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Extravasation mucocele involving the ventral surface of the tongue (glands of Blandin–Nuhn)

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Summary. *Background*. Mucocele is a lesion that involves the salivary glands and respective current ducts caused mainly by traumas in the affected area. Two different histological forms can be found: extravasation phenomenon and mucus-retention cyst where the former is the most frequently observed involving minor salivary glands such as the glands present in the anterior portion of the ventral surface of the tongue (glands of Blandin–Nuhn).

Case Report. This report describes a large lesion involving the ventral surface of the tongue that was definitively diagnosed by histological examination as extravasation mucocele.

Conclusion. Important concepts are reviewed to help clinicians correctly diagnose and treat this pathology.

Introduction

Mucocele is a common benign lesion of the oral mucosa etymologically meaning a cavity filled with mucus (muco meaning mucus and coele meaning cavity), which is the secretory product of salivary glands. The mechanisms for the development of these lesions are two, mucus extravasation and mucus retention [1,2]. The former is the primary cause of mucocele formation involving accessory salivary glands as a result of a physical trauma. The fluid from the ruptured ducts or acni leaks and accumulates into the adjacent tissue leading to swelling [3]. The resulting pool of glandular secretion is first surrounded by inflammatory cells and later by reactive granulation tissue consisting of fibroblasts as a result of an immune response. Although there is no epithelial lining surrounding the mucin, it becomes well encapsulated by this granulation tissue and it is therefore categorized as a false cyst or pseudocyst.

In contrast, a mucus retention phenomenon is a true cyst as it is lined with epithelium. This type of cyst, referred to as mucus-retention cyst, appears to be caused by epithelial proliferation of a partially obstructed salivary duct, which becomes unable to adequately drain the saliva produced, leading to ductal dilatation and swelling [1]. Complete obstruction of a salivary duct by a calcified mass is called a sialolith, also known as a salivary calculus or stone.

The most common site of occurrence of mucoceles is the lower lip (*c*. 80%) followed by the ventral surface of the tongue, represented mainly by the extravasation type [4]. Retention mucoceles are usually located in the cheek, palate, and floor of the mouth (ranulas), which is the third most frequent location (*c*. 6%) [4], with equal incidences in both the sexes [5,6]. There appears to be no age predilection, with lesions being present at birth to the ninth decade. Some studies, however, disprove this notion with lesions appearing mostly in the first three decades of life [5,6]. The disparate site and age incidences of extravasation and retention mucoceles suggest that these two types are not related and have different pathogenesis [4].

On clinical presentation, a mucocele usually appears as an asymptomatic nodule, with a normal or bluish colour depending on the size of the lesion, its proximity to the surface, and the elasticity of the overlying tissue [1]. It is fluctuant and movable because of its mucinous contents. The diameter may range from a few millimeters to a few centimeters. If left without intervention, an episodic decrease and increase in size may be observed, corresponding to the rupture and subsequent mucin accumulation [7].

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Treatment usually consists of surgical excision including the servicing mucous glands with evacuation of its contents. Other options, especially for large lesions, include the creation of a pouch (marsupialization) inside of the lesion, freezing (cryosurgery), laser ablation, and micromarsupialization. Investigators have also suggested the use of steroid injections as an option to surgery [8].

The present report describes the clinical features of a large mucocele involving the ventral surface of the tongue (glands of Blandin–Nuhn) and reviews important concepts to help clinicians correctly diagnose and treat this pathology.

Case report

A 9-year-old girl was brought to the Pediatric Dentistry Clinic at the University of São Paulo State Araraquara Dental School by her mother with the chief complaint of swelling on the ventral surface of the tongue, which appeared 2 months before the consultation. Extraoral examination revealed no asymmetry of the neck and no cervical lymphadenopathy. On intraoral examination, the mass was superficial, protruding, fluctuant, soft on palpation, and approximately 20×20 mm in size (Fig. 1). The lesion did not blanch under digital pressure. The patient had been aware of the swelling for about 2 months but denied any episodic reduction and increase in size as well as any episodes of trauma to the maxillofacial region. There was no evidence of calcification or retained foreign body in a radiograph of the soft tissue in this area.

The established clinical diagnosis was mucocele. The micromarsupialization technique was first performed, passing a 4·0 silk suture through the internal part of the lesion along its widest diameter after previous disinfection of the area with 0·12% chlorhexidine solution and delivery of local anaesthesia, making a surgical knot. An immediate extravasation of mucus while passing the suture helped sustain the clinical diagnosis of mucocele what could have also been performed using a fine-needle aspiration technique. Seven days later, the lesion showed no signs of regression and an excisional biopsy was performed, including the removal of all marginal glands before primary closure. The wound was closed with 4–0 sutures (gut for deep closure and silk superficially).

