Case Report

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Oropharyngeal dysphagia in type I Chiari malformation: a clinical case series

Disfagia orofaríngea na malformação de Chiari tipo I: série de casos clínicos

Ramon Cipriano Pacheco de Araújo[1], *Ana Karoliny Pizate de Macedo*[2], *Cynthia Meira de Almeida Godoy*[3], *Juliana Fernandes Godoy*[4], *Hipólito Magalhães*[4]

ABSTRACT

Type I Chiari malformation (MCI) is a generally congenital condition of herniation of the cerebellar tonsils below the foramen magnum, which can lead to compression of structures close to the posterior cranial fossa or obstruction of the flow of ventral cerebrospinal fluid. Type I, although rare, is the most commonly found in the population. This study aimed to report the findings of video endoscopy of swallowing, speech assessment, level of oral intake, nutritional risk and therapeutic planning in three cases of Type I Chiari malformation. Data were collected regarding the assessment of mobility and strength of tongue, Maximum Phonation Time (MPT), cough efficiency and the Eichner Index. The level of oral intake and pharyngeal signs of dysphagia were analyzed in four food consistencies, according to the International Dysphagia Diet Standardization Initiative (IDDSI) classification, using swallowing video endoscopy. For analysis and classification of pharyngeal residues, the Yale Pharyngeal Residue Severity Rating Scale (YPRSRS) was used, while the Malnutrition Screening Tool (MST) was used to track nutritional risk. Reduced tongue mobility and strength and reduced MPT were observed, while pharyngeal signs varied between cases, with the presence of incomplete glottic closure, posterior oral escape, multiple swallows, pharyngeal residues and laryngeal penetration.

Keywords: Arnold-Chiari malformation; Deglutition disorders; Deglutition; Pharynx; Case reports

RESUMO

A malformação de Chiari tipo I é uma condição geralmente congênita de herniação das tonsilas cerebelares abaixo do forame magno, que pode levar à compressão das estruturas próximas da fossa craniana posterior ou à obstrução do fluxo do líquido cefalorraquidiano ventril. O tipo I, apesar de raro, é o mais comumente encontrado na população. Este estudo teve como objetivo relatar os achados da videoendoscopia da deglutição, avaliação fonoaudiológica, nível de ingestão oral, risco nutricional e o planejamento terapêutico em três casos de malformação de Chiari tipo I na fase adulta. Foram coletados dados com relação à avaliação fonoaudiológica de mobilidade e força de língua, Tempo Máximo de Fonação, eficiência da tosse e o Índice de Eichner. Foram analisados o nível de ingestão oral e os sinais faríngeos de disfagia em quatro consistências alimentares, de acordo com a classificação da *International Dysphagia Diet Standartisation Initiative*), por meio da videoendoscopia da deglutição. Para análise e classificação dos resíduos faríngeos, foi utilizado o *Yale Pharyngeal Residue Severity Rating Scale*, enquanto que, para rastrear o risco nutricional, foi utilizado o *Malnutrition Screening Tool*. Observou-se redução mobilidade e força de língua e no Tempo Máximo de Fonação, enquanto que os sinais faríngeos variaram entre os casos, com presença de fechamento glótico incompleto, escape oral posterior, deglutições múltiplas, resíduos faríngeos e penetração laríngea.

Palavras-chave: Malformação de Arnold-Chiari; Transtornos de deglutição; Deglutição; Faringe; Relatos de casos

Study carried out at Departamento de Fonoaudiologia, Universidade Federal do Rio Grande do Norte – UFRN – Natal (RN), Brasil.
[1]Programa Associado de Pós-graduação em Fonoaudiologia (Mestrado), Universidade Federal do Rio Grande do Norte – UFRN – Natal (RN), Brasil.
[2]Curso de Fonoaudiologia, Universidade Federal do Rio Grande do Norte – UFRN – Natal (RN), Brasil.
[3]Hospital Universitário Onofre Lopes, Universidade Federal do Rio Grande do Norte – UFRN – Natal (RN), Brasil.
[4]Departamento de Fonoaudiologia, Universidade Federal do Rio Grande do Norte – UFRN – Natal (RN), Brasil.
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Corresponding author: Ramon Cipriano Pacheco de Araújo. E-mail: ramon.pacheco.016@ufrn.edu.br
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INTRODUCTION

