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| ORIGINAL

Oral mucocele exhibiting an extraoral swelling: a case report of an atypical presentation

Mucocele oral exibindo edema extraoral: relato de caso atípico

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ABSTRACT

Despite being common pathological entities found in the oral cavity, oral mucoceles can present distinct features, raising several diagnostic possibilities and treatments. In this study, we report the case of a 34-year-old man with an asymptomatic increased volume in the left periorbital region of more than a year, without remission or associated trauma. An aspiration biopsy was performed, followed by an excisional biopsy, and the specimen was sent for histopathological examination. Results led to the diagnosis of an oral mucocele. Surgical removal was performed in an excisional biopsy. Subsequently, the patient recovered promptly without further complications. Although oral mucoceles recur relatively often, its prognosis is good. This case emphasizes the importance of obtaining a detailed disease history, knowledge of its clinical features, and etiopathogenesis combined with complementary examinations to establish diagnostic hypotheses and converge on an adequate and individualized treatment plan

Indexing terms: Case report. Differential diagnosis. Mucocele.

RESUMO

Apesar de serem entidades patológicas comumente encontradas na cavidade oral, as mucoceles podem apresentar características distintas, o que pode sugerir diversas possibilidades diagnósticas e de tratamento. Paciente do gênero masculino, 34 anos, compareceu ao ambulatório com queixa de aumento de volume assintomático na região periorbitária esquerda há mais de um ano, sem remissão ou trauma associado. Foi realizada biópsia aspirativa seguida de biópsia excisional e a amostra encaminhada para exame histopatológico, que confirmou o diagnóstico de mucocele oral. O paciente evoluiu sem queixas e sem recidiva. Apesar da alta taxa de recorrência, mucoceles orais têm um bom prognóstico. Este relato de caso enfatiza a importância da obtenção de uma história detalhada da doença, do conhecimento de suas características clínicas e da etiopatogenia, para estabelecer hipóteses diagnósticas e convergir para um plano de tratamento adequado e individualizado.

Termos de indexação: *Relatos de casos. Diagnóstico diferencial. Mucocele.*

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INTRODUCTION

Oral mucoceles (OM) are benign pseudocystic lesions characterized by the rupture or obstruction of the salivary gland duct with mucous retention in the adjacent soft tissue [1-3]. OM usually present as small dome-shaped blue, purple, gray, or pink masses, which are painless, with a soft consistency, and measure <1 cm in diameter [1,2]. OM is more prevalent in young adults and most commonly affect the lower lip mucosa. They may also appear in sites where there are larger and smaller salivary glands such as the buccal mucosa and the floor of the mouth [1,4-6].

The etiology of OM is unknown, but it is typically associated with a history of local trauma resulting in ductal injury [1,2]. Thus, mucin accumulation triggers an inflammatory reaction that results in granulation and fibrous tissue that surround the liquid circumscribing the cavity [6]. Histologically, it is characterized by a cavity filled with eosinophilic constituents and inflammatory infiltrates surrounded by a granulation tissue, instead of an epithelial layer; therefore, it can be defined as a pseudocyst.

Although asymptomatic, OM can impair mastication and speech and cause cosmetic issues, depending on the lesion location and size. The standard treatment involves aspiration biopsy and total surgical removal, followed by histopathological analysis for appropriate diagnosis and outcome.

CASE REPORT

The patient signed a consent form to publish clinical photographs, and the case report was approved by the Institutional Ethical Committee under Protocol no. #5.638.565.

A 34-year-old man was referred to the Oral and Maxillofacial Surgery Clinics at School of Dentistry at University of Sao Paulo for evaluation of volume increase in the left jugal region. He did not have any relevant medical history or previous local trauma. His primary concern was aesthetics and function because it interfered with mastication. He noticed an increase in volume for over a year but was unable to determine the exact date of appearance of the first sign. The lesion increased in volume, with no signs of remission, leading to difficulty in mastication over the last few months because of the increasing size.

