International Journal of Pediatric Otorhinolaryngology 162 (2022) 111213

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Cochlear implantation outcomes in children with global developmental delay

Leila Monshizadeh [a], Seyed Basir Hashemi [a,b,*], Mehdi Rahimi [c], Mostafa mohammadi [b]

[a] Otolaryngology Research Center, Khalili Hospital, Shiraz University of Medical Sciences, Shiraz, Iran
[b] Department of Otorhinolaryngology, Khalili Hospital, Shiraz University of Medical Sciences, Shiraz, Iran
[c] Yazd University, Yazd, Iran

ARTICLE INFO

Keywords:
Cochlear implantation
Global developmental delay
Children
Hearing impairment

ABSTRACT

Objective: As the number of hearing loss cochlear implant candidates who suffer from global developmental delay has dramatically increased, we aimed to study the prognosis of implantation in this group.

Materials and methods: In this cross-sectional case-control study, we utilized the Ages and Stages Questionnaire third edition (ASQ-3) to investigate the prognosis of cochlear implantation and its rehabilitation in 26 congenitally deaf children who suffered from global developmental delay compared with those in 25 non-delayed cases with the same conditions in two time periods, namely the first diagnosis of hearing loss and 18 months after the surgery and rehabilitation program. The data were analyzed using Statistical Package for Social Sciences, version 21 (SPSS-21).

Results: By the time of hearing loss diagnosis (six months old), the performance of all the global developmentally delayed hearing loss children in five subtests of the ASQ-3 scale was significantly lower than that of their non-delayed peers. Meanwhile, they improved significantly in two gross motor and social development subtests 18 months after the surgery and rehabilitation.

Conclusion: Along with the general improvement of delay developed children with sensorineural hearing loss after cochlear implantation, global developmental assessment in the process of candidacy and after implantation is an essential factor that needs to be considered.

1. Introduction

Cochlear implantation is a common standard procedure through which children with profound sensorineural hearing loss can acquire hearing. As a result, they would achieve speech perception and production skills. The strong effect of certain factors, such as the age of implantation and additional disabilities, on a child's prognosis after cochlear implantation, has led the medical teams to exclude several cochlear implant candidates in the early years of implantation with the assumption of uncertain prognosis. For example, over a decade ago, in the UK, numerous candidates with even one moderate additional disability were deprived of implantation [1]. This pattern was seen in Canadian cochlear implant candidates in 2000 as well [1,2].

However, the number of documented children with sensorineural hearing loss who suffered from the global developmental delay resulted in an expansion in cochlear implantation candidacy criteria to include children with complex developmental delay.

Global developmental delay is a condition under which a child is delayed in one or more milestones categorized into motor skills, communication skills, cognitive skills, and social and emotional development [3]. According to various studies, approximately 40% of the children with sensorineural hearing loss experience global developmental delay as well [4,5]. In this situation, the delay can usually be overcome through early intervention, such as cochlear implantation, speech and language therapy, and physiotherapy or occupational therapy [6,7]. The permanent delay in the development, which occurs following genetic disorders (Down syndrome and Fragile X), or severe metabolic disorders might prevent cochlear implantation due to the child's confirmed poor prognosis. Meanwhile, the prevalence of various degrees of mild to moderate delay development in sensorineural hearing loss children has resulted in flexibility in the selection criteria of cochlear implantation. These days, many cochlear implanted children

* Corresponding author. Khalili Hospital, Khalili St. Department of Otorhinolaryngology, Khalili Hospital, Shiraz University of Medical Sciences, Shiraz, Iran.
E-mail addresses: leilamonshizadeh1008@gmail.com (L. Monshizadeh), hashemib@yahoo.com (S.B. Hashemi), mehdirahimi@yazd.ac.ir (M. Rahimi), mostafamohamadi95@gmail.com (M. mohammadi).

https://doi.org/10.1016/j.ijporl.2022.111213
Received 6 November 2021; Received in revised form 7 June 2022; Accepted 11 June 2022
Available online 28 June 2022

include a group who has experienced difficulties in fine and gross motor movement, social interaction, or cognitive abilities [8–10].

Accordingly, the health authorities encourage parents to monitor their child's development to inhibit its adverse effects on the child's performance [6,7]. However, both parents and professionals must adjust their expectations for implantation results and incline to provide global developmentally delayed hearing loss children with a situation in which they access cochlear implantation devices in the early years of diagnosis.

