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Clinical evaluation of cochlear implantation in children younger than 12 months of age

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ABSTRACT

Importance: Cochlear implantation (CI) is an effective therapy for patients with severe to profound sensorineural hearing loss. It remains controversial whether children younger than 12 months of age should undergo CI.

Objective: To evaluate the safety and effectiveness of CI in children younger than 12 months of age.

Methods: We performed a retrospective study of clinical data of pediatric patients younger than 12 months of age who underwent CI and were followed up for 1 to 2 years. Patients' developmental levels were evaluated by the Gesell score before CI. Intraoperative and postoperative complications were recorded to evaluate the safety of CI. Auditory and speech abilities were scored by the LittlEARS® auditory questionnaire (LEAQ), categories of auditory performance (CAP), speech intelligibility rating (SIR), infant-toddler meaningful auditory integration scale (IT-MAIS), and meaningful use of speech scale (MUSS) at 1, 2, 3, 6, 9, and 12 months after CI. The associations between clinical characteristics before CI and postoperative scores at 1 year after CI were analyzed by the linear mixed-effects model.

Results: Eighty-nine children (47 boys and 42 girls) were included in this study (mean age at CI, 9.2 ± 1.6 months). Sixteen patients were diagnosed with cochlear malformation and 16 underwent bilateral CI. No severe complications occurred in any patients. The mean developmental quotient of the Gesell score was 78.00 ± 10.03. The median LEAQ scores were 0, 5, 10, 16, 22, 26 and 30 before and at 1, 2, 3, 6, 9, and 12 months after CI, respectively. These findings implied that the LEAQ score greatly improved in the first year after CI. The overall CAP, SIR, IT-MAIS, and MUSS scores also increased with increasing duration after CI. No significant associations were detected between clinical characteristics (age, sex, implant number, pre-CI Gesell score, and inner ear malformation) and LEAQ outcomes at 12 months after CI.

Interpretation: With increasing duration after CI, auditory and speech behavior dramatically improve in young children. Our findings indicate that CI is feasible for children younger than 12 months of age.

KEYWORDS
Cochlear implantation, Age, Children, Outcome

INTRODUCTION

Cochlear implantation (CI) is regarded as an effective therapy for patients with severe to profound sensorineural hearing loss. Approximately 400 000 people worldwide receive CI; more than half of them are children. As of 2018,

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approximately 50 000 patients in China had received CI; more than 30 000 newborns in China are eligible for CI each year.[1] To obtain the maximum benefit of intervention regarding speech and language, many researchers are investigating the performance of CI in young children.

Various factors may affect the outcome of CI; age is considered an important preoperative factor. Some young children with meningitis are advised to undergo CI as soon as possible; however, it remains controversial whether a child younger than 12 months of age should undergo CI in routine clinical practice. Some researchers consider age to be the most important factor influencing the outcome of CI, because early CI can lead to better and more rapid postoperative outcomes.[2] However, other researchers suspect that early CI may increase the risk of complications.[3] Therefore, this study was performed to evaluate the safety and efficacy of CI in children younger than 12 months of age.

METHODS

Ethical approval

This study was approved by the Ethical Committee of Beijing Children's Hospital. Written formed consent was obtained from the patients' parents or guardians.

Patients

This study included pediatric patients who underwent CI before the age of 12 months at Beijing Children's Hospital, Capital Medical University from October 2012 to March 2018. Exclusion criteria were less than 1 year of follow-up, as well as the presence of non-congenital deafness (e.g., progressive hearing loss or meningitis). Computed tomography of temporal bone and magnetic resonance imaging of the inner ear were performed to diagnose inner ear malformations before CI.

Gesell score

The Gesell score includes five subscales: adaptability, fine motor, gross motor, language, and social skill evaluation. The developmental quotient (DQ) was calculated as follows: (development age/actual age) × 100. A DQ ≥ 86 indicates a normal level, DQ ≥ 76 and ≤ 85 indicates suspected neurological delay, DQ ≥ 55 and ≤ 75 indicates mild delay, DQ ≥ 40 and ≤ 54 indicates moderate delay, DQ ≥ 25 and ≤ 39 indicates severe delay, and DQ < 25 indicates profound neurological delay.[4]

Safety evaluation

Intraoperative complications, such as facial nerve injury (e.g., chorda tympani nerve injury), were recorded. Postoperative (short-term) complications were also recorded; these included balance disturbance, wound infection, scalp hematoma, and skin bruises. Furthermore, data were collected regarding long-term postoperative complications, such as implant displacement, meningitis, and cerebrospinal fluid otorrhea.