Extraoral examination revealed facial asymmetry, with an increase in volume in the lower left perioral region, extending from the labial commissure to the anteroinferior portion of the mandible, in the region of the labiomentonian sulcus (figure 1A,B). Intraoral examination revealed a localized volume increase in the left buccal region, firm to palpation, painless, underlying the mucosa, with normal color and texture (figure 1 C,D). No signs of infection were observed, and no drainage of secretions of any kind was noticed.

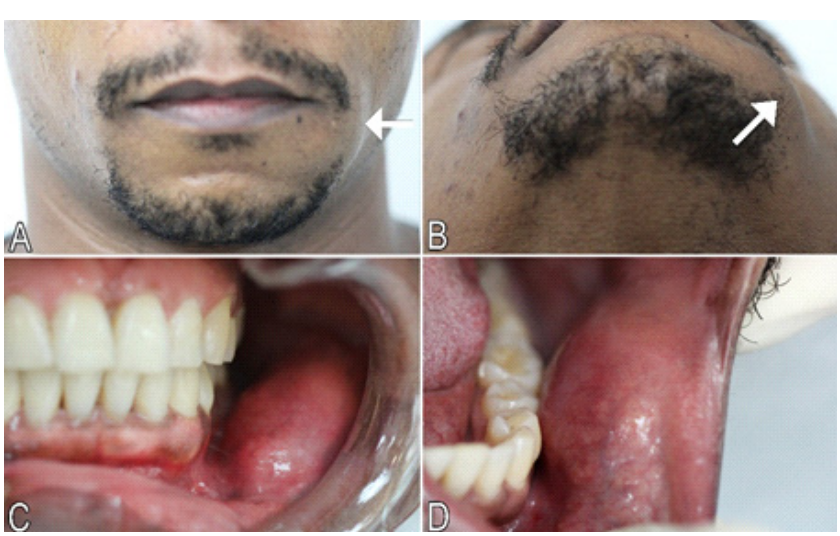

Figure 1. Clinical aspect of the lesion. A-B: Increased volume in the left cheek region (white arrows). C-D: Increased intraoral volume underneath the normal mucosa.

An aspiration biopsy was initially performed under local anesthesia (lidocaine 2% with epinephrine 1:200.000) (figure 2A). Approximately 1 mL of aspirate was obtained, and it was viscous, yellowish-transparent, resembling saliva (figure 2B), indicating salivary gland involvement. Thereafter, a linear incision was made, which was then followed by a blunt division of the soft mucosa and the underlying muscle fibers (figure 2C). The assistant used tactile pressure on the mucosa to facilitate the emergence of the lesion through the incision (figure 2D). The lesion was encapsulated, and the capsule was carefully dissected and excised (figure 2E, F). During the dissection procedure, the capsule was damaged, and the contents leaked. However, the entire lesion (approximately 30 mm in diameter) could be gently removed with associated minor glands (figure 2J), followed by the closure of the muscular and mucosal planes using resorbable sutures (figure 2G-I). The follow-up was uneventful, and the patient was satisfied with the outcome, without esthetic issues or functional concerns.

