

Neurological disorder in celiac patient after third molars extraction: is there a relation?

Distúrbio neurológico em paciente celíaco após extração de terceiros molares: existe relação?

Trastorno neurológico en un paciente celíaco después de la extracción de terceros molares: ¿hay alguna relación?

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Abstract

Celiac disease is an immune-mediated gluten-dependent systemic disorder characterized by a specific profile associated with small intestinal lesion. Some of the classical symptoms observed in patients with this condition are nutrient malabsorption, diarrhea, abdominal pain, fatigue and, more recently, neurological symptoms were associated with the disease. Therefore, the objective is to inform an unusual case of a patient with celiac disease that developed severe permanent paresthesia in the superior and inferior gingiva, tongue and palate after tooth extraction e to discuss the relation between both things.

Descriptors: Nervous System Disease; Celiac Disease; Molar, Third.

Resumo

A doença celíaca é um distúrbio sistêmico dependente de glúten imune mediado, caracterizado por um perfil específico associado à lesão do intestino delgado. Alguns dos sintomas clássicos observados em pacientes com essa condição são: má absorção de nutrientes, diarreia, dor abdominal, fadiga e, mais recentemente, sintomas neurológicos foram associados à doença. Portanto, o objetivo é informar um caso incomum de um paciente com doença celíaca que desenvolveu parestesia permanente grave na gengiva superior e inferior, língua e palato após a extração dentária e discutir a relação entre as duas coisas.

Descritores: Doenças do Sistema Nervoso; Doença Celíaca; Dente Serotino.

Resumen

La enfermedad celíaca es un trastorno sistémico dependiente del gluten mediado por el sistema inmunitario caracterizado por un perfil específico asociado con la lesión del intestino delgado. Algunos de los síntomas clásicos observados en pacientes con esta afección son malabsorción de nutrientes, diarrea, dolor abdominal, fatiga y, más recientemente, los síntomas neurológicos se asociaron con la enfermedad. Por lo tanto, el objetivo es informar un caso inusual de un paciente con enfermedad celíaca que desarrolló parestesias permanentes severas en la encía superior e inferior, la lengua y el paladar después de la extracción del diente e discutir la relación entre ambas cosas.

Descriptores: Enfermedades del Sistema Nervioso; Enfermedad Celíaca; Tercer. Molar.

INTRODUCTION

Celiac disease is an immune-mediated gluten-dependent systemic disorder associated with a wide range of symptoms, mainly gastrointestinal¹. Some authors also defend the association between celiac disease and neurological disorders². Concerning oral manifestation, an association with a delay in dental eruption, decreased salivary flow, recurrent stomatitis, angular cheilitis and dental enamel defects in both dentitions have been associated³. The prevalence of celiac disease in the healthy population is at least 1%⁴ and there are no accurate estimates of the prevalence of neurological manifestations, although some studies reveal a range between 10 and 22.5%⁵. Therefore, the objective is to inform a case of a patient with celiac disease that developed severe permanent paresthesia in the superior and inferior gingiva, tongue and palate after tooth extraction.

CLINICAL CASE

Female patient, 33 years old, was referred to the Department of Oral and Maxillofacial Surgery at Erasto Gaertner Hospital needing the extraction of her third molars due to recurrent episodes of pericoronitis. The patient reported that she had a late diagnosis of celiac disease at 30 years old, confirmed by an intestine biopsy and presence of circulating antibodies, and at the present moment she was on a

gluten-free diet. The patient related other medical conditions such as osteoporosis, epilepsy, depression, fibromyalgia, esophagitis, and lactose intolerance, treated with medication. An intraoral examination revealed the presence of permanent dentition with the lack of some teeth and a severe enamel defect, with brown and brittle teeth (Figure 1).

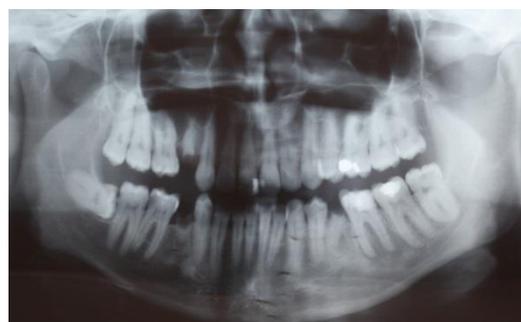


Figure 1: Panoramic X-ray showing tooth 38 in a vertical position, near the area of the mandibular channel, and tooth 48 in a horizontal position.

