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Speech Therapy for a Child with Neurosarcoidosis: A Case Report

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Case Report

Abstract

Introduction: Sarcoidosis is a multisystem granulomatous disorder with an unidentified origin. The prevalence of sarcoidosis in children is rare and also its frequency is unknown. There is some evidence of central nervous system (CNS) involvement in patients with sarcoidosis. The effect of rehabilitation, especially speech therapy, in these patients has not been reported before. This study reported the speech and language characteristics of a child with neurosarcoidosis before, during, and after speech therapy by formal and informal speech and language assessments.

Case Report: The case was a 4-year-old boy with neurosarcoidosis. The language comprehension and expression skills of the child were delayed and weak in comparison to their normal peers. During speech therapy, the child's language comprehension and expression skills were improving, which might indicate the effectiveness of speech therapy. The improvements in the child's language abilities were seen only during speech therapy, while after speech therapy, his language skills, speech intelligibility, and verbal output showed deterioration to some extent. Unfortunately, not only the improvement of the child's language was not maintained after the speech therapy, but also this decline was associated with the peak of the child's disease.

Conclusion: To conclude, more studies are needed about the time of providing speech therapy and its dose in children with neurosarcoidosis at different phases of disease progression.

Keywords: Neurosarcoidosis; Speech therapy; Language development; Pediatric; Case report

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Introduction

Sarcoidosis, a multisystem granulomatous disorder with unknown origin (1), shows higher prevalence in the Northern Europe and African-Americans (2, 3) and lower prevalence in Asia (4, 5); however, the global prevalence of sarcoidosis and neurosarcoidosis in children is rare and its extension is unknown (6). Most cases have been reported children with sarcoidosis between 13 and 15 years old (6). Moreover, it usually affects the lungs, skin, eyes, and liver (2). The most common symptoms of nervous system involvement are sensory problems and the major involvement is the central nervous system (CNS), including the parenchyma and meninges (7). In overall, there is much evidence of great involvement of the CNS, especially the facial, optic cranial (1), and the trigeminal nerve (8). Consequently, patients with sarcoidosis are expected to have some difficulties in their speech, language, and swallowing (9) and providing rehabilitation programs like speech therapy can be beneficial for them, especially in children patients who are in the golden time of their development. To our best knowledge, while there are some case reports about children with neurosarcoidosis, no study has reported the effect of speech therapy on a child with neurosarcoidosis. For one, nine children with neurosarcoidosis have been reported in 2015 and only three of these reports have briefly described the speech and language characteristics of them without specific details (6). The present study reported the speech and language profile of a child with neurosarcoidosis before, during, and after speech therapy.

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Case Report

History

The case was a 4-year-old boy with neurosarcoidosis. The child's family referred to a speech therapy clinic one year after the beginning of his disease and four months after his last hospitalization. The family's complaint was a drop of his speech intelligibility and a reduction in length of utterance. According to their report, the child also had a speech delay before the beginning time of disease. Before sarcoidosis, the mean length of utterance (MLU) of the child's sentences was about two words and it decreased to less than two words after sarcoidosis. At birth time, he was full-term and was born by normal vaginal delivery. He had a history of seizure at 26 months and at first, a brain abscess was diagnosed with magnetic resonance imaging (MRI). Parents had a healthy condition and also they had a first-degree family relationship. No history of disability was observed in relatives. Microscopic examination of brain and arachnoid tissues displayed that some sections of brain tissues had multiple noncaseous granulomas surrounded by multinucleated giant cells and lymphocyte. Moreover, foci of calcification and formation of Schaumann bodies were seen in the background and some parts of giant cells' cytoplasm. Furthermore, sections of neural tissue and connective tissue were seen in the arachnoid. After that, the child received ceftriaxone intravenously every 12 hours. In the next MRI, the brain's spot changed into the larger size; thus, the child was hospitalized and the length of hospitalization was 2 months.

Finally, neurosarcoidosis virus was diagnosed by magnetic resonance (MR) spectroscopy. Next, prednisolone 50 ml and phenobarbital were prescribed which reduced the inflammation of the spots
and turned them into a smaller size. The results of clinical examinations performed by the pediatric neurologist were as follows: muscle weakness in the right half of the body, drooling, decreased language comprehension, decreased motor balance, and decreased speech intelligence.

Clinical Findings

Speech and Language Assessment: After getting family's history, his speech and language abilities were assessed as below:

1. Oral/speech movement control (informal observational assessment of structure and function)
2. Diadochokinetic tasks (10)
3. Phonetic test (11)
4. Informal assessment of language comprehension and expression skills

According to the results (Tables 1 and 2), he had a moderate prognosis.

Table 1. Oral-motor function, resonance, and loudness before, during, and after speech therapy

Outcomes				Results		
				After speech therapy	During speech therapy	Before speech therapy
Oral motor structure/function						
1	Lips	Structure	Anatomy	Normal	Normal	Normal
			Symmetry/asymmetry	Normal	Normal	Normal
		Function	Power	Weak - there was no resistance to stretching	Normal	Untestable
			Range of motion	Normal	Normal	Untestable
2	Tongue	Structure	Anatomy	Normal	Normal	Normal
			Symmetry/asymmetry	Normal	Normal	Normal
		Function	Power	Normal	Normal	Untestable
			Range of motion	Slow	Slow	Untestable
3	Soft palate uvula	Structure	Anatomy	Normal	Normal	Normal
			Symmetry/asymmetry	Normal	Normal	Normal
		Function	Power	Normal	Normal	Untestable
			Range of motion	Normal	Normal	Untestable
4	Uvula		Structure	Normal	Normal	Normal
			Function	Normal	Normal	Untestable
5	Hard palate		Structure	Normal	Normal	Normal
			Function			Untestable
6	Teeth		Structure	Normal	Normal	Normal
7	Mandible		Structure	Normal	Normal	Normal
			Function			Untestable
8			Gag reflux	Hypersensitivity	Hypersensitivity	Untestable
9			Resonance	Hyper-nasality (conceptuality)	Hyper-nasality (conceptuality)	Untestable
10			Loudness	Decreased (level 3) (14)	Normal	Decreased (level 2) (14)