Post-CI outcome

The following tools were used to determine outcome post-CI. The LittlEARS® auditory questionnaire (LEAQ) contains 35 dichotomous questions. It was first proposed by Weichbold et al in 2005.[5] Higher LEAQ scores indicate advanced auditory and speech abilities. Categories of auditory performance (CAP) scores range from 0 to 7; 0 = no awareness to environmental sound, 1 = awareness to environmental sound, 2 = response to speech sound, 3 = identification of environmental sound, 4 = discrimination to some speech sounds without lip reading, 5 = understanding of common phrases without lip reading, 6 = understanding of conversation without lip reading, and 7 = telephone communication with a person.[6] Speech intelligibility rating (SIR) was used to evaluate speech ability; it was graded from 1 to 5 as follows: 1 = consequent sentences cannot be distinguished; 2 = consequent sentences cannot be understood, but a single word can be distinguished; 3 = consequent sentences can be distinguished, but the listener depends on lip reading; 4 = consequent sentences can be distinguished clearly by someone; 5 = consequent sentences can be distinguished by anyone.[7] The infant-toddler meaningful auditory integration scale (IT-MAIS) was used in this study; it contains 10 questions, each of which is scored from 0 to 4 (0 = never, 1 = seldom, 2 = sometimes, 3 = frequently, and 4 = always).[8] The meaningful use of speech scale (MUSS) was also used; this questionnaire contains 10 questions, each of which is scored from 0 to 4 (0 = never, 1 = rarely, 2 = occasionally, 3 = frequently, and 4 = always).[9] The postoperative outcomes of pediatric patients who underwent CI were evaluated using the LEAQ, CAP, SIR, IT-MAIS, and MUSS scores before and at 1, 2, 3, 6, 9, and 12 months after CI.

Statistical analysis

Continuous variables in this study are shown as means and standard deviations. These variables were compared by the two-sample *t*-test if data were normally distributed. Non-normally distributed data are shown as medians and interquartile ranges; these were compared using the nonparametric Wilcoxon–Mann–Whitney test. Categorical variables are shown as counts and proportions; these were compared using the χ^2 test. A linear mixed-effects model was used to analyze the relationships between clinical characteristics and post-CI scores. Statistical analyses were performed using SPSS Statistics, version 17.0 (IBM Corp., Armonk, NY, USA). A $P \leq 0.05$ was considered as statistically significant.

RESULTS

In total, 89 patients (47 boys and 42 girls) were enrolled in this study. All patients had been diagnosed with bilateral severe to profound sensorineural hearing loss. The mean age at CI was 9.2 ± 1.6 months. Sixteen patients (18%) had

been diagnosed with inner ear malformations (eight with large vestibular aqueduct syndrome, five with Mondini malformation, one with cochlear hypoplasia, and two with cochlear nerve dysplasia). Sixteen patients (18%) underwent bilateral simultaneous CI. Seventy-three patients (82%) underwent unilateral CI and had hearing aids on the other ear before CI. All patients were followed up for 1 to 2 years.

The developmental levels of the patients were evaluated by the Gesell score before CI. The total DQ of the Gesell score was 78.00 ± 10.03. The DQ of the gross motor subscale score was 84.78 ± 12.84, DQ of the fine motor subscale score was 80.42 ± 12.75, DQ of the adaptability subscale score was 84.13 ± 13.03, DQ of the language subscale score was 60.88 ± 19.21, and DQ of the social skill subscale score was 79.81 ± 11.40 (Figure 1). The DQ of the language subscale score was significantly lower (all $P < 0.001$) than the DQ values of the other four subscale scores (Table 1).

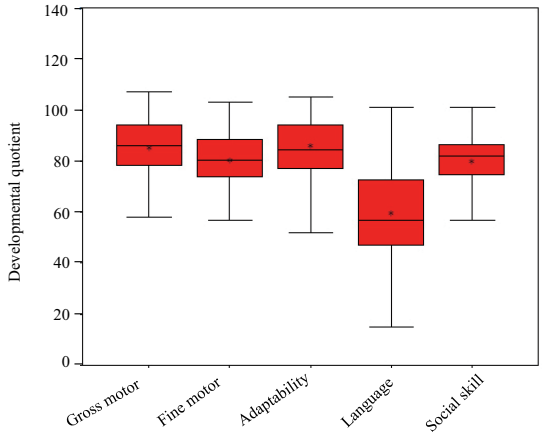

FIGURE 1 Developmental quotients of Gesell score subscales among patients before cochlear implantation. Mean values are indicated by asterisks (*).

