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Original article

Receptive speech in early implanted children later diagnosed with autism

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ABSTRACT

Introduction: Incidence of children with autism spectrum disorder (ASD) is rising through the years with estimated 1 in 68 in the US in 2014. This incidence is also rising in the population of congenitally deaf children. Favorable outcome after early cochlear implantation is expected due to plasticity and reorganization capacity of brain in infants and toddlers, but outcomes could be significantly modified in children with diagnosed ASD. Current methods of screening for autism have difficulties in establishing diagnosis in children who have both autism and other developmental delays, especially at such an early age. The aim of the study was to assess the development of auditory perception and speech intelligibility in implanted children with profound congenital hearing loss who were diagnosed with ASD comparing to those who were typically developing.

Material and methods: Fourteen children underwent cochlear implantation; four were later diagnosed with ASD and ten were typically developing. All children underwent intensive postoperative speech and hearing therapy. The development of auditory perception and speech intelligibility was assessed using the Categories of Auditory Performance (CAP) and the Speech Intelligibility Rating (SIR) during the 5-years follow-up.

Results: In children later diagnosed with ASD, auditory processing developed slowly. Depending on the individual capabilities, by the age of six they could identify environmental sounds or discriminate speech sounds. Speech Intelligibility in children with ASD was at best rated as category 2, with very little or no progress up to the age of six, despite extensive speech and language therapy. Communication skills were strongly affected by a degree of autistic features expression.

Conclusion: Preoperative psychological assessment in congenitally deaf infants should be expanded by the use of validated instruments for early detection of autism. The possibility of developing ASD should be kept in mind by all professionals involved in programs for cochlear implantation.

1. Introduction

Incidence of children with autism spectrum disorder (ASD) is rising through the years. In the 2014, the Autism and Developmental Disabilities Monitoring (ADDM) Network estimated that about 1 in 68 children with ASD were identified in the United States. Population of children with congenital hearing loss is not the exception to this trend. ASD is defined by clinical assessment and onset atypical social behavior; disrupted verbal and non-verbal communication; and unusual patterns of highly restricted interests and repetitive behaviors. Being a highly heterogeneous disorder, there are significant differences in children's developmental course, where some manifest signs of the disorder from early infancy and others experience behavioral regression in the second or third year of life [1,2]. Screening for autism was recommended in all children between the ages of 18 and 24 months [3] and, in the absence of any biological markers for autism, is focused on behavioral assessment. Current methods of screening may have difficulties in establishing diagnosis in children who have both autism and other developmental delays especially at such an early age [4].

On the other hand, early diagnosis of deafness became of at utmost importance for timely cochlear implantation and postoperative speech and language development. In the last decade there are increasing number of studies indicating that cochlear

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implantation in children younger than 24 months enables better results in receptive and expressive language development and speech perception, than in children implanted after 24 months of age [5,6] Guidelines for audiological assessment of children with developmental delays, especially those with delays in social and language development, are clear [7]. Results of behavioral assessment could be misinterpreted in children with profound sensoneural hearing loss younger than 18 months who carry a certain risk for ASD. Outcomes of cochlear implantation in deaf children who are later diagnosed with ASD are, at least, uncertain and unpredictable. This represents a still not well-researched aspect of cochlear implantation and rehabilitation with a growing number of patients.

In our study, auditory perception and speech intelligibility were assessed in cochlear-implanted children with profound congenital hearing loss who were later diagnosed with ASD. The aim of the study was to follow the development of these skills and to compare that progress to auditory perception and speech intelligibility skills in typically developing children with profound congenital hearing loss.

2. Material and methods

Prospective study was conducted on fourteen children who underwent cochlear implantation according to pediatric cochlear implantation program on Clinic for otorhinolaryngology and maxillofacial surgery of Clinical Center of Serbia during 2008 and 2009. Informed consent was obtained from the parents of all participating children and the study was approved by the Institutional Ethical Board. Patients involved in the study underwent preoperative medical evaluation (ENT exam, magnetic resonance imaging (MRI) and computed tomography (CT) of the endocranium and temporal bone); audiological and hearing aid evaluation; psychological and speech and language assessment.