Teeth 18, 28, 38 and 48 were present, the last two being partially covered by gingival tissue. X-rays showed tooth 38 in a vertical position, near the area of the mandibular channel, and tooth 48 in a horizontal position (Figure 1). The extraction of the 4 teeth was performed with no intercurrents, but later the patient developed paresthesia on the right side, in the region innervated by the posterior superior alveolar nerve, major palatine nerve, lingual nerve, inferior

alveolar nerve and buccal nerve. Treatment with laser therapy and physical therapy was proposed without success. A MRI was done to investigate brain tumour, but no alterations were found (Figure 2). Laboratory exams ruled out the possibility of malnutrition or a vitamin deficiency. Another possibility considered was neurotransmitters release dysfunction as cause of the paresthesia, but both dopamine and serotonin exams were within normal limits. One year after the surgery, the patient continues the follow up and still has paresthesia and cannot distinguish temperatures or textures on the affected site, even when poked with needles.

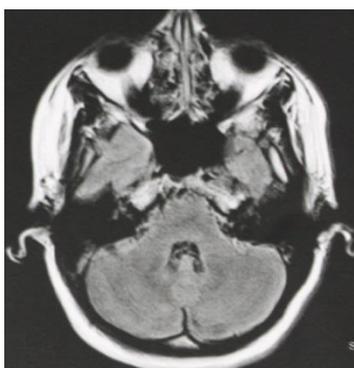


Figure 2: MRI scan in an axial view showing normality.

DISCUSSION

The estimated prevalence of persistent paresthesia (lasting at least 6-9 months) after third molar extraction ranges from 0 to 0,4%, a very small percentage⁶. When there is a visual resection of the nerve, it is possible to consider the surgical repair⁶, which was not indicated in the case reported in this paper, since the nerves were not exposed during surgery. No literature about paresthesia of palatal and posterior-superior alveolar nerve was found. The current understanding of the pathogenesis of neurologic findings in patients with celiac disease is limited⁷. Nutritional deficiencies may play a role in the development of neurological deficits, especially in those patients with a proved malabsorption of nutrients such as vitamin B-12, vitamin E, calcium and folic acid and that does not adhere to the correct diet⁸. The patient in the case presented had a late diagnosis of celiac disease, meaning that she lived a long period with implications of the systemic disorder. She presented other conditions that might have been potentialized by long term gluten intolerance, even though, she currently follow a gluten-free diet and has normal laboratory exams, without the lack of the nutrients mentioned above. A study by Chin et al.⁹ with a sample of 20 patients with neuropathy and biopsy-confirmed celiac disease found that all of the patients experienced burning, tingling, sensory loss and numbness in their distal extremities and 45% had diffuse paresthesias involving the face, trunk and lumbosacral region. Alaedini et al¹⁰ tested 27 patients looking for the

presence of antiganglioside antibodies, which are associated with autoimmune neuropathies. Neurological examinations in the six patients who tested positive revealed the presence of distal sensory loss, consistent with the diagnosis of peripheral neuropathy. Clinical tests showed that the same six had numbness or paresthesias with sensory loss in their hands and feet. Three of them suffered from neuropathic pain. Electromyography and nerve conduction studies showed discrete abnormalities in two of them, corresponding to what had been previously described by Oh et al.¹¹ as a possibility. In one of the patients, who was prescribed a nerve biopsy, chronic axonopathy was observed. None of the patients were found to have a nutritional deficiency or other possible causes of neuropathy.

CONCLUSION

Therefore, based on the available literature, the accurate incidence of neuropathy in celiac disease remains unclear and this diagnosis may be neglected if the patients are not properly and fully evaluated. Thereby, patients with celiac disease that present neurological symptoms should receive special attention to avoid further complications. The professional should be aware of this possible outcome when deciding for an oral surgery.

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CONFLICTS OF INTERESTS

The authors declare no conflicts of interests.

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