Nevertheless, studies focusing on global developmental delay in cochlear implanted children remain scarce and mostly go back to recent years [11–13]. Therefore, the main aim of this study was to compare the performance of the two groups of cochlear implanted children with/-without global developmental delay at two times; the first diagnosis of hearing loss (six months old) and 18 months after the surgery when in fact, cochlear implantation and its following rehabilitation program were carried out.

2. Materials and Methods

The objective of this cross-sectional case-control study was to evaluate the effect of global developmental delay on pediatric cochlear implantation performance. We selected the sample by matching all the congenitally deaf children with cochlear implants who suffered from global developmental delay (26 patients) to 25 of their non-delayed implanted peers. Six out of 26 developmentally delayed candidates suffered from severe difficulties, including motor and cognitive delays, while the others were approximately equal and had experienced mild to moderate degrees of developmental delay. For note, all the subjects' mean chronological age by hearing loss diagnosis was 6.03 ± 0.89 months old.

We selected the subjects from the children who participated in the rehabilitation program before and after the surgery. An expert speech therapist taught them lip-reading before the surgery. After the surgery, the same therapist provided the accepted traditional intervention program for cochlear implanted children named auditory-verbal therapy (AVT).

All the participants' parents signed a general informed written consent form to permit using the data of their medical records with consideration of their privacy. Additionally, the study protocol was reviewed and approved by the ethics committee of our university.

The inclusion criteria for global developmentally delayed cochlear implanted children included delay in one or more milestones, such as motor skills, cognitive skills, and social and emotional development. These symptoms were approved by developmental pediatrics and neurologist's examinations and reports from their parents while doing daily activities, like washing, dressing the child, or playing with him [6].

By the time of hearing loss diagnosis (about six months old), all the children were assessed

via the Ages and Stages Questionnaire third edition (ASQ-3). It is necessary to indicate that we used a version of (ASQ-3) that is culturally adapted for Persian-speaking children [14]. The second assessment was carried out after about 18 months, while the children were almost 24 months old and had received a cochlear implant device and its follow-up rehabilitation program.

The ASQ-3 is a developmental screening tool comprising five separate sub-scales of communication, gross motor, fine motor, problem-solving, and personal-social interaction, by which the developmental progress can be determined in children between the ages of 1 month–5 ½ years old. Each sub-scale indicates the activities expected to be feasible for the relevant age group. The essential data were gathered by asking the parents to fill in the ASQ-3 questionnaire with "regularly," "sometimes "or" not yet" doing the activity according to their child's abilities.

After data collection, we conducted statistical analysis through Statistical Package for Social Sciences, version 21 (SPSS-21). Evaluation of the group's difference concerning age distribution was performed through the independent sample *t*-test. Since we intended to compare the groups in terms of the five sub-scales of the ASQ scale, the Multivariate Analysis of Variance (MANOVA) was performed. The statistical significance was accepted at $P < 0.05$.

3. Results

According to Table 1, the mean age of diagnosis of hearing loss and age of implantation in the two groups were almost equal. The independent sample *t*-test showed no significant difference between the two groups concerning age distribution.

The subjects' mean scores of the five subtests of the ASQ-3 scale by the time of hearing loss diagnosis and 18 months afterward are demonstrated in the second and third tables, respectively.

Based on the data represented in Table 2, the pre-implantation scores were statistically different for all subtests in the two groups of children, with typically developing peers outperforming those with developmental delay. The post-implantation scores were unchanged for typically developing children and unchanged in three sub-tests for those with developmental delay (Table 3). However, there was an improvement in 2 sub-tests (gross motor and social development) for those with developmental delay.

We did the multivariate analysis of variance to examine the differences between the groups by the time of diagnosis of hearing loss (Table 4) and 18 months later (Table 5).

Table 4 concerns the significant differences between the subjects in terms of their performance in all the five subtests of the ASQ-3 scale by the time of hearing loss diagnosis (six months old). Moreover, the mean scores of the two groups, which are demonstrated in Table 2, showed better performance of the normally developed hearing loss children compared with that of the delay developed hearing loss children.

Nonetheless, after 18 months of cochlear implantation and rehabilitation program (Table 5), the multivariate analysis of variance results showed significant improvement in 2 domains (gross motor and social development) for those with the developmental delay with similar post-implant performance to typically developing peers ($P < 0.001$). This finding could also be confirmed by comparing the mean scores of the two groups after 18 months of implantation and rehabilitation programs (Table 3).