Arnold-Chiari malformations belong to a group of anomalies that involve the structures at the junction between the skull, cerebellum, and spinal cord, with different etiology, pathophysiology, and clinical manifestations, classified as types I, II, III, and later IV[1]. Chiari type I malformation (CIM) is a generally congenital condition of herniation of the cerebellar tonsils, with an excess of 3 mm to 5 mm below the foramen magnum, which can compress structures close to the posterior cranial fossa or obstruct the flow of ventral cerebrospinal fluid[2]. Type I, although rare, is the most found in the population[2].

The prevalence of CIM has been estimated at 0.8% to 1% in the adult population[2], which makes its evidence predominantly concentrated in retrospective studies and case reports. Its clinical manifestations begin insidiously, late, and progressively in adolescence or adulthood, even if it is possible to detect it in childhood by magnetic resonance imaging, and are accompanied by classic symptoms of headache after exertion, neck pain, weakness in the upper limbs, instability in the lower limbs with gait ataxia, oropharyngeal dysphagia, and less common symptoms, such as tinnitus and hoarseness[3,4].

Oropharyngeal dysphagia is the most important clinical condition among all symptoms due to the risk of death and its negative impact on the patient's quality of life[5]. The prevalence of dysphagia has been estimated at 36% to 47% of cases, depending on the assessment method used[5]. Some reports described cases in which dysphagia was the only manifested symptom of CIM in adulthood, which could be treated after surgical intervention to decompress the posterior cranial fossa. However, in other cases, this was not possible due to the long-term damage to cranial nerves[6]. When detected in childhood, there are manifestations of developmental delay, malnutrition, and recurrent aspiration pneumonia due to oropharyngeal dysfunction, with palate weakness and tongue atrophy[6,7].

Laryngotracheal food aspiration after swallowing is the most serious clinical manifestation in swallowing safety and can result in aspiration pneumonia, malnutrition, dehydration, prolonged hospitalizations, and death[4]. The pathophysiological mechanisms of dysphagia in these individuals are relative, and some studies suggest different severities of dysphagic manifestations for each patient with the diagnosis due to the angle of compression and the time to start treatment[5]. Thus, oropharyngeal dysphagia becomes an emerging symptom at some stage of the disease, persisting after surgical intervention[5,8]. This study aimed to report the findings of fiberoptic endoscopic evaluation of swallowing (FEES), speech-language-hearing (SLH) evaluation, oral intake level, nutritional risk, and therapeutic planning in three CIM cases in adulthood.

METHODOLOGY

This retrospective clinical case study collected data from medical records between 2021 and 2023. The research was carried out at the Otorhinolaryngology Outpatient Clinic of the Onofre Lopes University Hospital, in Natal (RN), Brazil. It met the ethical principles, and all participants or their guardians signed a standard informed consent form provided by the service before the exam procedures. The study was approved by the Research Ethics Committee of the Federal University of Rio Grande do Norte – CEP/UFRN, under evaluation report no. 6.169.294. The sample included three adults with a late diagnosis of CIM, treated at the service, and referred by other hospital departments due to clinical swallowing complaints. The exclusion criterion was the inability to carry out the commands. No individual had a history of orotracheal intubation, tracheostomy, head and neck neoplasia, or other neurological diagnoses. Data were collected regarding their history, previous SLH evaluation, FEES findings, oral intake level, and nutritional screening after the exam.

PROCEDURES

All participants complained of difficulty eating and were in the process of investigating oropharyngeal dysphagia. Their ages ranged from 46 to 66 years: case 1 was 46 years old, case 2 was 50 years old, and case 3 was 66 years old. The SLH evaluation was carried out before the instrumental examination when the patient was admitted to the outpatient clinic. This evaluation used the service's protocol, analyzing orofacial myofunctional aspects involved in swallowing, such as tongue mobility and strength, oral status, phonation, and coughing efficiency.