The biopsy sample was immediately fixed in 10% formalin and sent for histological evaluation. Six-micron-thick sections mounted on glass slides were stained with haematoxylin–eosin (HE). The

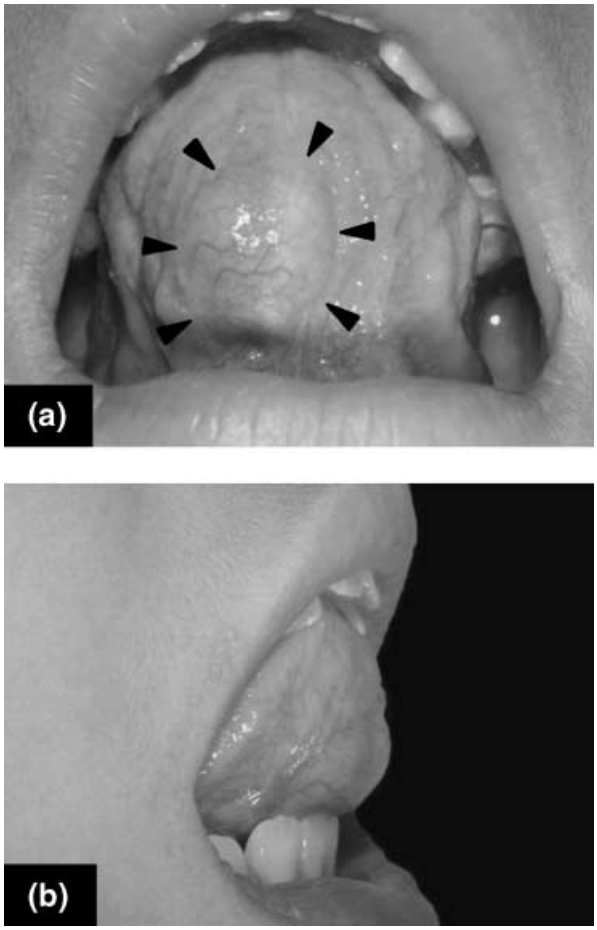

Fig. 1. Clinical aspect of the large lesion located in the ventral surface of the tongue (arrow heads). (a) Frontal view and (b) lateral view. The lesion was asymptomatic, pink-coloured and presented a fluid-filled consistency. The clinical diagnosis was mucocele.

histological evaluation of the sections by light microscopy revealed the presence of a granulation tissue lining the mucus cavity surrounded by a condensation of connective tissue rich in inflammatory cells and congested blood vessels (Figs 2 and 3). Mucus pool related to foamy cells was observed within the lumen of the lesion (Fig. 3b). Salivary glands and dilated ducts were also seen at the base of the lesion (Fig. 2). The definitive diagnosis was mucus extravasation mucocele on the ventral surface of the tongue.

The patient was re-examined every 3 months after the surgical excision of the lesion and review at 12 months showed no signs of recurrence (Fig. 4).

Discussion

The differential diagnosis in a case such as the reported should include lesions known to cause

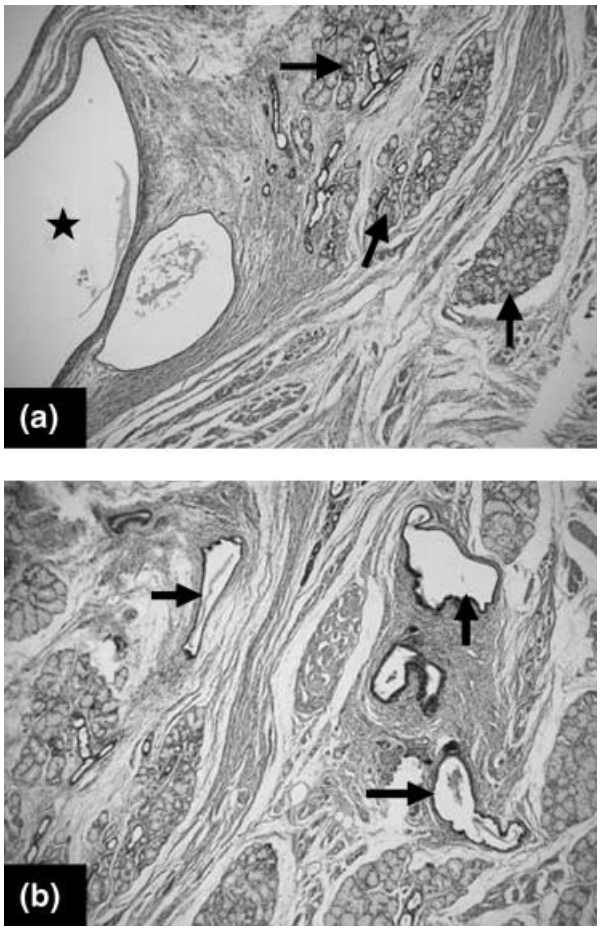

Fig. 2. (a) Salivary glands (arrows) present at the base of the lesion (★). HE; × 64. (b) Several dilated salivary ducts (arrows) are observed adjacent to the glands. HE; × 64.

