

Submandibular Tumefaction with Unexpected Diagnosis

Tumefação Submandibular com Diagnóstico Inesperado

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Abstract

Objective: Reporting the case of a 68-year-old patient, referred for evaluation of a possible odontogenic tumor. The patient had a submandibular nodule with 2-year evolution, approximately 2 cm, hardened, mobile and asymptomatic in the right submandibular region. After careful clinical examination and panoramic radiography analysis, the presumptive diagnosis was sialolith. Then, surgical removal was performed without complications, and, after histopathological analysis, the clinical diagnosis was confirmed. **Conclusion:** Clinical, radiographic, and histological evaluations are always a challenge for the correct diagnosis of hard lesions in the oral cavity.

Descriptors: Medical History Taking; Salivary Gland Calculi; Oral Diagnosis.

Resumo

Objetivo: Relatar o caso de uma paciente de 68 anos, encaminhada para avaliação de possível tumor odontogênico. A paciente apresentava nódulo submandibular com 2 anos de evolução, cerca de 2 cm, endurecido, móvel e assintomático em região submandibular direita. Após cuidadoso exame clínico e análise de radiografia panorâmica, o diagnóstico presuntivo foi de sialólito. Em seguida, a remoção cirúrgica foi realizada sem complicações e, após análise histopatológica, o diagnóstico clínico foi confirmado. **Conclusão:** As avaliações clínicas, radiográficas e histológicas são sempre um desafio para o correto diagnóstico de lesões duras em cavidade oral.

Descritores: Anamnese; Cálculos das Glândulas Salivares; Diagnóstico Bucal.

Resumen

Objetivo: Informar el caso de un paciente de 68 años remitido para evaluación de posible tumor odontogénico. El paciente presentaba un nódulo submandibular de 2 años de evolución, de unos 2 cm, endurecido, móvil y asintomático en región submandibular derecha. Después de un cuidadoso examen clínico y análisis de radiografía panorámica, el diagnóstico presuntivo fue sialolito. A continuación, se realizó la extirpación quirúrgica sin complicaciones y, tras el análisis histopatológico, se confirmó el diagnóstico clínico. **Conclusión:** Las evaluaciones clínicas, radiográficas e histológicas son siempre un desafío para el correcto diagnóstico de lesiones duras en la cavidad bucal.

Descritores: Anamnesis; Cálculos de las Glándulas Salivales; Diagnóstico Bucal.

INTRODUCTION

Salivary lithiasis is a common condition, characterized by excretory duct obstruction of salivary glands by a calculus, which is called sialolith¹. Clinical manifestations are increased local volume, pain, infection of the affected area, resulting in salivary ectasia and subsequent glandular dilation². The most affected gland is the submandibular, followed by the parotid, sublingual and, more rarely, minor salivary glands³. It can occur at any age, although rare in children.

The size of the calculus varies widely, from less than 1 mm to a few centimeters in length. The vast majority of sialoliths have dimensions less than 10 mm, lengths greater than 15 mm are rare and, when such condition is seen, it is called giant sialolith². Duct obstruction can have several causes, such as direct trauma, gland compression, radiation, Sjögren Syndrome, sialoadenitis and tumors

such as granular cell and cystic adenoid carcinoma⁴.

Sialoliths located in the anterior two-thirds of Wharton's duct are easily palpated and radiographic examination such as the panoramic can assist in the diagnosis¹, but the final diagnosis is only confirmed with histopathological examination⁵.

Treatment varies according to the affected gland, the size and location of the calculus. When small, calculi can be removed by extirpation, and when they are larger, surgical removal is necessary, and sialoadenectomy can even be performed⁶.

The purpose of this article is to report a case of a patient referred with a diagnostic hypothesis of a tumor in the submandibular region, and to emphasize the importance of careful clinical examination, radiography, and histopathological examination in the correct final diagnosis.

CLINICAL CASE

Male patient, 68 years old, referred for evaluation of submandibular lesion with presumptive diagnosis of odontogenic tumor. He complained of a “foreign element that appeared in my mouth” 2 years ago with a nodule in the left submandibular region. The nodule was hard, presenting symptoms associated with episodes of inflammation that led to an increase in submandibular volume and pain. On extraoral physical examination, a nodule in the left submandibular region was observed, hardened and asymptomatic on palpation. On intraoral physical examination, an orifice and a yellowish nodule were observed in the left mouth floor, in the molar region, hardened, mobile and painless on bidigital palpation (Figure 1).



