Robertson (2013; <i>N</i> = 10 Irish children with severe-to-profound hearing loss and ASD, but pre- and post-implant data presented for 1 child only) <sup>19</sup>							
RO-1	3;2 (7)	ASD	Mild ID	+CAP (No awareness pre-CI changed to understands common phrases post-CI)	No data	Spoken words and signs, +SIR (No spoken language pre-implant changed to "unintelligible" speech post-implant, but familiar listeners could understand him.)	The family's attitude to the cochlear implant was very positive.
CAP = Categories of Auditory Performance; CBCL = Child Behavior Checklist; CDI = MacArthur Communicative Development Inventory; EARS Battery = The Evaluation of Auditory Responses to Speech test battery; ESP = Central Institute for the Deaf Early Speech Perception Test; EVT = Expressive Vocabulary Test; GASP-W and GASP-S = Glendonald Auditory Screening Procedure for words and sentences respectively; LNT = Lexical Neighborhood test; MAIS = Meaningful Auditory Integration Scale; MLNT = Multisyllabic Lexical Neighborhood Test; NU-CHIPS = Northwestern University Children's Perception of Speech test; PBK = Phonetically Balanced Kindergarten word test; PDD = Pervasive Developmental Disorder; PPVT-III = Peabody Picture Vocabulary Test (3rd Edition); RDLS = Reynell Developmental Language Scales; SIR = Speech Intelligibility Rating. a Data from individual participants are reported when available. A participant's ID combines the first two letters of the first author's surname with their subject number from the source article. b Implant age (use) = age at implant in years and months, and implant experience in years. c "+" indicates improved performance from pre- to post-cochlear implantation on the specified assessment. d Expressive or receptive not specified. e No children with severe intellectual disability were included in the population. f Relevant speech perception categories based on research by Geers and Moog <sup>26</sup> : 3 = consistent word identification; 2 = some word identification; 1 = some pattern of speech perception; 0 = no pattern of speech perception.							

### Other outcomes

Table 1 also provides a summary of other relevant information regarding the perceived benefits of cochlear implantation for participants in three studies<sup>9,20,23</sup>.

Two points are worth noting. First, a wide range of benefits was identified, including reduced anxiety, happiness with noise, enjoyment of music, and improved communication,

environmental awareness and social interaction. Second, although not reported in table 1, five out of seven parents in Donaldson et al.'s study indicated that they would recommend an implant to other families<sup>9</sup>.

### Discussion

The authors have referenced some of their own studies in this review. These referenced studies have been

conducted in accordance with the Declaration of Helsinki (1964) and the protocols of these studies have been approved by the relevant ethics committees related to the institution in which they were performed. All human subjects, in these referenced studies, gave informed consent to participate in these studies.

This review of the literature addressed two research questions. First, do children with hearing loss and ASD

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benefit from cochlear implants? Findings from seven studies reporting pre- and post-implant assessment results suggest that they do, although not to the same extent as children with hearing loss and no additional disability<sup>25</sup> or children with hearing loss and other types of additional disabilities, such as mild or moderate intellectual delay, cerebral palsy, attention deficit hyperactivity disorder, and learning disability<sup>24</sup>.

In regard to the aspects of performance most affected by cochlear implantation, six of the studies reported outcomes for speech perception, three reported outcomes for language, and five reported on communication mode. Although the results reveal a general pattern of improvement in all three aspects following implantation, the observed improvements cannot be attributed to the use of cochlear implants per se, because none of the studies included a “no intervention” or “hearing aid” control group. Hence, it is possible that these children could have shown the same levels of improvement without an implant. In addition, as has been noted frequently in the literature, children varied widely in their response to cochlear implantation, with some showing marked improvements and others very little change<sup>9,18,19</sup>.

In regard to traditional correlates of cochlear implant success, age at implantation did not appear to relate directly to children’s outcomes; and although there was a tendency for children who used some spoken language to have greater experience with their devices than children who used gesture only, participant numbers are too small to conduct statistical analysis or draw firm conclusions. Finally, the association between outcomes and cognitive ability could not be determined because most studies did not provide a separate measure of children’s cognitive functioning, perhaps because of difficulties in assessment of children with a dual diagnosis<sup>6</sup>.

A further aim in conducting this review was to identify additional research that needs to be undertaken in order to provide parents with well-informed advice regarding the potential benefits, or otherwise, of cochlear implantation for their children with hearing loss and ASD.

Despite recent interest in the outcomes achieved by children with this dual diagnosis, many aspects require further investigation.

Future research would benefit from a direct comparison of children with and without cochlear implants, separate assessment of children’s cognitive ability, and the inclusion of assessment procedures likely to minimise the amount of missing data; for example, the Preschool Language Scale<sup>27</sup>, which has recently been used successfully with children who have hearing loss and ASD<sup>28</sup>. Further detail on the nature and severity of ASD in individual children is also essential, and should be facilitated through development of the Comprehensive and Brief International Classification of Functioning, Disability and Health Core Sets for ASD<sup>29</sup>.

### Conclusion

The findings obtained in seven studies reporting pre- and post-cochlear implant assessment results for children with a dual diagnosis of severe-to-profound hearing loss and ASD suggest that these children benefited from implantation, although outcomes were variable and, as noted by previous researchers, only a small subgroup used spoken communication post-implant<sup>13</sup>.

Future research should use assessment procedures that have been shown to be effective for children with a dual diagnosis in order to address the question of why some children achieve better outcomes than others, taking into account aspects such as the nature and severity of ASD, and levels of cognitive ability.

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