Sialolithiasis of minor salivary gland in labial mucosa: a clinical finding

Sialolitíase de glândula salivar menor na mucosa labial: um achado clínico

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Abstract

Introduction: sialolithiasis is the most common form of obstructive sialadenitis caused by a mixture of different calcium phosphates and an organic matrix. It is one of the most common salivary gland diseases, often attributed to the submandibular gland, with no relation to age or gender. However, it is rarely reported in the minor salivary glands. Objective: the present study aims to report auncommon clinical finding case of a sialolithiasisof minor salivary gland in labial mucosa. Case report: a 43-year-old female patient presented with a single, yellow and asymptomatic nodule in the labial mucosa at clinical examination. The clinical hypotheses were lipoma and fibrous hyperplasia. The lesion was biopsied, and the histopathological analysis showed a mineralized tissue. The final diagnosis was sialolithiasis and the patient remained under follow-up (8 months) without relapse. Conclusion: this case shows that sialolithiasis should be included in the diagnostic hypotheses when occur in a minor salivary glands area and emphasizes the importance of a complete clinical examination since it was not complaint of the patient.

Keywords: Sialolith. Sialadenitis. Minor salivary glands.

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Introduction

Sialolithiasis or salivary gland calculus is a pathology that involves the formation of calculi, or sialoliths, within the salivary ducts or glandular parenchyma which can cause pain, inflammation, discomfort and blockage of saliva[1-4]. It is a common salivary disease, reported as the main cause of the described obstructive sialadenitis, occurring mostly in the major salivary glands (SGs), such as the submandibular glands (80 to 92%) and parotid glands (16 to 19%)[4-6].

However, sialolithiasis of the sublingual gland and minor salivary glands (MSGs) is rare (less than 2% of cases)[7,8] andmanifests clinically like a solitary (multiple sialolithsare rare), asymptomatic and small submucosal nodule with hard/firm consistency. There is no predilection for sex and may occur at any age with aincidence peak at 50 years old according to the epidemiological studies[2,8,9].

Anatomically, the MSGs are located in lips, cheeks, tongue, floor of the mouth, hard and soft palate. Thus,all thesesites can exhibit a sialolith formation, being labial and buccal mucosa the most frequently affected sites[8,10].Although the etiology is poorly understood, many factors may be associated with an increased risk of developing sialoliths, such as local trauma, acidosis and stasis salivary,hypoptyalism and increased calcium concentration in saliva[3].

Furthermore, for sialolithiasis of MSGs, thehistopathological analysis is necessary to confirm radiological and clinical diagnosis[8]. Thetreatment comprehends the complete surgical excision under local anesthesia (sialolith and affected gland) and the recurrence is rare[6].

Therefore, this study aims to present a case of sialolithiasis in a minor salivary gland emphasizing the importance of a complete clinical evaluation and clinical/histopathological findings to correct diagnosis.

Case report

A 43-years-old blackfemale patient attended the Oral Diagnosis Center of the School of Dentistry of the University of Sao Paulo, without any complaining, for dental care. Intraoral examination revealed ayellowish 2mm nodule at labial mucosa adjacent tothe right upper canine, with fibrous consistency, smooth surface, sessile and well-defined margins (Figure 1A), which was asymptomatic and unnoticed by the patient. The clinical differential diagnosis included lipoma and fibrous hyperplasia. An excisional biopsy was performed,and an indurate encapsulated yellow tissue was obtained (Figure 1B)and submitted for histopathological analysis.

The microscopic evaluation showed a structure characterized by the presence of acellular mineralized tissuearranged in concentric laminations interpreted as sialolith (Figure 1C). Based on the findings, the final diagnosis of sialolithiasis in the minor salivary gland was established. The patient remains in follow-up and, after eight months of the surgical removal, there are no signs of relapse.