Figure 1: Orifice and a yellowish nodule observed in the left mouth floor.

Panoramic radiography was performed, in which a radiopaque image with a radiolucent halo delimited at the angle of the mandible on the left side was observed (Figure 2).



Figure 2: Radiopaque image with a radiolucent halo delimited at the angle of the mandible on the left side

The presumptive diagnosis was sialolith. The dental procedure consisted of surgical removal of the nodule, under local anesthetic infiltration with vasoconstrictor (Mepivacaine 2%), leaving the surgical site bloody, uneventful, and, at the end, the patient received postoperative care guidelines (Figure 3).



Figure 3: Surgical site, uneventful.

Chlorhexidine 0.12% aqueous mouthwash was prescribed every 8 hours. The removed material (Figure 4) was sent for histopathological analysis where it showed basophilic layers permeated by other eosinophil layers, denoting alternating deposits of mineral and a disorganized matrix (Figures 5 and 6). Therefore, the clinical, radiographic, and histopathological examination confirmed the diagnosis of sialolith. In the 7 and 15-day postoperative consultation, he had no complaints or signs of infection and was discharged.

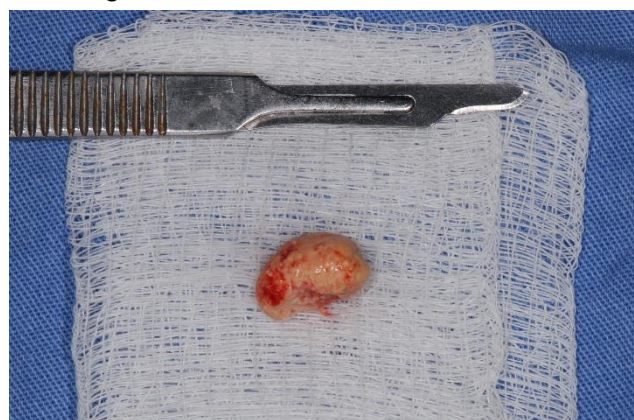


Figure 4: Removed material.

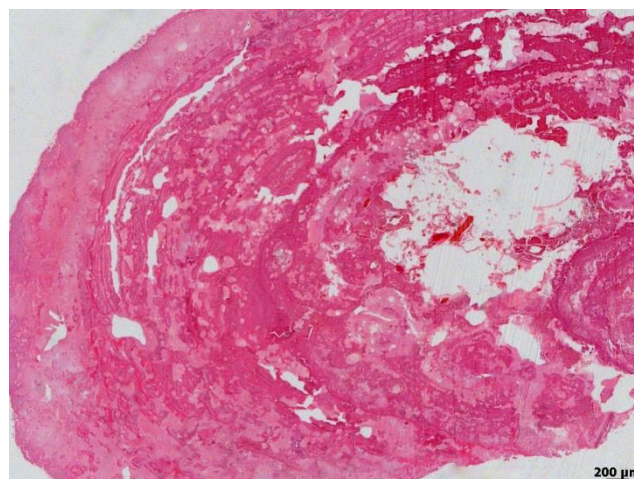


Figure 5: Basophilic layers permeated by other eosinophil layers, denoting alternating deposits of mineral and a disorganized matrix.

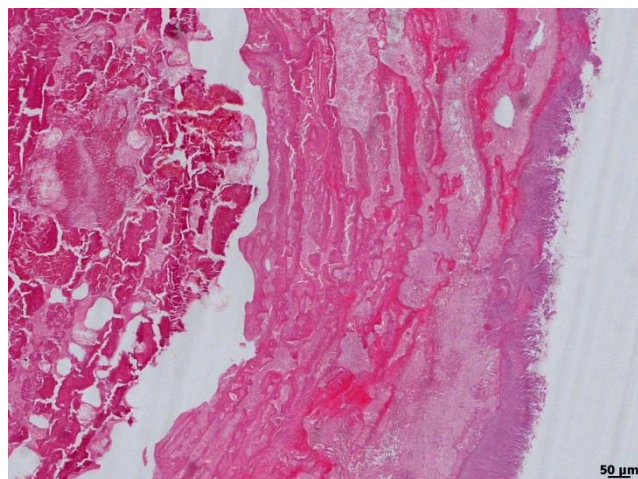


Figure 6: Basophilic layers permeated by other eosinophil layers, denoting alternating deposits of mineral and a disorganized matrix.