No severe complications were observed in any patients. Facial nerve monitoring was used to avoid nerve injury. Intravenous antibiotics were provided to reduce the risk of infection. Balance disturbance occurred in two patients who had been diagnosed with Mondini malformation; they recovered without intervention, within 3 days after surgery. Skin bruising occurred in one patient; the patient exhibited spontaneous recovery within 1 week. Scalp hematoma occurred in one patient; the patient recovered after a single treatment involving puncture and liquid removal. Long-term postoperative complications (e.g., implant displacement, meningitis, and cerebrospinal fluid otorrhea) did not occur.

Postoperative outcomes were evaluated in our CI center at 1, 2, 3, 6, 9, and 12 months after CI. The median (Q1–Q3) scores for each evaluation tool (i.e., LEAQ, CAP, SIR, IT-MAIS, and MUSS) at each time point after CI are summarized in Table 2. The scores before CI and 1 year after CI were significantly different ($P < 0.05$). LEAQ scores showed the most obvious changes after CI.

A linear mixed-effects model with a compound symmetric variance structure was used to analyze associations between clinical characteristics (i.e., age, sex, implant number, pre-CI Gesell score, and inner ear malformations) and LEAQ scores at 1 year after CI. There were no significant associations between clinical characteristics and scores at 1 year after CI (all $P > 0.05$). Furthermore, no differences in LEAQ scores were observed between patients who underwent unilateral CI and those who underwent bilateral CI, as well as between patients with cochlear malformation and patients with normal cochlear formation (all $P > 0.05$).

DISCUSSION

In China, the guidelines for CI were first published in 2003[10] and then updated in 2013[11]; these guidelines greatly aided in standardization of CI in China. The guidelines will be further modified because of the continued progress of CI. Clinical practice guidelines for CI increasingly focus on the appropriate minimum age for surgery. In 2000, the US Food and Drug Administration approved CI in children older than 1 year of age; however, surgeons can elect to perform CI in children younger than 1 year of age if necessary.[12] In France, CI is advised before the age of 1 year.[13]

The safety of CI is the most important aspect of treatment for patients younger than 1 year of age. Globally, the rate of complications is approximately 16%.[14] Severe complications refer to conditions that may require additional surgery or prolong the hospital stay; no severe complications occurred in our study. This is consistent with a prior study in which patients younger than 12 months of age were not found to experience increased risk of surgical complications during CI.[15] However, some mild complications occurred

TABLE 1 Developmental quotient (DQ) of language subscale score, compared with DQ values of the other four subscale scores, in patients younger than 1 year of age who underwent cochlear implantation

Comparable subscale	Mean difference	Standard error	95% confidence interval of the difference		*t*	*P*
			Lower	Upper		
Gross motor vs language	23.9028	2.4688	18.9802	28.8253	9.682	< 0.001
Fine motor vs language	19.5417	2.4390	14.6784	24.4049	8.012	< 0.001
Adaptability vs language	23.2500	2.2376	18.7883	27.7117	10.391	< 0.001
Social skill vs language	18.9306	1.9861	14.9704	22.8907	9.532	< 0.001

TABLE 2 Evaluation tool scores at 0, 1, 2, 3, 6, 9, and 12 months after cochlear implantation

Variable	Month after cochlear implantation							P^*
	0	1	2	3	6	9	12	
LEAQ	0 (0–1)	5 (3–9)	10 (7–14)	16 (10–18)	22 (17–26)	26 (24–32)	30 (27–33)	< 0.001
CAP	0 (0–1)	1 (1–2)	2 (1–2)	2 (2–3)	3 (2–3)	3 (2–3)	4 (3–5)	< 0.001
SIR	1 (1–1)	1 (1–1)	1 (1–1)	1 (1–1)	1 (1–1)	1 (1–2)	2 (1–3)	0.002
IT-MAIS	0 (0–0)	4 (2–7)	9 (9–17)	13 (10–18)	17 (12–24)	21 (17–24)	24 (17–25)	0.007
MUSS	0 (0–0)	1 (1–7)	2 (1–9)	2 (2–12)	3 (3–14)	3 (3–15)	4 (3–18)	0.015

