Oropharyngeal dysphagia may be a consequence of necrotizing otitis externa: case report

Disfagia orofaríngea pode ser uma consequência da otite externa necrotizante: relato de caso

Disfagia orofaríngea puede ser una consecuencia del externa del otitis necrotizante: Presentación de un caso

Karen de Oliveira dos Passos^{*} Aline Gasparin Rabaioli^{**} Bárbara Luisa Simonetti^{**} Sheila Almeida^{**}

Abstract

Individuals with episodes of necrotizing otitis externa not treated properly can come to present changes not only in the external auditory canal, but also in other structures such as the cranial nerves. Taking into account these characteristics and the fact that the individual can manifest changes in swallowing biomechanics this study aims to report a case of a female patient of 55 years, accompanied by the ORL team and forwarded to the speech therapy team during her hospitalization complaining of difficulty in swallowing, in use of a nasogastric feeding tube, vocal and facial paralysis. She performed Videolaringoscopy and Videofluoroscopy of Deglutition with results compatible with the complaints. The patient had good adhesion to speech therapy and clinical treatment of necrotizing external otitis, received systematic speech therapy care with orofacial exercises for mobility and tone to facial expressions, vocal exercises for vocal fold vibration and glottal closure, as well as head postural maneuvers during the

**Universidade Federal de Ciências da Saúde de Porto Alegre - UFCSPA - Porto Alegre - RS - Brazil

Authors' contributions; KOP - data collection, interpretation and data analysis, organization of the scientific article. AGR - data collection, organization of the scientific article. BLS - organization of the scientific article and literature review / update of the subject matter. STA - Project management, writing and critical review of the study, final approval of the content to be published.

Work displayed as poster shape at the Congresso Brasileiro de Fonoaudiologia Hospitalar, from May 21 to 23, 2015, Rio de Janeiro.

Correspondence address: Sheila Almeida - sheilat@ufcspa.edu.br Received: 12/09/2016 Accepted: 28/11/2016



^{*}Hospital Universitário ULBRA Mãe de Deus - HU - Canoas - RS - Brazil

time of swallowing. This treatment was carried out 3 times a week totaling twelve attendances in her first hospitalization and eleven in the second, in a period of 4 months. After this period she was discharged with a prescribed diet for overall consistency orally without restriction. In this case, in particular, it was notable the impact on swallowing and the importance of early speech therapy and multidisciplinary approach to help a favorable prognosis of the patient.

Keywords: Deglutition Disorders; Otitis Externa; Facial Paralysis

Resumo

Indivíduos com episódios de otite externa necrotizante não tratada adequadamente podem vir a apresentar alterações não só no conduto auditivo externo, mas também em outras estruturas, como nos pares cranianos. Levando em consideração essas características e o fato de que o indivíduo pode manifestar alterações na biomecânica da deglutição, o presente estudo tem como objetivo relatar um caso clínico de uma paciente do sexo feminino de 55 anos, hipertensa e diabética, com hipertireoidismo e diagnóstico atual de otite externa necrotizante. Esta foi acompanhada pela equipe de Otorrinolaringologia e encaminhada para a equipe de Fonoaudiologia durante sua internação hospitalar com queixa de dificuldade de deglutição, em uso de sonda nasoenteral para alimentação, alteração vocal e paralisia facial. Realizou Videolaringoscopia e Videofluoroscopia da Deglutição com resultados compatíveis com as queixas. A paciente apresentou boa adesão à fonoterapia e ao tratamento clínico da otite externa necrotizante, recebia atendimentos sistemáticos de fonoterapia com exercícios orofaciais de mobilidade e tônus para mímica facial, exercícios vocais de vibração de pregas vocais e coaptação glótica, bem como manobras posturais de cabeça no momento da deglutição. Esse atendimento foi realizado três vezes por semana totalizando doze atendimentos em sua primeira internação e onze na segunda, em um período de quatro meses. Após esse período, recebeu alta hospitalar com alimentação prescrita por via oral total sem restrição de consistência. Nesse caso, em especial, foi notável o impacto na deglutição e a importância da atuação fonoaudiológica e multidisciplinar precoce como auxílio a um prognóstico favorável da paciente.