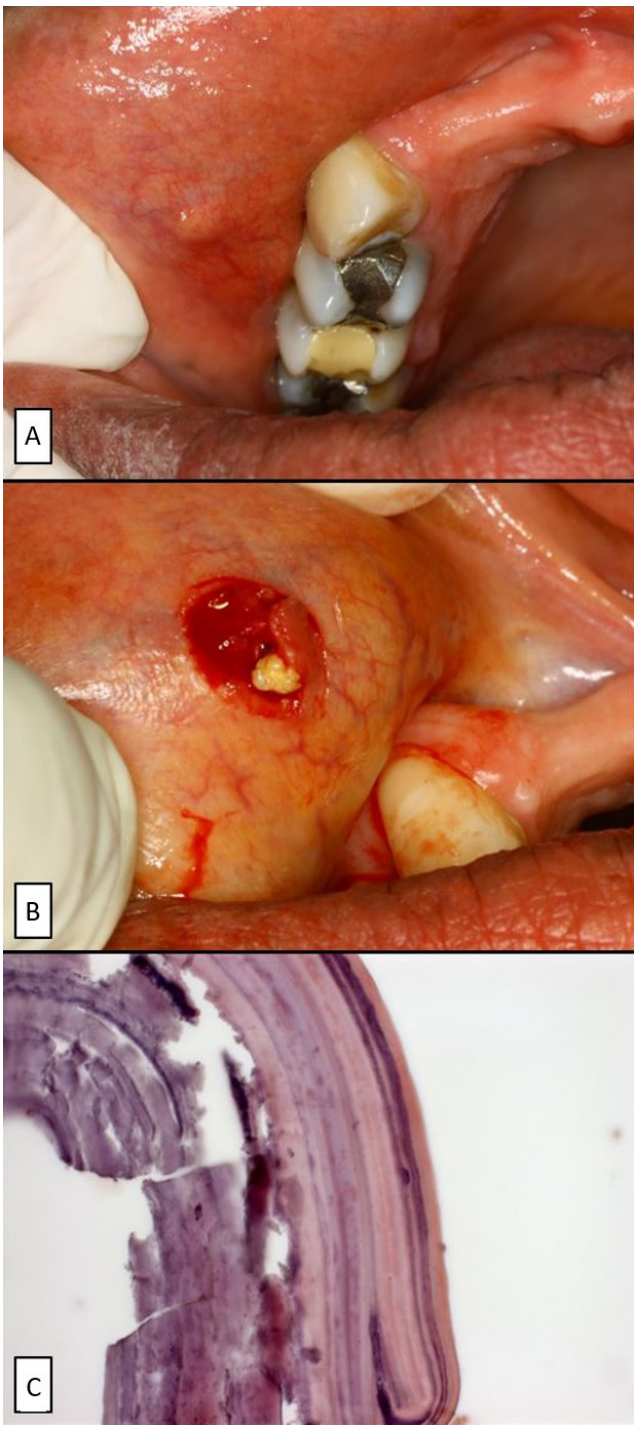

Figure 1 – Initial clinical aspect of sialolith in upper labial mucosa (A) and intraoperative surgical excision of a small yellowish circumscribed specimen (B); H&E stain at x200 magnification showing acellular calcified material arranged in concentric laminations (C)

Source: FOUSP Stomatology Clinic.

Discussion

There is no definitive cause to explain the formation of sialolith so far. Some authors suggest its origin as a physiological process, others associate local trauma, morphoanatomy and salivary composition as predisposing factors[10]. Lagha *et al*.[8] (2005) describe the process of *calculi* formation in two phases: the first one is the presence of irritant factors causing spasmodic contraction of the salivary gland duct and, consequently, stasis of saliva, and the second phase is the physicochemical promotion of calculi development.

Sialolithiasisusually occurs in major salivary glands and in MSG is considered rare, accounting for less than 1% of the cases[10]. Although occurring in both genders, there is a slight male predominance, affecting more commonly middle-aged patients[1]. The present case affected the age group and location usually reported in the literature for MSGs sialolithiasis[2,12,13].

Location and size of the sialoliths constitute the mainly factors related to the severity of signs and symptoms, directing the clinical diagnosis. In major SGs, the main complaint is the sudden onset of a painful swelling of the affected gland, which is most pronounced at meals (before, during and after). However, the lesion can be asymptomatic and detected accidentally by a routine radiographic exam[11,14].

Sialoliths in MSGs are usually asymptomatic causing, if noticeable, an increase in volume, which is usually clinicallymisdiagnosed in most cases. Due to its rarity, is often diagnosed as sialadenitis, mucocele, fibroma, lipoma or other SGs tumors[6,8].Besides this, other reasons for the underestimation of MSGssialolithiasisinclude spontaneously resolution of some cases and absence of the sialolith in the histopathological sections[6].

Sialolithiasis of major SGs can be observed in panoramic and intraoral occlusal radiographies as a solitary radiopaque body. However, in MSGs, the lesion is normally small and may not be observed through radiographic evaluation[2].

Moreover, due to the rarity of this lesion and the above discussed misdiagnose, the radiographic examination is seldomindicated.

Especially in the present case, the palpation of this lesion, due to its size, do not revealed a solid pattern, which could lead the suspicion of a calcified lesion and, consequently, the indication of the radiographic examination. The patient did not notice the lesion and thisfinding took part of a detailed clinical examination associated with the histopathological exam.

Through histopathological analysis, this lesion is characterized by the presence of a mineralized tissuearranged in concentric layers or a solid calcified mass within a minor salivary gland. Other findings, such as acinar atrophy, ductal ectasia and periductal inflammation can be observed[6]. In present case, histologic findings showedamineral fragment arranged in a concentric lamellar structure alternating layers of inorganic substances without the presence of inflammatory cells and gland structures.

The treatment of choice for sialolithiasis of MSGs is the excision of the sialolith and the associated MSG, avoiding the possibility ofrecurrences, which are unusal[8,11]. In our case, the lesion was surgically removed under local anesthesia and the patient remains under followed-up and there are no signs of relapse.

Conclusion

In summary, sialolithiasis of MSGsare uncommon and misdiagnosed. This entity should be included as a clinical differential diagnose of submucosal nodulesaffecting labial or buccal mucosa, even if the lesion were not indurated, including indication of radiographic and histopathological evaluation and emphasize the importance of a careful oral examination.

Resumo

Introdução: a sialolitíase é a forma mais comum de sialadenite obstrutiva causada por um composto de diferentes produtos, como fosfato de cálcio e matriz orgânica. É uma das doenças mais comuns das glândulas salivares, geralmente atribuídas à glândula submandibular, sem relação com idade ou sexo. No entanto, raramente é relatada nas glândulas salivares menores. Objetivo: reportar um achado clínico incomum de sialolitíase em glândula oral menor na mucosa labial. Relato de caso: uma paciente do sexo feminino, de 43 anos, apresentou nódulo único, amarelo e assintomático na mucosa labial durante o exame clínico. As hipóteses clínicas foram lipoma e hiperplasia fibrosa. A lesão foi encaminhada para biópsia e a análise histopatológica mostrou um tecido mineralizado. O diagnóstico final foi de sialolitíase e o paciente permaneceu em acompanhamento por 8 meses sem recidiva. Conclusão: este caso mostra que a sialolitíase deve ser incluída nas hipóteses diagnósticas de lesões em áreas de glândulas salivares menores e enfatiza a importância de um exame clínico completo, pois não se tratava da queixa principal da paciente.

Palavras-chave: Sialolito. Sialadenite. Glândulas salivares menores.

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