The oral status involved intraoral inspection of salivary stasis and assessment of the occlusal support distribution in the molar region, according to the Eichner Index (EI)[9]. This index was determined by the vertical contact components existing between bilateral molars and categorized into three types: class A, contact between four occlusal support zones; class B, contact between one and three occlusal support zones; and class C, no occlusal contact between molars. EI considered the usual occlusal support for chewing, using prosthetic oral rehabilitation, when available.

Tongue mobility and strength were subjective aspects assessed by the SLH pathologist. Patients were asked to perform tongue protrusion and lateralization movements and tongue protrusion against the resistance of a gloved finger. The normality criteria were correctly executing these commands and maintaining isometric strength against finger resistance. After showing an example, the evaluator asked patients to emit the vowel sound "a" with the maximum phonation time (MPT), in which the normality criteria were emissions of 14 s for women and 20 s for men. Then, the auditory-perceptual evaluation of voice was carried out, verifying whether roughness was present during the vowel emission. They were also asked to cough strongly and spontaneously to evaluate the subjective efficiency in producing a cough on command (efficient/weak) for eventual pharyngeal clearing. All changes were described and noted to continue the instrumental assessment of swallowing.

A resident physician performed the FEES accompanied by the head otorhinolaryngologist and an SLH pathologist with experience in oropharyngeal dysphagia, following the institution's protocol. A flexible fiberoptic rhinolaryngoscope manufactured by Olympus®, 3.2 mm in diameter, model LF-P, with a micro-camera and attached light source was introduced into the nasal cavity down to the hypopharynx. Patients were instructed to sit upright, and no topical anesthetic was used to introduce the instrument into the nasal cavity down to the hypopharynx.

After the physician carried out standard analyses of pharyngeal structures and sensitivity, the SLH pathologist offered foods artificially colored with blue aniline in different consistencies in the following order: level 2 (mildly thick liquid),

level 4 (extremely thick liquid), level 0 (thin liquid), and level 7 (regular solid food), according to the International Dysphagia Diet Standardization Initiative (IDDSI) classification[10]. Liquids were offered three times in 5-mL servings in a metal spoon, and the solid food was served in a single portion. The liquid was artificially flavored diet juice, thickened with an instant cornstarch food thickener, while the solid food was a single portion of 8-g crackers as requested.

The three previously mentioned professionals with experience in the examination were responsible for interpreting and evaluating simultaneously it, by consensus, and concluding whether there were pharyngeal signs of dysphagia, such as multiple swallowing, posterior oral leakage, pharyngeal residue in the valleculae and/ or pyriform sinuses, according to the Yale Pharyngeal Residue Severity Rating Scale (YPRSRS)[11] (1 - none; 2 - trace; 3 - mild; 4 - moderate; 5 - severe), laryngeal penetration, and laryngotracheal aspiration. The following parameters were considered for analysis, starting from the first serving: multiple swallows – i.e., two or more attempts to swallow the same serving[12]; posterior oral leakage, due to the presence of premature food leakage in the hypopharynx before triggering the swallowing reaction[12]; pharyngeal residue, by identifying the residual presence of colored food in the valleculae and/or pyriform sinuses after swallowing the first serving onwards[12]; laryngeal penetration, via observation of residual presence of colored food in the vocal folds[12]; and laryngotracheal aspiration when there was residue of colored food below the vocal folds[12]. All analyses were performed in real-time, and the images were stored on a computer at the outpatient clinic to be reviewed as many times as the professionals deemed necessary after carrying out the exam.

After the examination, the professionals assessed the oral intake level with the Functional Oral Intake Scale (FOIS) [13], based on the examination analysis and the existence and need for liquid thickening. Nutritional risk was assessed by a nutritionist, using the Malnutrition Screening Tool (MST)[14], in which values equal to or greater than 2 represent nutritional risk and the need for more detailed nutritional assessment.

