

Language development in the pediatric cochlear implant patient

Robert J. Ruben, MD 

Objective: To access the long-term outcomes of children implanted during most sensitive period for language development.

Study design: Literature review.

Method: An initial PubMed search was carried out using the search terms language development and cochlear implant resulted in 1149 citations. A second search was carried out on the initial citations using the criterion of implantation in the period of birth to 24 months, which identified 386 articles. These were analyzed to determine those studies in which linguistic outcome was measured at least three or more years following implantation.

Results: Twenty-one reports published from 2004 to 2017 that met the criteria. The range of follow-up was from 3 years to an excess of 10 years. Four => 10-year follow-up reports were consistent in showing that the earlier the subject is implanted the better the outcome. Many, but not all, of these children did obtain age-appropriate language. There were 17 reports with follow-up from 3 to less than 10 years. In 7 of the 11 studies, the children's expressive language was reported to have reached an age level of less than 80%. The results for receptive language showed that 4 of the 11 studies found that the children achieved a receptive language age level of less than 80%. There were 8 studies which documented the effect of implantation before 12 months of age and between 12 and 24 months of age and they all found that the earlier the implantation, the better the outcome for language.

Conclusion: The cochlear implant is efficacious in the amelioration of receptive and expressive language deficits in most congenitally deafened children implanted before the age of one. The language outcomes for those implanted after the age of one decline as the age of implantation increases.

Key Words: cochlear implant, language outcome.

Level of Evidence: N/A.

INTRODUCTION

Cochlear implantation in children was first reported in 1983.¹⁻³ These initial reports were utilized to obtain approval by the Federal Drug Administration in 1989 for children older than 2 years and in 2000 for children 12 months old or older.⁴ This initial approval was based upon reported improvement in speech detection and discrimination but not in language. In 1992 at the first European Symposium on Paediatric Cochlear Implantation, held at the University of Nottingham, UK⁵ (Fig. 1), a paper entitled "The pediatric cochlear implant" was presented that emphasized that language should be the outcome measure for pediatric cochlear implantation in those patients whose language had not yet developed at the time of implantation, that is those under one year of age.⁶ Four years later, there was a follow-up conference

entitled The International Conference on Language Development in Cochlear Implanted Children, Lyon, France, 1996⁷ (Fig. 2). There were 67 podium presentations and 6 posters covering a broad spectrum of issues of language development including psychophysical measures, neural functional imaging, and event-related potentials.

Language as an outcome measure for assessing a medical/surgical intervention was a new concept⁸ which was rapidly assimilated into the care of the prelingually deafened child. Heretofore, the study of language lay in the analysis of the characteristics of receptive and expressive language. The structure of language had come to be defined in terms of prosody, syntax, and semantics. The measures of the development of prosody, syntax, and semantics were now brought to bear to measure the efficacy of the cochlear implant for the development of language. It has now been more than one-third of a century since cochlear implantation has been utilized to promote the development of language in the prelingually deaf child. Several long-term outcome studies⁹⁻¹² have looked at not only the effects of the cochlear implant on language development but also its effects on broader issues of the quality of life, including educational, economic, and social attainment.

The objective of this study is the assessment of the long-term outcomes of children who were implanted during what is considered the most sensitive period for language development, the age of =<24 months, a determination based on several outcome reports that found that the language outcomes were poorer for children

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made.

From the Departments of Otorhinolaryngology-Head and Neck Surgery and Pediatrics, Albert Einstein College of Medicine, Montefiore Medical Center, Bronx, New York, U.S.A.

Editor's Note: This Manuscript was accepted for publication 12 March 2018.

Send correspondence to Robert J. Ruben, MD, Departments of Otorhinolaryngology-Head and Neck Surgery and Pediatrics, Albert Einstein College of Medicine, Montefiore Medical Center, Greene Medical Arts Pavilion, 3400 Bainbridge Avenue, 3rd Floor, Bronx, NY 10467. Email: robert.ruben@einstein.yu.edu.