Palavras-chave: Transtornos de Deglutição; Otite Externa; Paralisia Facial

Resumen

Los individuos con episodios de otitis externa necrotizante no se trata adecuadamente puede llegar a presentar cambios no sólo en el conducto auditivo externo, sino también en otras estructuras como los nervios craneales. Teniendo en cuenta estas características y el hecho de que el individuo puede manifestar cambios en la biomecánica tragar este estudio tiene como objetivo presentar un caso de una paciente femenina de 55 años, acompañados por el equipo de ORL y remitidos al equipo terapia del habla durante su hospital quejándose de dificultad en la deglución, en el uso de una sonda de alimentación nasogástrica, parálisis vocal y facial. Celebrada videolarin- goscopia y fluoroscopia de deglución con resultados consistentes con las quejas. El paciente tuvo una buena adherencia a la terapia del habla y el tratamiento clínico de la otitis externa necrotizante, recibió terapia del habla sistemática cuidado con los ejercicios orofaciales para la movilidad y el tono de las expresiones faciales, ejercicios vocales de vibración vocal pliegue y cierre de la glotis, así como la cabeza de maniobras posturales en deglución tiempo. Este tratamiento se llevó a cabo 3 veces por semana por un total de doce asistencias en su primera hospitalización y once en el segundo, en un período de 4 meses. Después de este período fue dado de alta con una dieta prescrita para la coherencia global por vía oral sin restricción. En este caso, en particular, fue notable impacto en la deglución y la importancia de la terapia del habla temprana y enfoque multidisciplinario para ayudar a un pronóstico favorable del paciente.

Palabras clave: Trastornos de Deglución; Otitis Externa; Parálisis Facial



Introduction

Necrotizing external otitis (NEO) is an infection most frequently caused by Pseudomonas aeruginosa and when treated inadequately may affect the squamous epithelium of the auditory canal and adjacent areas of soft tissue, blood vessels, cartilage and bone. Elderly, diabetic, immunosuppressed and debilitated patients¹ are more susceptible to this infection.

Initially the infectious process is limited to the external auditory canal and then extends to the osteocartilaginous junction towards the temporal bone. The facial nerve is usually affected. The infection may progress toward the base of the skull and affect the glossopharyngeal, vagus and accessory nerves causing the "jugular foramen syndrome" and eventually affect the hypoglossal nerve².

As previously mentioned inflammatory processes of the middle ear are among the causes of peripheral facial paralysis, resulting from lesion of the VII cranial nerve and is the most common of the cranial nerve pair pathologies³.

Peripheral facial palsy (PFP) is characterized by temporary or non-temporary reduction of facial nerve function in the peripheral segment, resulting clinically in changes in facial mobility, salivary and lacrimal secretion, and facial sensitivity, among others. Treatment of PFP is specific for each etiology, but other measures may be necessary, regardless of cause. Speech therapy aims to restore and prevent changes in chewing, swallowing, speech and facial expression.⁴

Methods

Clinical case presentation

A 55 year-old female patient with a history of diabetes, systemic arterial hypertension, hyperthyroidism and NEO complicated by jugular foramen syndrome, due to the evolution of the condition requiring two hospitalizations, also accompanied by a Otorhinolaryngology team.

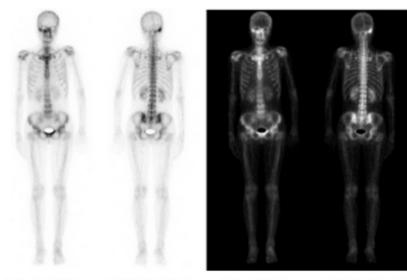
The patient underwent diagnostic exams such as an ear biopsy, in which squamous mucosa was visualized with chronic purulent inflammation. The Skeletal Scintigraphy (Figure 1) identified increased uptake in projections of the right parietal, temporal, and sphenoid bones. Late images showed an intense increase in activity at these sites. From the examination (Figure 1) the findings on the skull were suggestive of an active inflammatory process. After Gallium Scintigraphy, radiopharmaceutical uptake in the temporal bone projection, petrous and mastoid portions, and increased activity in soft tissues of the cervical region near the right side of the base of the skull were found. Correlating these findings with the bone scintigraphy, an inflammatory process was observed, possibly infectious, in the right temporal area, with extension to adjacent soft tissues.