CLINICAL CASE PRESENTATION

Case 1

Female patient, 46 years and 10 months old, breathing naturally, and with FOIS 7. She went to the hospital to investigate headache episodes and persistent neck pain. She was initially diagnosed with recurrent atlantoaxial subluxation with myelopathy. Two years later, she was followed up by the neurosurgery team for occipitocervical fixation and decompression operations, which resulted in the late diagnosis of CIM. Her postoperative period was accompanied by weakness in the upper limbs and complaints of choking when eating, leading to a referral for physiotherapeutic rehabilitation and the oropharyngeal dysphagia outpatient clinic. She was seen at the outpatient clinic 2 years after diagnosis. In the SLH evaluation, she presented EI class B, with missing teeth, no dentures, preserved tongue mobility and strength, absence of salivary stasis, inefficient spontaneous cough, and reduced MPT without roughness. The FEES showed preserved pharyngeal sensitivity to touch, moderate pharyngeal residues (YPRSRS 4) in the valleculae and pyriform sinuses, and laryngeal penetration, both with level 4 (extremely thick liquid). After the examination, she had FOIS 5 (total oral route with multiple consistencies, with the need for special preparation or compensations) and MST 2, which suggests nutritional risk and the need for detailed nutritional assessment. Therapeutic planning at the time of admission targeted swallowing safety, using maneuver strategies for voluntary readjustment of swallowing biomechanics with functional liquid training. The patient's SLH therapy was discontinued due to consecutive absences, and she subsequently continued to complain of dysphagia and hand weakness.

Case 2

Male patient, 50 years and 3 months old, breathing naturally, and with FOIS 7. He was diagnosed with CIM late, underwent occipitocervical decompression neurosurgery in the same year, and continued to complain of choking when eating and worsened hoarseness in the immediate postoperative period. He was referred by the neurology team 3 years after the surgical procedure, due to permanent swallowing and speaking complaints. He was seen at the outpatient clinic 5 years after diagnosis. The SLH evaluation verified EI class B, with missing teeth, no dentures, reduced tongue mobility and strength, absence of salivary stasis, efficient spontaneous cough, and reduced MPT with roughness and breathiness. The FEES verified preserved laryngeal sensitivity, oral residue after swallowing in all consistencies evaluated, pharyngeal traces (YPRSRS 2) in the valleculae, and multiple swallows, both with level 4 (extremely thick liquid). After the exam, he had FOIS 5 (total oral route with multiple consistencies, with the need for special preparation or compensations) and MST 0, with no nutritional risk. Therapeutic planning targeted orofacial myofunctional training and swallowing safety, with isometric and isotonic exercise strategies on the tongue, lips, and cheeks, and head posture strategies with functional liquid training. He remained in SLH therapy for 3 months, with clinical stability, with no coughing or choking complaints, until his therapy was discontinued due to consecutive absences. He was later monitored by the physiotherapy team.

Case 3

Female patient, 66 years and 5 months old, breathing naturally, and with FOIS 7. She was diagnosed with CIM late and underwent neurosurgery in the same year, with clinical stability and a good prognosis. She returned to the hospital after 5 years, with a progression of dysphonia and weakness in the lower limbs, which resulted in falls at home due to imbalance. During this period, the patient underwent physiotherapy sessions but did not seek SLH therapy. After the period without SLH therapy, she returned with tremors in her upper limbs, associated with complaints of choking on liquids. She was seen at the outpatient clinic 15 years after diagnosis. The SLH evaluation verified EI class A, using upper and lower partial dentures, reduced tongue mobility and strength, tongue tremor at rest, difficulty clicking the tongue, absence of salivary stasis, efficient spontaneous cough, reduced MPT with roughness and breathiness, and difficulty in swallowing saliva. The FEES verified preserved pharyngeal sensitivity, and posterior oral

leakage with levels 0 (thin liquid), 2 (mildly thick liquid), and 4 (extremely thick liquid). After the exam, she had FOIS 5 (total oral route with multiple consistencies, with the need for special preparation or compensations) and MST 0, with no nutritional risk. Therapeutic planning targeted orofacial myofunctional training and swallowing management, with isometric and isotonic exercise strategies on the tongue, lips, and cheeks, and compensatory interventions on food consistencies to increase swallowing safety. Her therapy was later discontinued due to consecutive absences, and she was monitored at the movement disorders outpatient clinic.