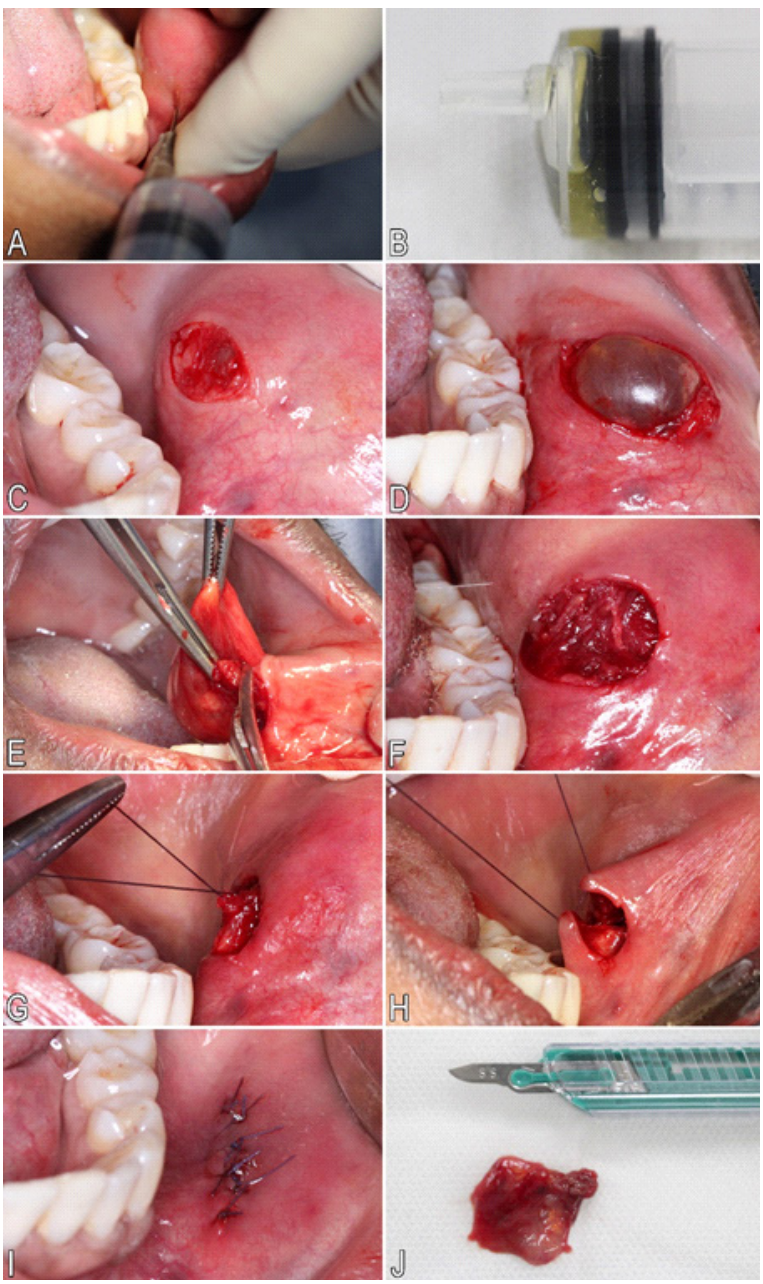

Figure 2. Intraoperative images. A-B: An aspiration biopsy obtained a viscous content, resembling saliva. C-E: A linear incision followed by a blunt division of the soft mucosa and the underlying muscle fibers to excise the capsule. F: Wound after the lesion removal. G-I: Wound closured by planes. J: Clinical aspect of the removed lesion.

Histological examination revealed mucosal fragments lined by parakeratinized stratified squamous epithelium. The lamina propria represented a narrow strip of fibrous connective tissue, underlying irregular mucin extravasation exhibiting foamy macrophages with foci of chronic inflammatory infiltrate and multinucleated giant cells. Altogether, the clinical aspect and histological description confirmed the diagnosis of OM (figure 3).

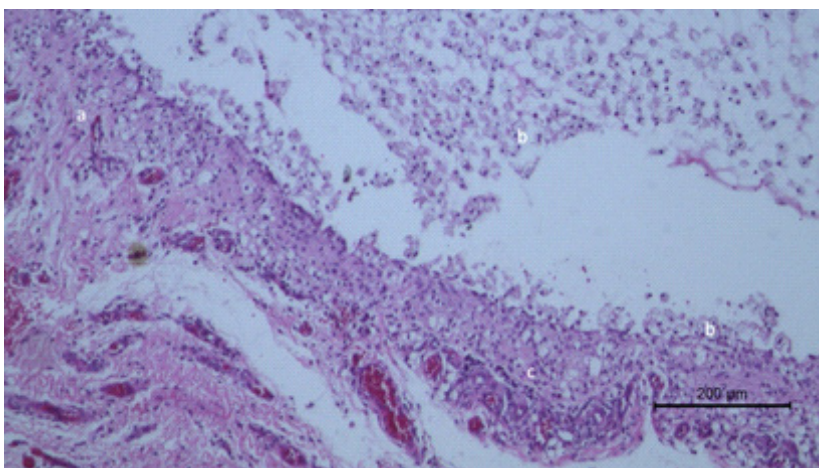

Figure 3. Photomicrography of the lesion fragment evidencing (a) fibrous connective tissue capsule surrounding extravasated mucin showing (b) foamy macrophages and (c) peripheral granulation tissue with multinucleated giant cells (original magnification 400X; hematoxylin and eosin).