Data are shown as median (Q1–Q3). *P values are shown for comparisons between before and 12 months after cochlear implantation. LEAQ, LittlEARS® auditory questionnaire; CAP, categories of auditory performance; SIR, speech intelligibility rating; IT-MAIS, infant-toddler meaningful auditory integration scale; MUSS, meaningful use of speech scale.

in our study. Balance disturbance occurred in two patients who had been diagnosed with Mondini malformation by computed tomography. This disturbance in balance was caused by intraoperative outflow of cerebrospinal fluid. Skin bruising occurred in one patient because of excess pressure on the skin. Scalp hematoma occurred in one patient because of insufficient pressure on the skin. A relatively small mastoid cavity is a large challenge for surgeons during CI. Additionally, uncontrolled bleeding should be carefully monitored because of the low blood volume in infants. Cochlear malformations increase the risk of surgical complications, including facial nerve injury and cerebrospinal fluid gusher; however, these complications can be avoided by facial nerve monitoring and insertion of temporal muscle tissue. Evaluation of surgical complications from CI has differed among published studies. The majority of surgical complications after CI in patients younger than 1 year of are soft tissue-related; they may be caused by the small skull size and thin skin flap. In our study, the soft tissue-related complication rate was 2.25%, which is similar to the rate published in a prior study (2.74%).[15]

A positive correlation has been reported between age and developmental delay.[16] In our study, the mean DQ values of subscale scores were nearly normal, with the exception of the language subscale score. The mean DQ of the language subscale score was 60.88 ± 19.21, which indicated mild delay. This finding also suggested that the performance of CI is appropriate in patients younger than 1 year of age. Presumably, earlier CI is useful because plasticity declines in the auditory cortex when sound stimulation is absent. Hearing disability may influence overall development; the auditory cortex will assume other functions if no intervention is performed.[17] Functional changes in the auditory cortex may affect the outcome of CI. Cortical processing in deaf patients reportedly differs from such processing in normal hearing individuals.[18] Similar conclusions have been reached based on findings in animal studies.[19] CI is an effective treatment for helping patients to restore auditory input and for establishing a functional auditory network. Optimal outcomes can be achieved if CI is performed before the age of 1 year for most CI candidates.[20]

There is a need for evaluation tools to monitor early auditory development in children who undergo CI. In the present study, the LEAQ scores improved more rapidly, compared with other scores, with increasing duration after CI during the first year postoperatively. The LEAQ has been previously validated[21]; a significant correlation has been reported between hearing age and LEAQ scores in children who undergo CI. Rapid changes in LEAQ scores will aid in evaluation of patient progress after CI; they will also aid parents in following a rehabilitation program. Performance of CI in patients younger than 1 year of age has been shown to result in better auditory and speech scores, compared with CI performed in older patients.[22] In our study, the median LEAQ score at 1 year after CI was 30; this was better than the score of 24 previously observed in a group of children with a mean age of 22.27 ± 10.66 months.[23] Therefore, earlier CI could be advised for patients with congenital sensorineural hearing loss.[24]

In our study, there were no significant associations between clinical characteristics and LEAQ scores at 1 year after CI. Advantages of bilateral CI include sound source localization and speech recognition in a noisy environment. However, patients who undergo bilateral CI require long-term follow-up. The abovementioned advantages cannot be identified in a short period at a young age[25]; they also cannot be clearly identified by using our evaluation tools. The percentage of patients with inner ear malformations was 18% in our study; most of these malformations may not affect the outcome of CI (e.g., large vestibular aqueduct syndrome and Mondini malformation).[26]

There were some limitations in our study, including its small sample size and short follow-up period, as well as the lack of a control group of patients who underwent CI after 1 year of age. In addition, no tools were used that could evaluate postoperative sound source localization and speech recognition. We will address these limitations in future studies.

In conclusion, our study showed that early CI is feasible for children under 1 year of age with severe to profound sensorineural hearing loss. All surgical procedures must

be performed carefully. The ideal age for CI must be determined based on each patient's individual situation.

CONFLICT OF INTEREST

The authors declare no conflicts of interest.

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