DOI: 10.1002/liv.2.156



PAEDIATRIC COCHLEAR IMPLANTATION



Medical School
Queen's Medical Centre
Nottingham, UK
24-27 September 1992

PROGRAMME

Fig. 1. Programme—The first European Symposium on Paediatric Cochlear Implantation, September 22–27, 1992.

implanted at >24 months than for those implanted before their second birthday.^{13–18} These empirical observations are congruent with other sensitive period studies.^{19–23} For the present study, follow-up of three or more years post implant was determined as the minimum time for evaluation of the outcome measure, based on findings of the fewest number of years after implant in an infant = <24 months of age to achieve approximate age appropriate language.^{24,25}

METHODS

A PubMed search was carried out using the search terms *language development AND cochlear implant* as the initial search criterion. This resulted in 1149 citations. A second search was carried out on the initial 1149 citations using the additional criterion of implantation in the period of birth to 24 months, which identified 386 articles. These 386 articles were then analyzed to determine those studies in which, in addition to implantation occurring before 24 months of age, linguistic outcome was measured at least three or more years following implantation, and that excluded other morbidities, eg, CMV, Down Syndrome, etc. Twenty-one reports met these final criteria.^{9,10,12–18,24–36}

RESULTS

There have been 21 reports published from 2004 to 2017 that met the criteria. They represent a heterogeneous group of articles from 12 different countries. The range of follow-up was from 3 years to an excess of 10 years. There was no consistency in the types of instruments that were used to evaluate various aspects of language. For the present study, each language report was characterized as representing either expressive or receptive language. The reports covered cochlear implantation in infants as early as 1989.⁹ The type and/or sequence of implantation was not analyzed in the present study because there was the use of many different forms of implants, which included

pg 12264

**INTERNATIONAL CONFERENCE
ON LANGUAGE DEVELOPMENT
IN COCHLEAR IMPLANTED CHILDREN**

Hôtel SOFITEL
20, quai Gailleton - 69002 LYON

PROGRAM

LYON - FRANCE
December 8-9, 1996

Official languages : English/French

Chairman : Alain MORGON

Scientific Secretariat :

Patricia CHUTE, Lionel COLLET, Gérald O'DONOGHUE,
Eric TRUY, Suzan WALTZMAN

General Secretary :

Professeur A.H. MORGON, Hôpital Edouard Herriot,
Pavillon U,
Place d'Arsonval, 69003 LYON - FRANCE
Phone. (33) 04.72.11.05.35 Fax (33) 04.72.11.05.34

SATURDAY DECEMBER 7 : COCKTAIL RECEPTION AS OF 5-7 PM

Fig. 2. Program—International Conference on Language Development in Cochlear Implanted Children, December 8–9, 1996.

TABLE I.
Outcome 71 Subjects at 10+ Years Post-Implant—4 Studies.

Study	# subjects	Expressive	Receptive	Comment
Ramos ³⁴ Spain	25			The subjects implanted at =<24 months performed significantly better on all tests than children who were implanted at an older age. Results not compared to normal hearing age matched controls.
Pisoni ³⁰ United States	3			These data show that the processing of auditory information is less than seen in normal children at more than 10 years of follow-up.
Colletti ¹⁰	40	At 10 years all the children implanted before 12 months of age were at the normal level. Those implanted later, between 13 and 35 months, did not reach age level	At 10 years all the children implanted before 12 months of age were at the normal level. Those implanted later, between 13 and 35 months, did not reach age level	
Uziel ⁹	3			All are oral and in regular schools

devices from single-channel, to multichannel, to bilateral simultaneous, to sequential bilateral implantation.

The => 10-Year Follow-Up Reports

The four => 10-year follow-up reports encompassed a total of 71 subjects (25, 3, 40, and 3 subjects, respectively) who were implanted at =<24 months of age.^{9,10,30,34} These subjects do not represent all that were implanted at =<24 months as there were those who were lost to follow-up for a variety of reasons. The outcomes for these 71 subjects are shown in Table I.

These four reports of 71 subjects followed for at least 10 years post-implant are consistent in showing that the earlier the subject is implanted the better the outcome. Many, but not all, of these children did obtain age-appropriate language.

In the Uziel⁹ report, of the 82 children implanted at different ages, three were implanted before the age of 24 months and followed for at least 10 years. Of the 82 children, there was a complete assessment for all but one child who moved to a different country. The value of these data is considerable in reflecting a complete clinical experience, a real-world result, for it assessed the outcome of an entire population of implanted deaf children, with implantation occurring at different ages, with differing technologies, and included comorbidities. Uziel et al. found that for expressive language, 54 of 82 children (66%) developed connected speech intelligible to the average listener and that, again for expressive language, 62 (75%) of 82 children scored below the median value of their hearing peers of equivalent age. Study of the subjects' receptive language found that words identified correctly on the Phonetically Balanced Kindergarten (PBK) Test word test was 72% (SD, +/-26%; range, 0 to 100%; median, 82%).