Comparative description of the assessment results and therapeutic planning

In the SLH evaluation, two cases had reduced tongue mobility and strength, and one had tongue tremors at rest. Regarding the oral status, two cases had EI class B, with significant tooth absences in the molar region, while one case used a well-fixed complete denture. All cases had reduced MPT, and one of them had a weak spontaneous cough to eventually clear the larynx. Regarding the FEES findings, two cases had incomplete glottic closure, and all cases had pharyngeal signs of dysphagia with liquids and thickened liquids, predominantly level 4 (extremely thick liquid). Moreover, the case with tongue tremors also had signs of posterior oral leakage with other liquid consistencies (levels 0, 2, and 4). No signs of dysphagia were observed with solid food (level 7) in any of the cases. The oral intake level for all individuals was FOIS 5, and one of them was at nutritional risk due to relevant weight loss. Table 1 presents the data regarding the SLH assessment and FEES findings.

The therapeutic planning for the cases at the time of their admission to the oropharyngeal dysphagia outpatient clinic focused on the oral and pharyngeal phases of swallowing, according to the assessment findings, mobility and tongue strength improvement, compensatory measures, strategies with head posture maneuvers and voluntary swallowing readjustments. The objectives of conventional swallowing therapy were used to improve the biomechanics of swallowing, such as strengthening the tongue muscles, increasing hyolaryngeal excursion movement, and decreasing pharyngeal residue in recesses (Chart 1).

DISCUSSION

The process through which clinical manifestations of dysphagia trigger in individuals with this malformation is still unclear, as swallowing difficulties affect those with the malformation in different ways[15]. However, it has been observed that lower cranial nerve paralysis due to compression could be associated with nerve traction and small sectioning inherent to the disease[3]. Therefore, this study aimed to report the FEES findings, SLH assessment, oral intake level, nutritional risk, and therapeutic planning of three cases of CIM in adulthood.

The cause of oropharyngeal dysphagia in this population is still being investigated. However, there is strong evidence that a larger cranial spinal angle is associated with the severity of late dysphagic manifestations in adults[2]. This is due to compression of the brainstem and cerebellum below the foramen magnum,

Table 1. Description of characteristics and findings of speech-language-hearing evaluation and fiberoptic endoscopic evaluation of swallowing between cases

Variables	Case 1	Case 2	Case 3
Age (years)	46	50	66
Sex	Female	Male	Female
Time since diagnosis (years)	2	5	15
Speech-language-hearing assessment findings			
Eichner Index	Class B	Class B	Class A
Tongue mobility	Adequate	Reduced	Reduced
Tongue strength	Adequate	Reduced	Reduced
Tongue tremor	Absent	Absent	Present
Salivary stasis	Absent	Absent	Absent
Maximum phonation time	Reduced	Reduced	Reduced
Roughness	Absent	Present	Present
Spontaneous cough	Weak	Efficient	Efficient
FEES findings (IDDSI)			
Laryngeal sensitivity	Preserved	Preserved	Preserved
Glottal closure	Complete	Incomplete	Incomplete
Multiple swallows	Absent	Level 4	Absent
Posterior oral leakage	Absent	Absent	Levels 0, 2, and 4
Pharyngeal residues	Level 4	Level 4	Absent
Severity of residues (YPRSRS)	Moderate (4)	Trace (2)	Absent (1)
Laryngeal penetration	Level 4	Absent	Absent
Laryngotracheal aspiration	Absent	Absent	Absent
FOIS	5	5	5
MST	2	0	0