DISCUSSION

The literature is unclear about the use of the term "mucocele." It has been described as a mucus extravasation phenomenon accompanied by a mucus retention phenomenon [5]. OM are painless, asymptomatic swellings that have a relatively rapid onset and fluctuate in size. They may be present as a fluid-filled vesicle, a blister in the superficial mucosa, or as a fluctuant nodule deep within the connective tissue. Spontaneous drainage of the inspissated mucin, specifically in superficial lesions, followed by subsequent recurrence may occur. Some authors consider both retention and extravasation as OM [5]; others recognized OM as mucous extravasation, whereas retention phenomena are restricted to salivary duct cysts [7]. However, the main concern regarding these reviews or case series is the ambiguity whether epithelial-lined lesions are salivary duct cysts rather than OM [1,7]. Although being discussed with epidemiological importance, both entities should be surgically treated to minimize relapse [8].

Despite being a common lesion, this clinical case draws attention to its unusual history and clinical presentation, which can result in distinct differential diagnoses, especially for general dentists. OM are often found as localized swellings in the underlying mucosa at the lower lip, buccal mucosa, and soft palate regions [1,2]. However, when the extraoral volume increased, other diagnostic possibilities should be considered. Another important feature in the diagnosis of OM is the history of lesion evolution, which is usually associated with local trauma and episodes of swelling and remission [1,2]. In this case, the absence of history and progressive increase were considered in determining the diagnostic hypothesis. Thus, the lesion could be misdiagnosed as other pathologies such as lipoma, oral lymphangioma, mucoepidermoid carcinoma (mainly the cystic variant), pleomorphic adenoma, oral focal mucinosis, and myxoglobulosis [9-11]. The aspiration biopsy obtained liquid resembling saliva; thus, lipoma and pleomorphic adenoma were excluded. The histological analysis of the excised tissue ruled out other hypotheses and confirmed OM. This reinforces the importance of histopathological examinations of all tissues associated with the disease history to determine accurate treatment and follow-up regimen.

Among treatment options, complete surgical removal is a low-cost and effective treatment option, especially when combined with the removal of adjacent minor salivary glands to minimize recurrence. The procedure should be planned according to the lesion size, location, and age. However, the removal of large lip lesions may have aesthetic

issues [12]. Furthermore, uncooperative pediatric patients are a concern for this time-consuming surgical procedure. Among treatment options, micro-marsupialization and CO2 laser ablation might be considered [13-15].

Minor surgical complications such as postoperative infection and minor bleeding can occur, which are primarily caused by inadequate surgical technique. Recurrences are not rare, but can be avoided by removing associated minor glands [2] and local trauma. Despite the absence of a clear history in this case, persistent cheek biting could have contributed to the lesion's continuous and gradual growth. Overall, this case report highlights the value and importance of assessing the history of the lesion's evolution, combined with the clinical and histopathological examinations, to determine the appropriate diagnosis and treatment prognosis.

This case report was prepared in accordance with Surgical Case Report guideline [16].

Collaborators

L Morita, conceptualization (equal), visualization (equal), writing - original draft (equal), writing - review & editing (equal). VPS Santos, conceptualization (equal), visualization (equal), writing - original draft (equal). MCZ Deboni, conceptualization (equal), writing - original draft (lead), writing - review & editing (equal). EP Ferraz, conceptualization (equal), supervision (lead), writing - original draft (lead), writing - review & editing (lead).

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