4. Discussion

The present study results indicated that global developmental delay causes the poor performance of hearing loss children in motor skills, speech perception and production, cognitive skills, and social and emotional development. However, they become improved like normally developed cochlear implanted children in gross motor and social development after implantation and its follow-up rehabilitation program. We hypothesized that their growth in terms of social interaction and movement ability is due to the natural evolution and growth of self-esteem following the AVT program and the child's improvement in auditory perception performance, which allow the child to investigate the surrounding environment; this may, in turn, facilitate his/her learning process. This finding revealed the importance of a child's general development assessment in the process of candidacy and after implantation more than ever because significant developmental delay is a negative predictor of the child's performance in different aspects of cognitive and language abilities.

Edwards LC et al.; conducted a study in 2006 on 32 children before and after the implantation surgery and its follow-up rehabilitation. It implied a strong negative correlation between the severity of developmental delay exhibited by cochlear implanted children and their progress in speech intelligibility [11]. This pattern was also observed in the research by Archbold and colleagues (2015) and Eze and colleagues (2013) [15,16].

In other words, those who suffered from a significant delay in various areas of functioning depicted no progress in speech perception even

Table 1
The mean age of hearing loss diagnosis and implantation in the two groups.

Group	N	Age (months)	Mean ± SD	Minimum	Maximum	Significance (P-value)
Children with delayed development and hearing loss	26	Age of hearing loss diagnosis	6 ± 0.97	5	7	0.75
		Age of implantation	12.38 ± 2.11	12	15	
Normal developed hearing loss children	25	Age of hearing loss diagnosis	6 ± 0.81	5	7	0.41
		Age of implantation	12.76 ± 0.92	12	15	

Table 2
Descriptive statistics at the age of six months old.

ASQ subtests	Mean ± SD	
	Delay developed hearing loss children	Normal developed hearing loss children
Communication	20 ± 2	36 ± 5.9
Gross motor	17.69 ± 8	48 ± 2.8
Fine motor	24.4 ± 7.9	46.8 ± 4.8
Problem solving	21.15 ± 4.96	48.8 ± 3.8
Social development	21.34 ± 7.2	48.4 ± 4.26
N	26	25

Table 3
Descriptive statistics at the age of 24 months old.

ASQ subtests	Mean ± SD	
	Delay developed C.I children	Normal developed C.I children
Communication	21.34 ± 5.2	49.6 ± 4.54
Gross motor	49.23 ± 4.23	49.8 ± 4.67
Fine motor	20.19 ± 7.41	49.4 ± 4.16
Problem solving	19.8 ± 5.19	48.6 ± 3.39
Social development	47.69 ± 5.69	48.2 ± 4.3
N	26	25

though their cochlear implantation device was switched on and a rehabilitation program was in progress. According to Lindcey C. Edwards and colleagues, speech perception and intelligibility were related to the child's developmental status; This means the delayed developed cochlear implanted children's access to environmental sounds did not significantly improve their speech perception abilities compared with that of the normally developed cochlear implanted children. Moreover, it did not significantly ameliorate their quality of life [11]. Nevertheless, speech perception performance of mild global developmentally delayed children was almost equal to that of their non-delayed implanted peers. This finding is in line with the present study results, but we assessed the patients through the schedule of growing skills.

Another study in 2004 indicated that severe developmental delay, especially that observed in pervasive developmental disorders, is a condition in which small gains in speech perception and production skills might be accessed following implantation. Nevertheless, some progress in behaviors and interaction with others through implantation and its following rehabilitation program encouraged most parents to recommend cochlear implantation to other families under similar conditions [17].

Overall, the results obtained by different papers have implied that mild to moderate developmentally delayed cochlear implanted children can benefit from surgery. However, their progress might be lower than their normally developed peers [5,18,19]. Thus, it could be argued that general developmental status of young cochlear implant candidates is a predictor of their post-implantation performance, which should be considered for candidacy decision.

Despite the remarkable results obtained in this study, there are several study limitations that need to be taken into consideration; primarily the small sample size, particularly in the case group with only six severe developmentally delayed patients and 20 mild to moderate developmentally delayed patients. Furthermore, there was no access to a variety of appropriate diagnostic culturally adopted tests for Persian-speaking children in this age group.

