

Case Report

Speech and swallowing difficulties and rehabilitation in osmotic demyelination syndrome: A single case report

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ARTICLE INFO

Article history: Received 07-11-2021 Accepted 30-11-2021 Available online 25-01-2022

Keywords: Hyponatremia mutism dysphagia dysarthria

ABSTRACT

Objective: Aim of the work was to highlight the speech and swallowing difficulties that are associated with osmotic demyelination syndrome and also the importance of speech and language therapy in patients with osmotic demyelination syndrome.

Materials and Method: In this case report, a 68 years old female who developed osmotic demyelination syndrome as a result of hyponatremia correction has been described. The patient developed motor, speech and swallowing difficulties after 10^{th} day of hyponatremia correction. For assessing speech and language skills, Bedside Screening Test for Persons with Aphasia was used. Gugging Swallowing Screen was used for assessing the swallowing skills.

Results: The assessment results indicated that the patient's auditory comprehension skills were preserved and other verbal skills were affected as the patient had mutism. Also the patient had slight dysphagia with lower risk of aspiration. With speech-language intervention, improvement was observed in swallowing and verbal communication skills.

Conclusion: ODS is a rare clinical condition. In order to have a better perspective about speech, language and swallowing skills in individuals with ODS, replication of such studies are essential.

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1. Introduction

Osmotic demyelination syndrome (ODS) is a rare clinical conditionin which there is loss of myelin in many portions of central nervous system. Based on the occurrence of demyelination, ODS is classified into central pontine myelinolysis and extrapontine myelinolysis.^{1,2} Many factors that increase the risk of ODS include liver disease, alcoholism, malnutrition, duration of hyponatremia and hypokalemia. ODS is a neurologic condition which occurs after rapid correction of hyponatremia. Also, in patients with ODS due to hyponatremia where the serum sodium was lesser than or equal to 126 mmol/L at initial stages

were followed by hyponatremia correction (correcting the concentration of serum sodium).³ After overly rapid correction of severe hyponatremia, there is typically 2 to 6 days delay in the clinical manifestation of ODS. The clinical features of ODS includes movement disorders, confusion, paraparesis or quadriparesis, disorientation, dysphagia, dysarthria, obtundation, and coma. Severely affected patients may become "locked in" or patients are awake but they are unable to move or communicate.⁴

This case report highlights the speech and swallowing difficulties that are associated with ODS and also the importance of speech and language therapy in a case with ODS.

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2. Case Report

A case of 68 year old female with a medical history of osmotic demyelination syndrome which was developed as a result of correction of hyponatremia (Her initial sodium level was less than 105 mEq/L). The patient did not have any difficulties in speech and swallowing skills before the hyponatremia correction. After 10^{th} day of hyponatremia correction, she started to present clinical features of speech and swallowing difficulties. She had difficulty in moving right side of the body (right hemiplegia) and manifested unilateral facial palsy (right side). After 12^{th} day of hyponatremia correction, the patient was given nasogastric feeding (Ryles tube). Her MRI report findings revealed bilateral caudate nucleus and lentiform nucleus to appear bulky, thalamic hyperintensity, cerebelleum hyperintensity, bilateral fronto-parietal lobe hyperintensity. The aim and objective of the study was explained to the patient's caregiver and written consent was obtained from the caregiver.

2.1. Speech, language and swallowing evaluation

A comprehensive evaluation was carried out by the speech language pathologist. The native language of the patient was Kannada. She was monolingual and had completed 10th standard. Her hearing skills were normal. During the evaluation, the patient was conscious, alert and oriented though emotionally unstable. The patient presented mutism, and was unable to give verbal answers. The patient was able to follow simple verbal commands. Bedside Screening Test for Persons with Aphasia in Kannada⁵ was administered. On Bedside Screening Test, in auditory verbal comprehension domain, She got a score of 8 out of 10 in yes/no subsection (she answered through gesture), a total score of 10 in pointing task and auditory word recognition task, a score of 8 out of 10 in verification task (she answered through gesture), a score of 8 out of 10 in sequential commands. Overall, in auditory verbal comprehension domain, she got a score of 44 out of 50. Among all the domains (spontaneous speech, auditory verbal comprehension, repetition, naming, reading and writing), the patient was able to do only auditory verbal comprehension domain which included subsections such as yes/no questions, pointing task, auditory word recognition task, verification task and sequential commands. On oral mechanism examination, lip asymmetry was observed (right sided deviation) and lip seal was absent. Elevation, lateralization, protrusion of tongue were found to be present. Pooling of saliva was observed. In swallowing evaluation, the patient was able to do dry swallow but delayed hyolaryngeal elevation was observed during swallowing of pureed liquid and semi-solid and no cough, nasal regurgitation or throat clearing were observed. Prolonged oral transit time and stasis in oral cavity were observed. The

swallowing evaluation also included Gugging Swallowing Screen $(GUSS)^6$ to get information on the severity of dysphagia and aspiration risk. On GUSS, in the indirect swallowing test, the patient got a score of 3 points and in the direct swallowing test, a score of 13 points. So a total score of 16 was obtained. Based on the patient's total score, the severity code for the patient in GUSS was slight dysphagia with low risk of aspiration.