All children included in the study had congenital bilateral profound sensorineural hearing loss with minimal or no benefit from appropriate hearing fitting and limited acquired language skills provided by amplification, prior to cochlear implant surgery. All children were implanted between 12 and 18 months of age. All subjects used Medel PULSARci10 cochlear implant and Opus I speech processor. Four children were later diagnosed with ASD (three male and one female) and ten children were typically developing (four males and six females). All children underwent intensive postoperative speech and hearing therapy.

The development of auditory perception and speech intelligibility was assessed using the Categories of Auditory Performance (CAP) [8] and the Speech Intelligibility Rating (SIR) [9] The CAP classify auditory receptive ability into eight performance categories that increase in difficulty (from “no awareness of the environment” to the “use of the telephone with known users”). The SIR consists of a five point rating scale that measure speech intelligibility (from “unintelligible speech” to a child being rated as having “connected speech that is intelligible to all listeners”) increasing in levels of complexity along with the child’s speech production. Testing was done at the second year of age (6 to 12 months after cochlear implantation), and then every year until the age of 6.

All results were analyzed using the Statistical Package for Social Sciences (SPSS) 20.0 (SPSS Inc., Chicago, IL, USA). Statistical comparisons were made using the Mann–Whitney test. A P-value of < 0.05 was determined to be statistically significant.

3. Results

Fourteen children were included in the study, four children later diagnosed with ASD (three male and one female) and ten children who were typically developing (four males and six females). Information about gender, diagnosed hearing loss age, age at implantation, as well as CAP and SIR test scores (from the age of 2 to 6) for every child were given in the Table 1. Average age at time of diagnosing hearing loss was 10.4 months, and average age at time of cochlear implantation was 16 months.

Diagnosis of ASD was established from the age of 37 months to 61 months (median age 44 months). Average scores of CAP and SIR test of typically developing children and children later diagnosed with ASD tested from the age of 2 to 6 were shown in the Fig. 1. Considering both CAP and SIR scores, there is stable progress in typically developed children. In children with cochlear implants later diagnosed with ASD, auditory processing developed slowly. Depending on the individual capabilities of the children, by the age of six they could identify environmental sounds or discriminate speech sounds. Speech Intelligibility in children with ASD was at best rated as category 2, with very little or no progress up to the age of six, despite extensive speech and language therapy. There was a significant statistical difference between CAP and SIR scores between two groups on every testing during follow-up period (Mann–Whitney test, $P < 0.05$).

4. Discussion

Almost 40% of children with a diagnosed permanent hearing loss have an additional disability [10]. If there is co-occurrence of ASD and hearing loss, establishing both diagnosis early in the child’s development should be crucial for further therapy. Diagnosis of ASD could be complicated to establish at such an early age, especially because both ASD and hearing loss result in language and communication delays [11,12]. Median age of ASD diagnosis for children with hearing loss was described to be from 49 to 66 months [13–15], though those studies indicated that hearing loss would further delay diagnosis of ASD. In our study median age was lower than described by other authors. Among various instruments considered suitable for assessment of ASD in normal hearing population, M-CHAT have been outlined as most promising for early screening of autism [16,17]. Concerns were already raised considering these instruments and their use in children with hearing loss [18,19]. Further studies must be done to accurately calculate sensitivity and specificity of these instruments in population of children with congenital hearing loss, to validate these instruments for this population and establish more precise ways of scores interpretation.