Finally, based on this study's findings concerning the improvement of delay developed hearing loss children in gross motor and social interaction after implantation and its follow-up rehabilitation program, it is suggested to proceed with a bigger sample size to observe the real difference between the groups. Also, we recommend performing another appropriate diagnostic culturally adopted test for Persian-speaking children (i.e., Bayley scale of infant and toddler) that evaluates other child's developmental aspects.

Table 4
Test of between-subjects effects for ASQ-3 subscales in both groups by the time of diagnosis of hearing loss (six months old).

Source	Dependent variables	SS	Df	Ms	F	Significance
Group	Communication	3262.745	1	3262.745	168.289	0.001
	Gross motor	11707.089	1	11707.099	316.663	0.001
	Fine motor	6381.811	1	6381.811	148.179	0.001
	Problem solving	9741.204	1	9741.204	497.366	0.001
	Social development	9328.272	1	9328.272	259.136	0.001
Error	Communication	950.000	49	19.388		
	Gross motor	1811.536	49	36.970		
	Fine motor	2110.346	49	43.088		
	Problem solving	979.365	49	19.987		
	Social development	1763.885	49	35.998		
Total	Communication	43750.000	51			
	Gross motor	67550.000	51			
	Fine motor	72375.000	51			
	Problem solving	72150.000	51			
	Social development	72175.000	51			
Corrected total	Communication	4212.745	50			
	Gross motor	13518.627	50			
	Fine motor	8492.157	50			
	Problem solving	10720.500	50			
	Social development	11092.157	50			

Table 5
Test of between-subjects effects for ASQ subscales in both groups after 18 months of implantation and rehabilitation (24 months old).

Source	Dependent variables	SS	Df	Ms	F	Significance
Group	Communication	10174.155	1	10174.155	424.687	0.001
	Gross motor	4.130	1	4.130	0.21	0.132
	Fine motor	10872.707	1	10872.707	297.626	0.001
	Problem solving	10565.648	1	10565.648	544.94	0.001
	Social development	3.285	1	3.285	0.128	0.939
Error	Communication	1173.88	49	23.957		
	Gross motor	958.615	49	23.957		
	Fine motor	1790.038	49	19.564		
	Problem solving	950.038	49	36.531		
	Social development	1255.538	49	19.389		
Total	Communication	74525.000	51			
	Gross motor	125975.000	51			
	Fine motor	73400.000	51			
	Problem solving	70200.000	51			
	Social development	118475.000	51			
Corrected total	Communication	11384.039	50			
	Gross motor	962.745	50			
	Fine motor	12662.745	50			
	Problem solving	11515.686	50			
	Social development	1258.824	50			

Funding

This research received no fund or grant from any funding agency in the public, commercial, or non-profit sectors.

Declaration of competing interest

The authors declare no conflict of interest.

Acknowledgements

The authors would like to thank all the parents of cochlear implanted children who collaborated in data collection.