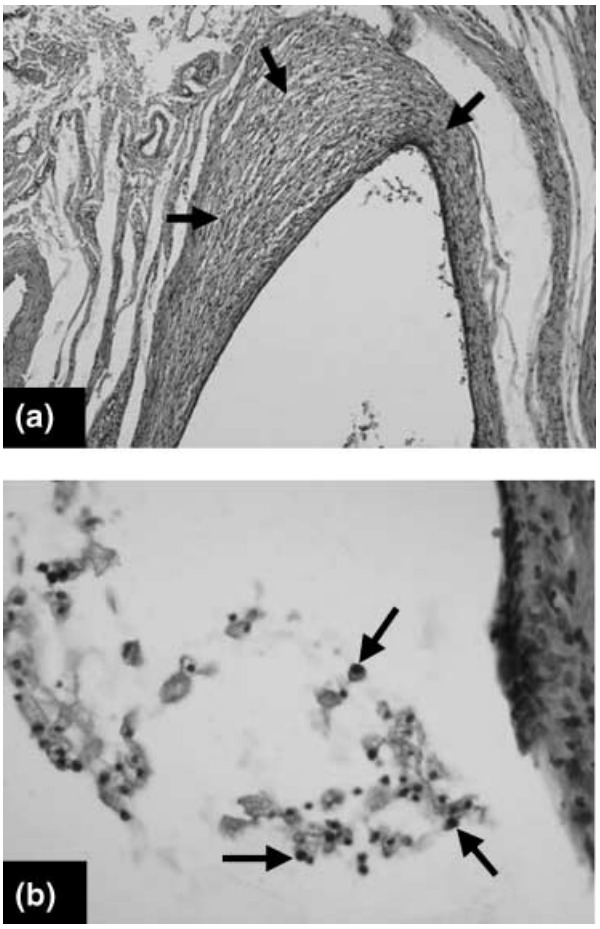

Fig. 3. (a) The pool of mucus leaked to the connective tissue is surrounded by granulation tissue (arrows). HE; × 82. (b) Detail of the mucus within the lumen of the lesion. Note the presence of foamy macrophages (arrows). HE; × 320.

swelling of the tongue. The tongue contains adipose, connective tissue, blood vessels, nerves, and salivary glands, so pathosis of any of these tissues is possible. Palpation of the lesion may aid in developing the differential diagnosis as some lesions such as lipomas and salivary gland tumours do not exhibit fluctuance, whereas cysts, mucoceles, abscesses, and haematomas have a fluid-filled consistency. Needle aspiration might be performed to rule out a vascular cause or a cystic mucoepidermoid tumour, as their clinical appearance may be similar [9].

Extravasation and mucus retention phenomenon are difficult to differentiate clinically as both lesions present identical characteristics. Regardless of their location, mucoceles present as a soft, painless swelling varying from deep blue to normal pink in colour [1]. Lesions superficial to the mucosa usually present with a bluish colour, whereas the overlying tissue of deeper lesions can have normal colouration [1], as observed in the present case. Usually blue in colour, these blanch under digital pressure, which distinguishes them from other pigmented lesions such as nevi, haematomas, and melanomas [10].

Mucocele of the anterior lingual salivary glands are uncommon [9,11], particularly large lesions as in the case reported. Three distinct sets of minor salivary glands can be found on the human tongue, the glands of von Ebner, the glands of Weber, and the glands of Blandin and Nuhn, which are mixed mucous and serous glands embedded within the musculature of the anterior tongue ventral surface [11] covered only by a thin mucous membrane [1]. Mucoceles involving the glands of Blandin–Nuhn are often histologically diagnosed as being extravasation type and likely to occur in young patients [1,9,11].