Early cochlear implantation provides favorable outcome, enables age-appropriate speech-language development and potential for mainstreaming and social integration [5]. In our study, expected age-appropriate auditory performance and speech intelligibility skills were obtained in a group of typically developing early implanted children by the age 4 or 5 years [20]. After receiving a cochlear implant, children with ASD had shown improvements in behavior, vocalization, eye contact, reaction to sound, and response to requests [21]. Also, children with ASD showed limited development in auditory perception and speech intelligibility after cochlear implantation compared to deaf patients with other additional disabilities [22,23]. In this study, all children were implanted by the age of 18 months, which could be considered early comparing to other studies [21,23,24]. Auditory performance skills were developed in various degree, and none of our subject developed intelligible expressive speech. Clinical picture and treatment responsiveness varies significantly among children with autism, and children who appear the same at certain age, could show different developmental trajectories at later ages [1]. The same variability in postoperative results after speech and language therapy in implanted children with ASD is to be expected. Speech intelligibility skills were rated very low during follow-up time, in

Table 1
Gender, age of diagnosed hearing loss, age at implantation, CAP and SIR test scores of children.

	Gender	Age of diagnosed hearing loss (months)	Age at impantation (months)	CAP score					SIR score				
				2 yr	3 yr	4 yr	5 yr	6 yr	2 yr	3 yr	4 yr	5 yr	6 yr
Typically developing children													
1	Female	14	18	2	3	4	5	6	2	3	4	5	6
2	Male	12	18	3	4	6	6	7	2	2	3	3	4
3	Female	14	18	3	4	4	5	5	2	3	3	4	4
4	Male	15	17	3	3	4	5	5	1	2	3	4	4
5	Male	2	9	3	4	6	7	7	2	3	5	5	5
6	Female	13	18	3	4	5	7	7	2	3	4	5	5
7	Female	2	15	2	3	4	5	5	1	2	3	3	4
8	Female	14	18	4	5	6	7	7	2	4	5	5	5
9	Female	12	16	4	5	6	7	7	2	4	5	5	5
10	Male	7	15	3	4	6	7	7	2	3	5	5	5
Children with ASD													
1	Male	9	13	1	1	2	3	3	1	1	2	2	2
2	Male	12	18	1	1	2	3	4	1	1	1	2	2
3	Female	8	14	1	1	1	1	1	1	1	1	1	1
4	Male	12	17	1	1	2	2	2	1	1	1	2	2

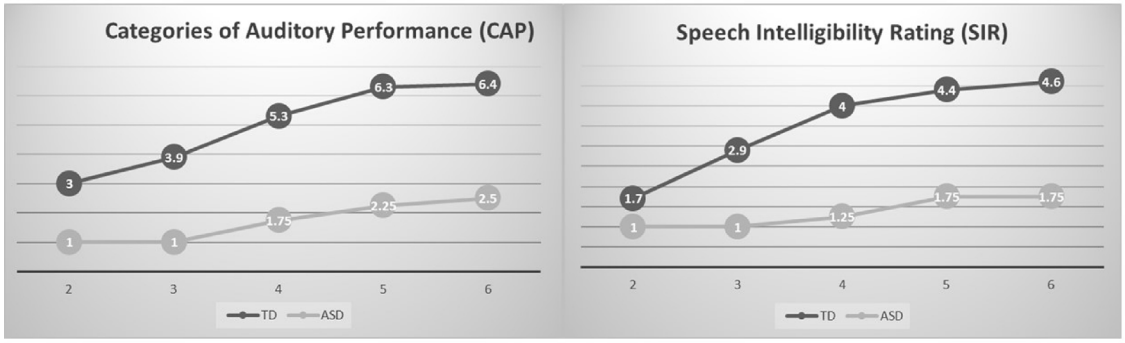

Fig. 1. Average scores of CAP and SIR test of typically developing children and children later diagnosed with ASD.

spite intensive postoperative speech therapy. In this study, communication skills, receptive and expressive language development were strongly affected by a degree of autistic features expression. In implanted children with ASD expectations and outcomes of cochlear implantation are significantly modified [25], and the main goal should be to set foundation for communication system which will eventually support the use of sound and speech [15].

5. Conclusion

Precise and valid screening instruments for infants and toddlers are needed in the future to lower the age of ASD detection in congenitally deaf children and should be included in routine preoperative psychological assessment before cochlear implantation. The possibility of developing ASD should be kept in mind by all professionals involved in counseling of the parents whose infants are selected for early cochlear implantation.

Disclosure of interest

The authors declare that they have no competing interest.

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