Subtitle: FEES = fiberoptic endoscopic evaluation of swallowing; IDDSI = International Dysphagia Diet Standardization Initiative; YPRSRS = Yale Pharyngeal Residue Severity Rating Scale; FOIS = Functional Oral Intake Scale; MST = Malnutrition Screening Tool

Chart 1. Description of therapeutic planning with goals, objectives, and strategies for the cases at the time of their admission to the oropharyngeal dysphagia outpatient clinic

	Goals	Objectives	Strategies
Case 1	- Pharyngeal phase of swallowing.	- To reduce the accumulation of pharyngeal residues with thickened liquid.	- Supraglottic swallowing with functional training with extremely thick liquid (level 4).
		- To increase hyolaryngeal excursion movement.	- Effortful swallowing technique.
Case 2	- Oral preparatory phase, oral phase, and pharyngeal phase of swallowing.	- To improve tongue mobility and strength.	- Tongue protrusion and lateralization exercises.
		- To provide head postural adjustments to improve hyolaryngeal excursion.	- Tongue counter-resistance exercises with a gloved finger and spatula.
			- Tip of the tongue tapering exercises.
			- Chin down postural strategy with swallowing training with liquid and thickened liquid (levels 0, 2. and 4).
Case 3	- Oral preparatory phase, oral phase, and pharyngeal phase of swallowing.	- To improve tongue mobility and strength.	- Tongue protrusion and lateralization exercises.
		- To use compensatory swallowing measures.	- Tongue counter-resistance exercises with a gloved finger and spatula.
		- To provide head postural adjustments to improve hyolaryngeal excursion.	- Tip of the tongue tapering exercises.
			- Chin down postural strategy with swallowing training with liquid and thickened liquid (levels 0, 2. and 4).
			- Hold the liquid in the oral cavity for 5 seconds and then start swallowing.

increasing the tension in the lower part of the structures, and resulting in minor damage and paralysis of the cranial nerves, evidenced in magnetic resonance images[5]. The greater angle comprises the angular lowering of the brainstem in relation to the line of extension of the occipital slope and of the floor of the fourth ventricle[5].

Surgical intervention with occipitocervical decompression (or posterior cranial fossa decompression) is common among individuals with the pathology and is performed by the neurosurgery team after confirming the diagnosis[3]. As the cases reported in this study were diagnosed late in adulthood, the decompression intervention was carried out at a time of peak tension and intermittent acute clinical manifestations during the medical investigation process, resulting in a long period of traction and minor damage to the adjunct cranial nerves[6]. Furthermore, the surgical process only relieved the constant tension in the posterior region, characterized by debilitating headaches and cervical tension symptoms in daily life. However, as described in the reports, it did not resolve the dysphagic condition, as two cases reported worsened swallowing and hoarseness in the immediate postoperative period.

The SLH evaluation found that the two individuals with the longest diagnosis time had a reduction in tongue mobility and strength in the requested tests, which suggests changes in the oral phase of swallowing. The tongue is known to play a significant role in swallowing, participating in the control, transport, and retropulsion of the food bolus from the oral cavity to the pharynx[2]. Reduced mobility and strength can affect swallowing efficiency, verified with instrumental examination, such as the oral residue observed after serving all food consistencies in case 2 and the incoordinated transfer of the food bolus to the subsequent phase, with premature leakage, observed in the liquid servings in case 3. These findings are compatible with studies that found that the time of diagnosis is related to important oropharyngeal changes, such as tongue atrophy and hypofunction of the velopharyngeal sphincter, due to a decline in the hypoglossal motor neurons (XII), originating in the middle of the bulb[5].