DISCUSSION

Sialolithiasis is the major cause of obstructive diseases of the salivary glands, being involved in approximately two thirds of cases⁷.

The submandibular gland is the region of most common occurrence as presented in this case, and there are at least five interconnected factors that explain this high frequency: (1) the Wharton duct has a long and winding path; (2) saliva drainage in the submandibular gland occurs against gravity; (3) salivary secretion has a more alkaline pH than that produced in the parotid; (4) it is a gland with predominantly mucous secretion and (5) phosphate and calcium rates in the submandibular saliva are higher than in the other glands⁸.

Sialoliths in submandibular glands can vary from less than 1 mm to several centimeters, with a case of sialolith removal up to 7.2 cm in length being reported in the literature⁹. The case reported here refers to the surgical removal of a sialolith measuring 2 cm in length, being classified as a giant sialolith, a name given to cases measuring more than 15 mm^{10,11}.

It is a condition that can affect any age group, but its incidence is higher in young and middle-aged adults^{12,13} as seen in this case. Inflammation and pain occur in the presence of benign tumors of the salivary glands as in sialoliths and can confuse the clinician in his diagnosis¹⁴.

A more accurate diagnosis is obtained by clinical and imaging tests association such as sialography, computed tomography, magnetic resonance imaging, scintigraphy; endoscopy is the most used radiographic examination¹⁵.

The patient's anamnesis, associated with careful inspection and palpation of the anatomical structures together with the

radiographic examination made it possible to observe the location, shape and approximate size of the calculus. That information associated with the symptoms reported by the patient, such as a considerable increase in volume, absence of pain and difficulty in eating sometimes did not require other tests. Although MRI and CT are very safe methods for diagnosis, in this case it was not necessary, being more indicated in cases that cannot be diagnosed by other methods.

The exact etiopathogenesis of sialolith remains uncertain, although the literature presents a wide variety of hypotheses. It is known that its occurrence results from the deposition of calcium phosphate around an initial organic matrix consisting of altered salivary mucins, bacteria, and desquamated epithelial cells. In the formation of calculus, intermittent stasis produces a change in the mucoid elements of saliva, which forms a gel. This gel reproduces an ideal environment for deposition of salts and organic substances creating sialolith. Traditional theories suggest that the process occurs in two phases: the formation of a central core and, later, the genesis of the layered periphery. The central core is formed by the precipitation of salts, which are bound by organic substances and, once formed, will serve as a base for the deposition of organic and inorganic material that, compacted, will form the layered periphery¹⁶.

Regarding treatment, a minimally invasive procedure is of utmost importance in preserving the function of the gland^{2,17}. The literature indicates some surgical procedures such as transoral sialolithotomy, sialendoscopy, shock with extracorporeal lithotripsy and resection of the gland^{12,18} being the most performed intraoral approach, as in this clinical case¹⁹.

There are three conditions that require resection of the submandibular gland: when sialoadenitis is recurrent, when the calculi are under the mylohyoid muscle and when there is a suspicion of an associated tumor¹⁴. In the reported case, the calculus was in the portion of the duct where it was possible to feel it on palpation and, despite insertion into the tissues, it was successfully removed by minimally invasive intraoral intervention, without damage to the anatomical structures, maintaining the gland and enabling a more comfortable post-operative for the patient.

Among the possible postoperative complications of the intraoral approach are injury

to the lingual nerve and Wharton's duct stenosis. During excision of the gland, there is a risk above 8% of permanent or temporary paralysis of the mandibular marginal nerve¹². There are only a few studies on the correlation between risk factors and specific features of the calculus, with proximal sialoliths causing more complications than distal sialoliths (Dong, S.H et al 2018). In this case, after 15 days postoperatively, there was no paresthesia of the lingual nerve and painful symptoms, with complete healing of the mucosa of the mouth floor, without complications.

CONCLUSION

The diagnosis of tumor in the submandibular region is a challenge, therefore, meticulous clinical and radiographic examination, in addition to knowledge of the anatomy and diseases that can affect the mouth floor and submandibular region leads to more reliable diagnostic hypotheses. The final diagnosis is always determined by histopathological examination.

ACKNOWLEDGEMENT

This study was financed in part by the Coordenação de Aperfeiçoamento de Pessoal de Nível Superior - Brasil (CAPES) - Finance Code 001.

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CONFLICTS OF INTERESTS

The authors declare no conflicts of interests.

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Received 03/11/2021

Accepted 04/01/2022