References

[1] H.M. Fortnum, D.H. Marshall, A.Q. Summerfield, Epidemiology of the UK population of hearing loss children, including characteristics of those with and without cochlear implants–audiology, aetiology, comorbidity and affluence, Int. J. Audiol. 41 (3) (2002) 170–179, https://doi.org/10.3109/14992020209077181.
[2] E. Fitzpatrick, L. Brewster, Pediatric cochlear implantation in Canada: results of a survey, Can. J. Speech Lang. Pathol. Audiol. 32 (1) (2008).
[3] M. Srour, B. Mazer, M.I. Shevell, Analysis of clinical features predicting etiologic yield in the assessment of global developmental delay, Pediatrics 118 (1) (2006) 139–145, https://doi.org/10.1542/peds.2005-2702.
[4] N. Wakil, E.M. Fitzpatrick, J. Olds, D. Schramm, J. Whittingham, Long-term outcome after cochlear implantation in children with additional developmental disabilities, Int. J. Audiol. 53 (9) (2014) 587–594, https://doi.org/10.3109/14992027.2014.905716.
[5] C.E. Corrales, J.S. Oghalai, Cochlear implant considerations in children with additional disabilities, Curr. Otorhinolaryngol. Rep. 1 (2) (2013) 61–68, https://doi.org/10.1007/s40136-013-0011-z.
[6] Y.Y. Choo, P. Agarwal, C.H. How, S.P. Yeleswarapu, Developmental delay: identification and management at primary care level, Singap. Med. J. 60 (3) (2019) 119–123, https://doi.org/10.11622/smedj.2019025.
[7] J.B. Moeschler, M. Shevell, Comprehensive evaluation of the child with intellectual disability or global developmental delays, Pediatrics 134 (3) (2014) e903–e918, https://doi.org/10.1542/peds.2014-1839.
[8] L. Monshizadeh, R. Vameghi, M. Rahimi, F. Sajedi, S.B. Hashemi, F. Yadegari, et al., Is there any association between language acquisition and cognitive development in cochlear-implanted children? J Int Adv Otol 17 (3) (2021) 195–199, https://doi.org/10.5152/iao.2021.8990.
[9] L. Monshizadeh, R. Vameghi, F. Sajedi, F. Yadegari, S.B. Hashemi, P. Kirchem, et al., Comparison of social interaction between cochlear-implanted children with normal intelligence undergoing auditory verbal therapy and normal-hearing children: a pilot study, J. Int. Adv. Otol. 14 (1) (2018) 34, https://doi.org/10.5152/iao.2018.3663.
[10] T. Vidranski, D. Farkaš, Motor skills in hearing impaired children with or without cochlear implant–A systematic review, Coll. Antropol. 39 (Suppl 1) (2015) 173–179. PMID: 26434027.
[11] L.C. Edwards, R. Frost, F. Witham, Developmental delay and outcomes in paediatric cochlear implantation: implications for candidacy, Int. J. Pediatr. Otorhinolaryngol. 70 (9) (2006) 1593–1600, https://doi.org/10.1016/j.ijporl.2006.04.008.
[12] K.C. Johnson, J.L. DesJardin, D.H. Barker, A.L. Quittner, M.E. Winter, Assessing joint attention and symbolic play in children with cochlear implants and multiple disabilities: two case studies, Otol. Neurotol. Off. Publ. Am. Otol. Soc. Am. Neurotol. Soc. Eur. Acad. Otol. Neurot. 29 (2) (2008) 246–250, https://doi.org/10.1097/mao.0b013e318162f1f3.
[13] Y.S. Chang, I.J. Moon, E.Y. Kim, J. Ahn, W.H. Chung, Y.S. Cho, et al., Social skills and developmental delay: importance in predicting the auditory and speech outcomes after cochlear implantation in children, Acta Otolaryngol. 135 (2) (2015) 154–161, https://doi.org/10.3109/00016489.2014.973531.
[14] R. Vameghi, F. Sajedi, A. Kraskian Mojembari, A. Habiollahi, H.R. Lornezhad, B. Delavar, Cross-cultural adaptation, validation and standardization of ages and Stages questionnaire (ASQ) in Iranian children, Iran. J. Public Health 42 (5) (2013) 522–528. PMID: 23802111.
[15] N. Eze, E. Ofo, D. Jiang, A.F. O'Connor, Systematic review of cochlear implantation in children with developmental disability, Otol. Neurotol. Off. Publ. Am. Otol. Soc. Am. Neurotol. Soc. Eur. Acad. Otol. Neurotol. 34 (8) (2013) 1385–1393, https://doi.org/10.1097/MAO.0b013e3182a004b3.
[16] S. Archbold, S. Athalye, I. Mulla, S. Harrigan, N. Wolters-Leermakers, J. Isarin, et al., Cochlear implantation in children with complex needs: the perceptions of professionals at cochlear implant centres, Cochlear Implants Int. 16 (6) (2015) 303–311, https://doi.org/10.1179/1754762815Y.0000000012.
[17] A.I. Donaldson, K.S. Heavner, T.A. Zwolan, Measuring progress in children with autism spectrum disorder who have cochlear implants, Arch. Otolaryngol. Head Neck Surg. 130 (5) (2004) 666–671, https://doi.org/10.1001/archotol.130.5.666.
[18] S. Vlahović, B. Sindija, The influence of potentially limiting factors on paediatric outcomes following cochlear implantation, Int. J. Pediatr. Otorhinolaryngol. 68 (9) (2004) 1167–1174, https://doi.org/10.1016/j.ijporl.2004.03.016.
[19] R.F. Holt, K.I. Kirk, Speech and language development in cognitively delayed children with cochlear implants, Ear Hear. 26 (2) (2005) 132–148, https://doi.org/10.1097/00003446-200504000-00003.