Phonation and coughing were important aspects during the evaluation, as the cases had reduced MPT, with incomplete glottal closure, and roughness and breathiness in vocal emission. These data demonstrate that individuals with CIM have low resistance to glottal closure, which has a negative impact on the protection of the lower airways from eventual aspirations. Case 1 was the only one that had a weak cough, to the same extent that there was reduced MPT and laryngeal penetration in the instrumental examination. Thus, these data confirm the understanding of unsafe swallowing, with little reflex laryngeal protection and low capacity to eject material that has penetrated the vocal folds[2,6,7]. These findings help to understand the pathophysiology involved in these individuals since there is no published data on phonation and cough efficiency in CIM, important parameters for SLH planning.

Pharyngeal signs of dysphagia were assessed with FEES, serving four different food consistencies, in which the three cases showed changes in the pharyngeal phase of swallowing. Cases 1 and 2 had different degrees of pharyngeal residues after swallowing the same food consistency, while case 2 required multiple swallows to ingest and clear the entire volume. Case 1 had laryngeal penetration due to the severe accumulation of remaining material in recesses after swallowing, which demonstrates a decrease in the motor component of hyolaryngeal

elevation and anteriorization to properly open the upper esophageal sphincter. These findings were used as a basis for therapeutic planning to reduce pharyngeal residue and increase hyolingual elevation. Head posture strategies and voluntary swallowing adjustment maneuvers were used to increase safety and pharyngeal clearing.

Difficulties in drinking fluids in all research participants resembled typical difficulties in neurogenic dysphagia, such as patterns observed in individuals with amyotrophic lateral sclerosis and myasthenia gravis[2,5,12], with eventual risk of laryngotracheal aspiration and nutritional risk due to low oral nutrient intake in case 1.

Orofacial myofunctional training was used in therapeutic planning in cases 2 and 3, who had reduced tongue mobility and strength. Isometric and isotonic tongue exercises are used to expand and strengthen the tongue movements necessary to contain and propel food in the oral cavity[6]. Changes in tongue functioning are frequently reported in clinical cases of CIM, with some milder signs, while others are associated with spasms, atrophy, and tremors. No reports showed a decrease in intraoral sensitivity[3]. Weakness and decreased fine motor movements are constant in this population and should be addressed in SLH rehabilitation[14,15]. The movements of lifting the tip of the tongue, clicking, dragging the tip of the tongue on the palate, and isometric resistance should be prioritized in SLH therapy.

All cases in this study were diagnosed late in adulthood, which may have interfered with the variation of oropharyngeal dysphagia clinical manifestations, as there was evidence of differences in clinical manifestations between age groups[5]. Late diagnosis can lead to a greater risk of hospitalization due to little protection of the lower airways, as seen in the inefficient cough in case 1, which increases the chances of silent aspirations, a more critical condition in dysphagia[5,13,14]. Therefore, early diagnosis and SLH monitoring during pre- and post-surgical intervention are important to initiate effective action and ensure adequate nutritional status, with functional swallowing.

The limitations of this study include the few participants in the sample, as it is a less frequent pathology in the clinic where it was carried out, and the lack of comparative results after the SLH intervention, as the patients abandoned the rehabilitation process. The strengths of the research include SLH evaluation, instrumental evaluation of swallowing, and nutritional screening of research subjects, as well as the description of the therapeutic planning used. Also, this was probably the first study to describe MPT and cough efficiency.

FINAL COMMENTS

The cases had reduced MPT and tongue mobility and strength. Pharyngeal signs of swallowing varied between cases, with incomplete glottal closure, posterior oral leakage, multiple swallows, pharyngeal residue, and laryngeal penetration. Dysphagia decreased the level of oral intake in all cases. Therapeutic planning was based on compensatory measures, orofacial myofunctional exercises, voluntary muscle conditioning adjustments, and head posture maneuvers.