The patient was referred to the Speech Therapy department during hospitalization, with symptoms of difficulty in swallowing with the use of nasoenteral tube for feeding, vocal alteration and facial paralysis in treatment with antibiotic therapy. The patient was evaluated by the team, with request for permission to use data collected for a research project approved by the Research Ethics Committee of the hospital under registration number CAAE: 01453912.4.0000.5335.

In the Sensory Motor-Orofacial evaluation, the following findings were observed; altered lip posture and mobility, altered tongue mobility and strength with preserved sensitivity, preserved facial sensitivity, altered and restricted jaw lateralization, saliva deglutition present, facial paralysis to the right, reduced maximum phonation times and hoarse and breathy voice quality.

In the clinical evaluation of swallowing in which pasty consistency was tested the following findings were observed: difficulty in ejection of food bolus, oral stasis of food and elevation alteration and anteriorization of the hyolaryngeal complex. In relation to clinical signs of penetration and aspiration wet voice, cough and throat clearing were observed. A clinical diagnosis of severe Oropharyngeal Dysphagia was concluded using the Gugging Swallowing Screening (GUSS) scale⁵, with subsequent restriction of any oral diet as well as solicitation of otorhinolaryngological evaluation and imaging examinations. In the videolaryngoscopy, findings compatible with vocal fold paralysis (VF) were found in adduction, with an important stasis of saliva in the right piriform sinus, in addition to cleft phonation. In the Videofluoroscopic Swallowing examination, the pasty and liquid consistencies were tested with barium sulfate contrast. Oral phase alteration was observed





ANTERIOR POSTERIOR ANTERIOR POSTERIOR

Figure 1. Bone scintigraphy of the presented case. Observed in the examination: increase uptake in the projections of the right parietal, temporal and sphenoid bones. Late images (right) show an intense increase in activity at the previously described sites.

due to inadequacy in the preparation, organization and ejection of the bolus, with premature leakage of food to the oropharynx in all consistencies; In the pharyngeal phase food stasis in the valleculae and piriform recess, in greater volume on the right side, decrease in the hyolaryngeal elevation, anteriorization and stabilization and presence of tracheal aspiration with the consistencies tested, due to pharyngeal food stasis was noted. The patient presented coughing with ejection of food bolus as she could not swallow, concluding the examination with diagnosis of Severe Oropharyngeal Dysphagia.

The patient started speech therapy with adequate treatment adherence. Orofacial exercises of mobility and tonus for facial expression and vocal exercises with VC (vocal cord) vibration and glottal closure were planned; Postural head maneuvers - protection maneuver: head facing the paralyzed side of the face at the moment of swallowing - and voluntary - efficiency maneuver: swallowing with effort, were also conducted. Improvement was noted in the aspects of orofacial mobility, lip closure, laryngeal elevation, progression in the ability to tolerate various food consistencies and improvement in vocal patterns. Systematic sessions of speech therapy were performed three times a week, totaling twelve visits in the first hospitalization and eleven in the second, in a period of 4 months. After this period, the patient was discharged with prescribed total oral feeding without restriction of consistency and clinical phono audiological diagnosis of Functional Deglutition, in addition to a new objective Videofluoroscopic Swallowing Exam, in which the pasty, semi-solid and liquid consistencies were tested. She presented alteration of the oral phase with premature posterior leakage of the liquid consistency and absence of pharyngeal phase changes (stasis / aspiration).

Discussion

The precise pathogenesis of necrotizing external otitis (NEO) is unknown. It most likely depends on the combination of two factors, including the underlying changes of the immune system and the specific characteristics of the bacterium that may find in that environment the ideal conditions to exert its pathogenic activity. Some cases of NEO are related to other states of immunosuppression such as hematological diseases (leukemia and severe anemia), treatment with cytotoxic or immunosuppressive drugs, AIDS and HIV-positive patients2.

In the literature review, we found other case reports in which patients developed external otitis



(EO) most likely as a result of poorly controlled type II diabetes mellitus ⁶.

Pseudomonas aeruginosa is a common cause of EO in diabetic patients and, despite local and systemic antibiotic therapy for EO, infection spreads from mastoid air cells to the base of the skull where it causes cranial nerve paralysis ⁷.