Table 2. Speech and language characteristics before, during, and after speech therapy

Outcomes	Results		
Speech assessment			
Diadochokinetic testing	13.05 seconds per minute	The same as before treatment	Untestable
Phonological errors	Distortion of /p, m, d, f, v, s, z, š, ẑ, ʤ, h/	The same as before treatment	Untestable
Phonological process	Deletion of /n, h/	The same as before treatment	Untestable
	Substitution /f, v, t, d, z, n, ĵ, g, ŝ, ẑ/	The same as before treatment	Untestable
Language assessment			
Language comprehension	Normal	Normal	Comprehending simple sentences, for example: come here, sit down, eat, etc.
Language expression			
MLU (word)	1.6	3	Not speaking spontaneously and just repeating some simple words with auditory and visual prompts
	Limited word diversity (commonly used nouns and verbs) -Limited use of morphological features -No use of pronouns -No inversion of pronouns -Mismatch between verb and subject -Lack of proper use of prepositions and conjunctions -Not using complex sentences	Expressing two- and three-word sentences with structure S + V S + O + V	-Echolalia -Decreased spontaneity in communication -Decreased initiation at the relationship
Imitation	Ability to fast imitate one- to three-syllable words	Ability to imitate three-word sentences	Weakening of imitation of simple single words (although presented with visual and auditory prompts)

MLU: Mean length of utterance

Speech and Language Therapy: Two sessions of speech therapy were provided for this case weekly and each session lasted for 45 minutes. In addition to speech therapy, he received two sessions of occupational therapy weekly at home and also his parents were getting counseling sessions weekly. Over first sessions of speech therapy, the treatment was started with a child-centered approach to familiarize him with the environment and helped him to feel more comfortable. After two months, he gradually switched to a hybrid approach (12) which was presented according to the child's language development. Its stages are in the following:

1. Focus stimulation (12)
2. Vertical structuring (12)
3. Inclusive education (milieu teaching) (12)
4. Script therapy (12)

Six months after starting speech therapy, his speech and language abilities were assessed again. Then speech therapy was continued till two months later when his disease reached a peak; therefore, his family could not participate in speech therapy sessions regularly because of his medical treatments. Consequently, he could not complete speech therapy program. One year after the first session, while he did not participate in speech therapy, their family referred again for re-assessment. By this way, three data points were achieved including before, during, and after speech therapy. According to the results (Tables 1 and 2), his expressive language improved to some degree during speech therapy (MLU: 1.6 to 3). This improvement probably indicates the effectiveness of the speech therapy. In addition, some improvement in speech, voice loudness, and comprehensive language was observed. Besides, the child's speech intelligibility increased from 60% to 100% in contrary to his oral motor skills which remained unchanged during the intervention period. Unfortunately, after the speech therapy, language comprehension and expression showed a significant decrease and his speech output dramatically dropped. In general, the child's speech and language

improvement was not maintained after the intervention period, and this decline was associated with the peak of his disease.

Recommendations

While the child was improved to some extent by speech and language therapy in the appropriate time, which could not be seen before and after this therapy, it is strongly recommended that practitioners refer these patients to a speech and language therapist as soon as possible. In this way, not only speech therapy could decrease the pace of the deterioration in communication abilities like speech, language, and swallowing in the patients, but also in some cases, it can improve these mentioned abilities through an organized plan.

Conclusion

The current report presents the speech and language profile of a child with neurosarcoidosis before, during, and after speech therapy by descriptive informal assessments of speech and language. It seems that providing on-time speech therapy as a rehabilitation program for a child with neurosarcoidosis can improve the child's speech and language abilities. Given that the early years of a child's development are the golden time of language development (12), early detection and early intervention for speech and language rehabilitation probably can prevent exacerbation of symptoms and possibly helps these patients to have better speech, language, and communication. Thus, providing speech therapy possibly slows down the deterioration of their speech, language, and communication skills. Of course, more studies are needed on the time and dose of speech therapy in these children. It is suggested that further studies examine the effect of speech therapy on patients with sarcoidosis in different ages. In addition, using formal speech and language assessments to report language profile of these patients in various stages of their disease is also recommended; hence, identifying these changes occurs with more details. Finally, because of the various involvement of different organs among different races with sarcoidosis (13) and also different involvement of the nervous system in patients with sarcoidosis (6), it is better for neurologists to refer these children at the beginning of diagnosis to a speech and language pathologist for a comprehensive assessment of speech and language. It leads to early detection of any delay or deviation in the development of speech and language, and also start of speech therapy if necessary. Thus, the golden time of the development period as an efficient agent in neuroplasticity was applied (14).

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Providing equipment and study sample: Masoumeh Amirkhani

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Responsibility for maintaining and integrity of the study from the beginning to publication and responsibility for responding to the opinions of referees: Masoumeh Amirkhani, Marzieh Ostadi, Amir Zalpour

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