The initial emphasis for the children assessed by the Uziel study was placed on mainstreaming and on oral/aural mode of communication: 74% of the children were placed in a mainstream program, 73% were using oral communication as the mode of communication, only 21% were enrolled in a school for the deaf, and 5% were

in part-time at a school for the deaf. At the time of the final evaluation, 79% were using oral communication, 20% total communication, and 1% sign; 78% were placed in mainstream, and 22% were attending a school for the deaf.

At the time of the Uziel study, 32 were at junior high school level, 14 at high school level, and 3 children were attending elementary schools. Six additional children were enrolled in a special unit for children with disability where they received general and professional education. Seventeen were in further noncompulsory education, studying a range of subjects at vocational levels. Six subjects attended universities. Three were already engaged in a professional activity.

The 3 to =<10-Year Follow-Up Reports

There were 17 reports^{12-18,24,26-29,31-33,35,36} with follow-up from three to less than 10 years, encompassing a total of 904 subjects that were implanted at =<24 months of age. Table II presents the results on the studies for which the follow-up was less than 10 years.

In 7 of the 11 studies, the children's expressive language was reported to have only reached an age level of less than 80%. The results for receptive language were somewhat better in that only four of the 11 studies found that the children achieved a receptive language age level of less than 80%. Only one study, it should be noted, showed both expressive language and receptive language to reach age level.

There were eight studies^{10,14-18,29,35} which documented the effect of implantation before 12 months of age and between 12 and 24 months of age. All the studies were consistent in that the earlier the implantation, the better the outcome for expressive and receptive language. The best outcomes were those in the children implanted before 12 months of age.

DISCUSSION

The analysis of the 21 publications that met the criteria, determined as described above, demonstrates noticeable consistency in the findings. The best success

TABLE II.
Number of Studies Reporting Outcomes from 3 Years to
<10 Years of Follow-Up.

	All at age level	Most* (>80%) at age level	Some† (<80%) at age level	None at age level
Expressive	1 ²⁹	3 ^{13,14,28}	7 ^{15,17,24-26,32,33}	
Receptive	1 ²⁹	6 ^{13,14,17,18,27,28}	4 ^{15,24,25,32}	
Overall		1 ¹⁴	2 ^{15,32}	1 ³³

*Either > 80% or as the author described the finding.

†Either < 80% or as the author described the finding.

in language acquisition occurs when the patient is implanted early, and the data suggest that this is optimally before the age of one. This observation recognizes the importance of the optimal sensitive period for language development.

Implantation at a later age does enable the development of expressive and receptive skills, although seldom to age level. The studies in general indicate that even the children implanted early, although they tend to do better than those implanted later, do not generally reach age level, although it can be noted that in the study of Wie et al,¹⁷ the implanted infants did achieve age level at 48 months. These findings are consistent with the concept that while there is a sensitive period meaningful for optimized language acquisition, the possibility of strengthening the development of language acquisition still remains after that most sensitive period: the central nervous system does not “shut down”, so to speak, but its capacity to adapt to an electrically induced afferent input remains.

The data indicate that for these implanted children, receptive language tests better than expressive language. Three studies, Niparko et al,²⁵ Uziel et al,⁹ and Yoshinaga-Itano et al,²⁴ which are characterized by excellent assessment of all subjects, demonstrated that very few of these children achieve age level in either expressive or receptive competence. It is also noted that in these and other studies, very few of the children exceed the average age level of competence of normal-hearing children. The study of Wie et al¹⁷ clearly demonstrated the advantage of early implantation for receptive and expressive language. The early implanted (5 to 12 months of age) subjects all obtained normal receptive and expressive language scores with variance similar to that of the hearing population at 48 months postimplantation. Those children implanted at 12 to 18 months of age did not achieve age level receptive or expressive language at 48 months postimplantation. Ching's recent article is congruent with the above findings.³⁸

The cochlear implant has created an entirely new area of medicine. For the first time, the structure and function of the central nervous system is being deliberately shaped as a medical intervention by the introduction of electrically induced afferent signals to a central nervous system that had previously not been stimulated or had ceased to be stimulated by sound. The cochlear implant is taking advantage of the capacity of

genetically provided pathways and of the plasticity of the central nervous system to adapt to this synthetic stimulation. The substitution of synthetic stimulation is less than perfect, but is a great step forward in enabling language development in deaf children.

The observation from the long-term studies of implantation during the early sensitive period emphasizes two major areas that need to be further developed. The first is that of the afferent electrical stimulation of this intervention. Over the past three decades, there have been many improvements in the form, number of electrodes, and coding of the electrodes implanted. This work will go forward to find how to optimize the electrical afferent so as to enable the recipient to obtain age-appropriate language abilities.