In the study by Climans et al (2013) there is a case report of a man with malignant external otitis and progressive dysphagia for solids and liquids for three weeks, accompanied by sporadic regurgitation and vomiting of partially digested foods8. He presented common comorbidities to the subject of the present study with both having type II diabetes mellitus and hypertension. As a common clinical complication, dysphagia with risk of aspiration is described for all food consistencies, requiring the use of an alternative means of feeding. Despite the similarity of the case reported with that of the present study, there was no speech therapy intervention in the cited study. In the case of Climans et al (2013) drug treatment was necessary for the subsequent regression of the symptoms and return to oral feeding occurring only after treatment⁸.

In the literature it is documented that in cases of NEO there is a possibility that the infection can affect the facial nerve with progression to the base of the skull affecting other cranial pairs responsible for facial expression, phonation and deglutition, with all these changes expected and consistent with those found in the case².

The most common symptom (90% of cases) is a painful, analgesic-resistant otalgia that radiates to the frontal-temporal and parietal regions worsening at night. In 45% to 100% of the cases it is accompanied by a fetid and purulent otorrhea. Sensitivity and peri-auricular edema are also common findings².

There is a relationship between broncho-aspiration and laryngeal and / or vocal fold paralysis, with failure of deglutition biomechanics and airway protection being observed in such cases^{9, 10}.

In this case, we observed a complex situation in which the favorable prognosis was dependent on the good adherence to speech therapy and the clinical treatment of NEO^{2, 8}.

Conclusion

Clinical complications related to external necrotizing otitis are well described in the literature. However, the relationship between these complications and the changes in deglutition biomechanics are still poorly reported. In this case, the severity of the repercussion in terms of deglutition and the importance of the early intervention with speech therapy and multidisciplinary assistance were noteworthy as an aid to a favorable prognosis of the patient in question.

References

1. Vieira ABC, Mancini P, Gonçalves DU. Doenças Infecciosas e perda auditiva. Rev Med Minas Gerais. 2010; 20(1): 102-6.

2. Antunes A, Macedo GRM, Leão RS, Caldas NCR. Otite externa necrotizante com paralisia facial periférica bilateral: relato de caso e revisão da literatura. Rev. Bras. Otorrinolaringol. 2004; 70(1): 137- 42.

3. Atolini Jr N, Jorge Jr JJ, Gignon VF, Kitice AT, Prado LSA, Santos VGW. Paralisia Facial Periférica: Incidência das Várias Etiologias num Ambulatório de Atendimento Terciário. Arq. Int. Otorrinolaringol. 2009; 13(2): 167-71.

4. Dib GC, Kosugi EM. Como Diagnosticar e Tratar Paralisia facial periférica. Moreira Jr Editora. 2003; 61(3): 110-7.

5. Trapl M, Enderle P, Nowotny M, Teuschl Y, Matz K, Dachenhausen A, Brainin M. Dysphagia bedside screening for acute-stroke patients The Gugging Swallowing Screen. Stroke. 2007; 38(11): 2948-52.

 Fonseca AS, Andrade NA, Neto MLA, Santos VMM. Paralisia bilateral do hipoglosso na evolução de otite externa necrotizante. Rev Bras Otorrinolaringol. 2007; 73(4): 576-76.

7. Sibai TA, Ben-Galim PJ, Eicher SA, Reitman CA. Infectious Collet-Sicard syndrome in the differential diagnosis of cerebrovascular accident: a case of head-to-neck dissociation with skull-based osteomyelitis. Spine J. 2009; 9(4): 6-10.

8. Handley TPB, Miah MS, Majumdar S, Hussain SSM. Collet-Sicard syndrome from thrombosis of the sigmoidjugular complex: A case report and review of the literature. Int J Otolaryngol. 2010; 2010:1-5.

9. Leder SB, SUiter DM, Duffey D, et al. Vocal Fold Immobility and Aspiration Status: A Direct Replication Study. Dysphagia. 2012; 27(2): 265-70.

10. Onofri SMM, Cola PC, Berti LC, et al. Correlation Between Laryngeal Sensitivity and Penetration/Aspiration After Stroke. Dysphagia. 2014; 29(2): 256-61.