REFERENCES

1. Milhorat TH, Chou MW, Trinidad EM, Kula RW, Mandell M, Wolpert C, et al. Chiari I malformation redefined: clinical and radiographic findings for 364 symptomatic patients. Neurosurgery. 1999 Maio;44(5):1005-17. http://doi.org/10.1097/00006123-199905000-00042. PMid:10232534.
2. Almotairi FS, Andersson M, Andersson O, Skoglund T, Tisell M. Swallowing dysfunction in adult patients with Chiari I malformation. J Neurol Surg B Skull Base. 2018 Maio 25;79(6):606-13. http://doi.org/10.1055/s-0038-1655758. PMid:30456032.
3. Passias PG, Naessig S, Kapadia BH, Para A, Ahmad W, Pierce KE, et al. Timing to surgery of Chiari malformation type 1 affects complication types: an analysis of 13,812 patients. J Craniovertebr Junction Spine. 2020;11(3):232-6. http://doi.org/10.4103/jcvjs.JCVJS_67_20. PMid:33100774.
4. Campisi R, Ciancio N, Bivona L, Di Maria A, Maria GD. Type I Arnold-Chiari malformation with bronchiectasis, respiratory failure, and sleep disordered breathing: a case report. Multidiscip Respir Med. 2013 Fev 22;8(1):15. http://doi.org/10.1186/2049-6958-8-15. PMid:23433005.
5. Lu F, Chen Z, Wu H, Jian FZ. Magnetic resonance imaging of chiari malformation type i in adult patients with dysphagia. BioMed Res Int. 2019 Maio 14;2019:7485010. http://doi.org/10.1155/2019/7485010. PMid:31218227.
6. Graham K, Black A, Brain P. Resolution of life-threatening dysphagia caused by caudal occipital malformation syndrome following foramen magnum decompressive surgery. Aust Vet J. 2012 Jul 24;90(8):297-300. http://doi.org/10.1111/j.1751-0813.2012.00952.x. PMid:22827623.
7. Albert GW. Chiari malformation in children. Pediatr Clin North Am. 2021 Ago;68(4):783-92. http://doi.org/10.1016/j.pcl.2021.04.015. PMid:34247709.
8. Doval Rodríguez A, Serramito García R, Menéndez Cortezón B, Prieto González A. Chiari type I malformation discovered through a glossopharyngeal neuralgia. Neurocirugia. 2022;33(6):398-401. http://doi.org/10.1016/j.neucie.2022.02.008. PMid:35256327.
9. Eichner K. Renewed examination of the group classification of partially edentulous arches by Eichner and application advices for studies on morbidity statistics. Stomatol DDR. 1990;40(8):321-5. PMid:2270610.
10. Cichero JAY, Lam P, Steele CM, Hanson B, Chen J, Dantas RO, et al. Development of international terminology and definitions for texture-modified foods and thickened fluids used in dysphagia management: the IDDSI framework. Dysphagia. 2017 Dez 2;32(2):293-314. http://doi.org/10.1007/s00455-016-9758-y. PMid:27913916.
11. Neubauer PD, Rademaker AW, Leder SB. The Yale pharyngeal residue severity rating scale: an anatomically defined and image-based tool. Dysphagia. 2015 Jun 7;30(5):521-8. http://doi.org/10.1007/s00455-015-9631-4. PMid:26050238.
12. Daggett A, Logemann J, Rademaker A, Pauloski B. Laryngeal penetration during deglutition in normal subjects of various ages. Dysphagia. 2006 Jan 10;21(4):270-4. http://doi.org/10.1007/s00455-006-9051-6. PMid:17216388.
13. Crary MA, Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. Arch Phys Med Rehabil. 2005 Ago;86(8):1516-20. http://doi.org/10.1016/j.apmr.2004.11.049. PMid:16084801.

14. Ferguson M, Capra S, Bauer J, Banks M. Development of a valid and reliable malnutrition screening tool for adult acute hospital patients. Nutrition. 1999;15(6):458-64. http://doi.org/10.1016/S0899-9007(99)00084-2. PMid:10378201.

15. Urbizu A, Ferré Á, Poca MA, Rovira A, Sahuquillo J, Martin BA, et al. Cephalometric oropharynx and oral cavity analysis in Chiari malformation type I: a retrospective case-control study. J Neurosurg. 2017 Fev 1;126(2):626-33. http://doi.org/10.3171/2016.1.JNS151590. PMid:27153161.