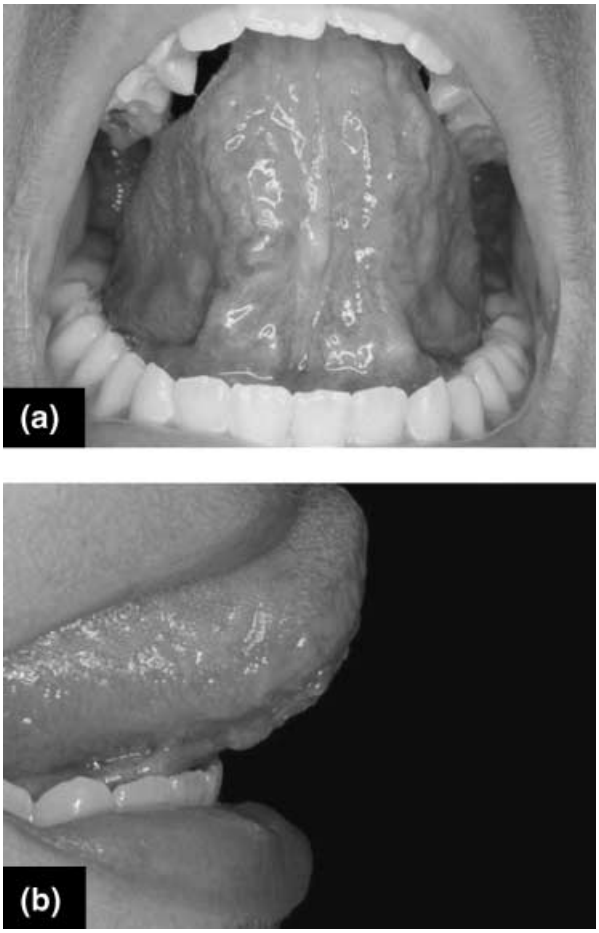

Fig. 4. Clinical aspect of the ventral surface of the tongue at the 12-month re-examination. (a) Frontal view and (b) lateral view.

Traumatic injury to a duct or ducts with partition of this structure is the most likely aetiologic factor leading to the development of these lesions [4]. Although a history of trauma is frequently associated with the onset of mucoceles, in several cases the patients are unable to recall any episodes of injuries to the tongue [9]. In the present reported case, the patient denied trauma and parafunctional habits were not noticed. The patient also denied any episodic decrease and increase in the size of the lesion. A mucocele may persist for as long as a year without rupturing, but most of the patients present within a few weeks of onset for evaluation. These lesions are often asymptomatic, however, as they grow in size, they can cause discomfort, external swelling, and interfere with speech and mastication [1]. A large mucocele of such long-standing duration as the lesion reported here is rare and can be very alarming.

In the present reported lesion, the mucus cavity was not lined by epithelium, instead there was a granulation tissue surrounding the mucus collection. The development of granulation tissue is important to limit the spread of the mucus and to facilitate its removal by macrophages. The molecular mechanisms, however, that are involved in the growth of extravasation mucoceles are still unclear [12]. The luminal fluid from extravasation mucoceles contains a large amount of matrix metalloproteinases, especially MMP-9, MMP-2 [12], and MMP-1 [13], when compared with saliva from Wharton's duct [12]. These proteolytic enzymes produced by macrophages and fibroblasts are involved in remodeling of the connective tissue and may contribute to the development of the lesion [12].

Superficial extravasation mucoceles may require no treatment [4,14], whereas the surgical approach to the management of large mucoceles includes techniques such as the unroofing procedure (marsupialization) prior to the dissection of the lesion along with the servicing mucous glands [1]. Micromarsupialization is an alternative technique that can be easily performed, aiming the drainage of the mucus and reduction of the lesion in size, which involves passing a thick silk suture through the internal part of the lesion along its widest diameter making a surgical knot after previous disinfection and anaesthesia [7]. The suture material is removed 7–10 days later, at which time the mucocele is expected to be resolved. The advantages of this technique include simplicity, minimum trauma, and relative lack of pain. There is only little information in the literature, however, regarding the rate of success of this technique which, in general, is not recommended for large lesions. In our case, despite the size of the lesion, micromarsupialization was first performed because of the simplicity of the procedure. Moreover, there is no evidence that this technique inflicts any harmful effects to the established lesion, even when it is not successful. Delbem *et al.* [7] reported a high rate of success after performing this procedure in small mucoceles with smooth surface, thin mucosa, sessile base, and flaccid consistency located in the lower lip, floor of the mouth, and ventral surface of the tongue.

Because of the possibility of a lesion recurrence, excision is warranted for definitive treatment and diagnosis. When possible, it is beneficial to identify and remove the glands associated with the lesion to reduce the rate of recurrence [1]. If a benign or

malignant tumour is diagnosed, then referral to an appropriate specialist is necessary, as further surgery (to obtain clear margins) and other more specific therapies may be indicated.

What this paper adds

- Mucoceles are relatively common lesions in pediatric patients, however, they can be very alarming when assuming a large volume.
- This paper shows the clinical approach to an unusual mucocele lesion and stresses the importance to validate the clinical diagnostic with a histopathological analysis.

Why this paper is important to paediatric dentists

- Paediatric dentists should be prepared to encounter variations of more predictable clinical appearances of mucoceles to adopt the best conduct regarding diagnosis and treatment.

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