The second area requiring our attention is the exploration of the properties of the plasticity of the central nervous system, its characteristics and capacities, so as to maximize the response of the central nervous system, whether implantation occurs before or after 12 months of age.

It is noteworthy that deeper scientific understanding of these two areas will, in all likelihood, illuminate problems and issues of other implantations of sensory systems in those who have been deprived of the normal input. It will also likely be of use or other modes of central nervous system interventions.

The optimal care for the deaf child today is dependent on early detection and implantation. Throughout much of the world, newborn infant screening by physiological means has become routine. The next step should be the use of genetic screens to determine the possibility of hearing loss as part of newborn infant screening. The latter should enable early detection of progressive disorders, and will also serve as a control for the physiological testing which has false negatives and false positives.

Medicine functions at three levels care, cure, and prevention. The cochlear implant today functions in many ways at all three levels: it cares for the patient, it does not totally cure patient of hearing loss, and it can enable language development and, to a greater or lesser extent, ameliorate language impairment. For the area of prevention, science turns to the cell biology of the inner ear and the identification of processes which result in hearing loss, genetic and otherwise, and the exploration of interventions to prevent these once they have been defined.

CONCLUSION

The cochlear implant is efficacious in the amelioration of receptive and expressive language deficits in most congenitally deafened children implanted before the age of one. The language outcomes for those implanted after the age of one decline as the age of implantation increases. The challenge today is to advance the technology of the implant, and deepen our understanding of how we can engage with the plasticity of the central nervous system so as to optimize language development in those who are hearing impaired.