2.2. Intervention

The patient attended a eight speech and language therapy sessions (45 minutes each) twice weekly over a period of four week. Since the patient presented mutism with language comprehension being preserved, speech and language therapy was initiated with the major goal of reestablishing verbal communication. The patient was using mostly head movement and gestures to express her needs. Since, the patient was not able to perform spontaneous speech, repetition, naming, reading and writing domains of the Bedside Screening Test, So, the first short-term goal was to improve functional verbal language. The language stimulation was initiated with verbal imitation of isolated sounds along with gestures and orthographic cues. Patient showed good progress in each speech and language therapy session. At the initial stage of speech and language therapy session, the patient started to utter sounds followed by words and then was able to produce simple phrases and sentence. The patient speech showed features of dysarthria like articulatory imprecisions, reduced loudness, affected prosody (monotonous and lack of stress while speaking) and reduced rate of speech. Though there was good improvement in verbal communication but total re-establishment of verbal fluency and speech intelligibility was not achieved. On Perceptual Speech Intelligibility rating scale (7 point rating scale was developed at Ali Yavar Jung National Institute of Speech and Hearing Disabilities, Mumbai, 2003) the patient's speech intelligibility was rated as 4 which indicated Intelligible with careful listening although some words were unintelligible.

Swallowing skills were also worked on simultaneously along with language stimulation during each session. The patient had oro-pharyngeal dysphagia. The major goal was to improve lip closure and also to improve laryngeal elevation. To achieve these goals, oro-motor exercises and swallowing maneuvers were carried out. Direct swallowing therapy were also carried out where different food consistencies (semiliquid, pureed liquid, liquid, semisolid and solid) were trialed. With these interventions, the patient's swallowing skills for different food consistencies improved considerably. Patient was able to maintain lip seal during swallowing. Drooling of saliva was controlled. The patient was using the nasogastric feeding for 25 days, since the oral feeding was enabled as a result of intervention, the ryles tube was removed. Therefore, with intervention the patient's speech and swallowing skills were improved.

3. Discussion

In this case study, the patient developed motor, speech and swallowing difficulties after 10^{th} day of hyponatremia correction. To know the importance of speech and language assessment and intervention in patient with ODS, discussion is divided into two subsection as assessment and intervention.

3.1. Assessment

In the present study, Bedside Screening test indicated that preservation of auditory verbal comprehension only and not any other verbal skills. This might be attributed to the fact that the patient had mutism. Earlier studies have also reported the presence of mutism in individuals with ODS.^{4,7} Writing was also affected due to right hemiplegia. GUSS swallowing assessment revealed slight dysphagia with lower risk for aspiration. However, severe dysphagic features were reported in the earlier studies.^{4,7} This difference might be due to the extent of the neurological problem and site of lesion. In Coca et al. $(2017)^7$ study the patient's brain MRI disclosed diffusion restriction scanning in some gyrus of parietal, frontal occipital and temporal regions and also bilateral basal ganglia. In Yuridullah et al. (2018)⁴ study patient's brain MRI images disclosed classic hyperintense or areas of demyelination of the pons consistent with osmotic demyelination syndrome. In this study, The patient's MRI findings indicated hyperintensity in Bilateral fronto-parietal lobe, Thalamus, Cerebelleum. Bilateral caudate nucleus and lentiform nucleus appears to be bulky. Therefore, the assessment of speech, language and swallowing skills are very essential in patients with osmotic demyelination syndrome to plan the intervention.

3.2. Intervention

With swallowing therapy, the patient's swallowing skills improved and Ryles tube was removed as the patient was able to do oral feeding. With speech-language intervention, improvements were found in verbal communication. After eight speech-language therapy sessions, patient was able to speak in sentences but her speech showed features of dysarthria. On Perceptual Speech Intelligibility rating scale, the patient obtained four point which denoted "intelligible with careful listening although some words were unintelligible". A few authors have mentioned a complete neurological recovery⁸ where as others have reported that there was partial recovery presenting neurological conditions.^{9,10} In the case report by Coca et al. $(2017)^7$ the patient's swallowing skills were improved with intervention and the nasoentric tube was removed and there was persistent dysarthria in the patient which is similar to the present case report. Yuridullah et.al. (2018)⁴ reported

with speech therapy, improvement found in swallowing and speech skills in the patient who developed osmotic demyelination syndrome as result of severe hyponatremia over correction. Therefore, in this study as a result of speech-language intervention, patient had a better prognosis in swallowing and verbal communication skills.

4. Conclusion

ODS is a rare clinical condition which occurs frequently as a result of hyponatremia correction. In order to have a better perspective about speech, language and swallowing skills in individuals with ODS, replication of such studies are essential.

5. Acknowledgments

We wish to express our sincere thanks to Dr. Amulya P Rao for the constant support in this study.

6. Source of Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors

7. Conflicts of Interest

The authors declare that there are no conflicts of interest regarding the publication of this paper.

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Cite this article: Monish V, Sreedevi N. Speech and swallowing difficulties and rehabilitation in osmotic demyelination syndrome: A single case report. *IP J Otorhinolaryngol Allied Sci* 2021;4(4):149-152.