BIBLIOGRAPHY

1. Eisenberg LS, Berliner KI, House WF, Edgerton BJ. Status of the adults' and children's cochlear implant programs at the House Ear Institute. *Ann N Y Acad Sci* 1983;405:323–331.
2. Eisenberg LS, Berliner KI, Thielemeier MA, Kirk KI, Tiber N. Cochlear implants in children. *Ear Hear* 1983;4:41–50.
3. House WF, Berliner KI, Eisenberg LS. Experiences with the cochlear implant in preschool children. *Ann Otol Rhinol Laryngol* 1983;92:587–592.
4. Cochlear Implants. 2017. National Institute of Health. Available at: <https://report.nih.gov/NIHfactsheets/ViewFactSheet.aspx?csid=83>. Accessed December 28, 2017.
5. O'Donoghue G, Koyunchu M. First European symposium on paediatric cochlear implantation Nottingham 24–27 September 1992. *J Laryngol Otol* 1992;106:1109–1110.
6. Ruben RJ. Language—the outcome measure for the linguistically developing cochlear implant patient. *Int J Pediatr Otorhinolaryngol* 1995;33:99–101.
7. Ruben RJ. Summary of the International Conference on Language Development in Cochlear Implanted Children, Lyon, France 8–9 December 1996. *Int J Pediatr Otorhinolaryngol* 1999;47(2):213–214.
8. Ruben RJ. Language screening as a factor in the management of the pediatric otolaryngic patient. Effectiveness and efficiency. *Arch Otolaryngol Head Neck Surg* 1991;117:1021–1025.
9. Uziel AS, Sillon M, Vieu A, et al. Ten-year follow-up of a consecutive series of children with multichannel cochlear implants. *Otol Neurotol* 2007;28:615–628.
10. Colletti L, Mandala M, Zoccante L, Shannon RV, Colletti V. Infants versus older children fitted with cochlear implants: performance over 10 years. *Int J Pediatr Otorhinolaryngol* 2011;75:504–509.
11. Sarant JZ, Harris DC, Galvin KL, Bennet LA, Canagasabay M, Busby PA. Social development in children with early cochlear implants: normative comparisons and predictive factors, including bilateral implantation. *Ear Hear* 2017. doi: 10.1097/AUD.0000000000000533. [Epub ahead of print]
12. De Raeve L, Vermeulen A, Snik A. Verbal cognition in deaf children using cochlear implants: effect of unilateral and bilateral stimulation. *Audiol Neurootol* 2015;20:261–266.
13. Kosaner J, Deniz H, Uruk D, Deniz M, Kara E, Amann E. Assessment of early language development in Turkish children with a cochlear implant using the TEDIL test. *Cochlear Implants Int* 2017;18:153–161.
14. Dettman SJ, Dowell RC, Choo D, et al. Long-term communication outcomes for children receiving cochlear implants younger than 12 months: a multicenter study. *Otol Neurotol* 2016;37:e82–e95.
15. van Wieringen A, Wouters J. What can we expect of normally-developing children implanted at a young age with respect to their auditory, linguistic and cognitive skills? *Hear Res* 2015;322:171–179.
16. Colletti L, Mandala M, Colletti V. Cochlear implants in children younger than 6 months. *Otolaryngol Head Neck Surg* 2012;147:139–146.
17. Wie OB. Language development in children after receiving bilateral cochlear implants between 5 and 18 months. *Int J Pediatr Otorhinolaryngol* 2010;74:1258–1266.
18. Tajudeen BA, Waltzman SB, Jethanamest D, Svirsky MA. Speech perception in congenitally deaf children receiving cochlear implants in the first year of life. *Otol Neurotol* 2010;31:1254–1260.
19. Tomblin JB, Barker BA, Hubbs S. Developmental constraints on language development in children with cochlear implants. *Int J Audiol* 2007;46:512–523.
20. Sharma A, Campbell J. A sensitive period for cochlear implantation in deaf children. *J Matern Fetal Neonatal Med* 2011;24(Suppl 1):151–153.
21. Benasich AA, Choudhury NA, Realpe-Bonilla T, Roesler CP. Plasticity in developing brain: active auditory exposure impacts prelinguistic acoustic mapping. *J Neurosci* 2014;34:13349–13363.
22. Friedmann N, Rusou D. Critical period for first language: the crucial role of language input during the first year of life. *Curr Opin Neurobiol* 2015;35:27–34.
23. Ruben RJ, Rapin I. Plasticity of the developing auditory system. *Ann Otol Rhinol Laryngol* 1980;89:303–311.
24. Yoshinaga-Itano C, Baca RL, Sedey AL. Describing the trajectory of language development in the presence of severe-to-profound hearing loss: a closer look at children with cochlear implants versus hearing aids. *Otol Neurotol* 2010;31:1268–1274.
25. Niparko JK, Tobey EA, Thal DJ, et al. Spoken language development in children following cochlear implantation. *JAMA* 2010;303:1498–1506.
26. Svirsky MA, Teoh SW, Neuburger H. Development of language and speech perception in congenitally, profoundly deaf children as a function of age at cochlear implantation. *Audiol Neurootol* 2004;9:224–233.
27. Lee KY, van Hasselt CA. Spoken word recognition in children with cochlear implants: a five-year study on speakers of a tonal language. *Ear Hear* 2005;26:30s–7s.
28. Wu CM, Sun YS, Liu TC. Long-term categorical auditory performance and speech intelligibility in Mandarin-speaking prelingually deaf children with early cochlear implantation in Taiwan. *Clin Otolaryngol* 2008;33:35–38.
29. Colletti L. Long-term follow-up of infants (4–11 months) fitted with cochlear implants. *Acta Otolaryngol* 2009;129:361–366.
30. Pisoni DB, Kronenberger WG, Roman AS, Geers AE. Measures of digit span and verbal rehearsal speed in deaf children after more than 10 years of cochlear implantation. *Ear Hear* 2011;32:60s–74s.
31. VanDam M, Ide-Helvie D, Moeller MP. Point vowel duration in children with hearing aids and cochlear implants at 4 and 5 years of age. *Clin Linguist Phon* 2011;25:689–704.
32. Boons T, Brokx JP, Frijns JH, et al. Effect of pediatric bilateral cochlear implantation on language development. *Arch Pediatr Adolesc Med* 2012;166:28–34.
33. Nittrouer S, Sansom E, Low K, Rice C, Caldwell-Tarr A. Language structures used by kindergartners with cochlear implants: relationship to phonological awareness, lexical knowledge and hearing loss. *Ear Hear* 2014;35:506–518.
34. Ramos-Macias A, Borkoski-Barreiro S, Falcón-González JC, Plasencia DP. Results in cochlear implanted children before 5 years of age. A long term follow up. *Int J Pediatr Otorhinolaryngol* 2014;78:2183–2189.
35. Murri A, Cuda D, Guerzoni L, Fabrizi E. Narrative abilities in early implanted children. *Laryngoscope* 2015;125:1685–1690.
36. Escorihuela Garcia V, Pitarch Ribas MI, Llopez Carratala I, Latorre Monteagudo E, Morant Ventura A, Marco Algarra J. Comparative study between unilateral and bilateral cochlear implantation in children of 1 and 2 years of age. *Acta Otorrinolaringol Esp* 2016;67:148–155.
37. Geers A, Brenner C. Background and educational characteristics of prelingually deaf children implanted by five years of age. *Ear Hear* 2003;24:2s–14s.
38. Ching TYC, Dillon H, Leigh G, Cupples L. Learning from the longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study: summary of 5-year findings and implications. *Int J Audiol* 2017:1–7.