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Mucocelles of the anterior ventral surface of the tongue and the glands of Blandin Nuhn: 5 cases

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Introduction

Oral mucocoeles (MC) are cystic diseases of the oral mucosa [1]. The most common are ranula and MC of the lower lip [2]. Blandin and Nuhn mucocoeles (BNMC), which develop at the ventral side of the tongue, are rare benign lesions [2]. They are often misdiagnosed and confused with MC of the floor of the mouth that extend to the ventral surface of the tongue. Their management with marsupialisation is not recommended. The objective of this study was to describe our cases and compare their clinical aspects, check-up, and management with those found in the literature.

Patients and methods

Five patients, 4 male and one female, were treated for Blandin and Nuhn mucocoeles in our department from 2011 to 2017. The age range was from 7 to 39 years, with an average age of 18.2 years. Four lesions presented as a submucosal swelling, the other one appeared papillary. All the lesions were located at the ventral surface of the tongue near the lingual frenulum. One lesion was medial and 4 were on the left side of the frenulum. One patient underwent an incomplete excision of the lesion performed in other hospital.

Results

We describe five clinical cases to detail their clinical history, care, and treatment. This study was conducted in accordance with the principles of the Helsinki Declaration (2013)

Case 1 (Fig. 1)

An 8-year-old male presented with a small lesion of the ventral face of his tongue. Due to the small size and lack of suspect criteria, no further imaging was performed. BNMC was completely removed under local anaesthesia with no complications. The patient was followed during two years without BNMC recurrence.

Case 2 (Fig. 2 and 3)

A 16-year-old male presented with a ventral tongue lesion gradually increasing in volume. Preoperative ultrasound and CT showed a unilocular 20 x 9 x 9 mm round avascular cyst with a large axial axis. MRI showed a cystic formation on the left anterior pelvic surface of the tongue in intermediate signal T1 and hypersignal T2. The cyst measured 19 mm wide, 9 mm in height, and 7.5 mm across. There was no gadolinium enhancement.

The lesion was surgically removed under general anaesthesia. The pathological results showed a cystic formation 22 x 10 x 10 mm with foamy histiocytes and mucoid material accumulation. On the outskirts, the corium was the seat of collagen and fibro-inflammatory changes. On the surface, there was a mature squamous, no disorganisation of the epithelial architecture, and no signs of malignancy. The histological appearance favoured a mucoid cyst. The patient was followed during one years without BNMC recurrence.

Case 3 (Fig. 4)

A 39-year-old male presented with a centimetric ventral tongue lesion. A CT showed a cystic lesion with liquid density. Surgical excision of the BN gland was done under general anaesthesia. The patient was followed during six months without BNMC recurrence.

Case 4 (Fig. 5)

A 21-year-old male presented with a swelling of the left ventral surface of his tongue. The lesion was indurated and close to the oral floor. Marsupialisation of the sublingual gland and its excision one month later failed. BNMC was initially confused with a ranula, explaining the lack of imaging and the sublingual surgery failure. After excision of the Blandin and Nuhn gland, the patient presented with one recurrence of the mucocele that was treated by a second surgical resection. The patient was then followed during 6 months without recurrence.

Case 5 (Fig. 6)

A 7-year-old girl presented with a lesion on the left ventral surface of her tongue. Puncture of the lesion was attempted but failed. She underwent a surgical resection without complications. The patient was then followed during 1 year without recurrence

Discussion

Blandin and Nuhn mucocèles must be understood and recognised to propose complete excision of the Blandin and Nuhn gland and avoid recurrence. BNMC are not very well-known even if they are more common than suspected [1]. BN glands open in the oral cavity by five to seven small ducts laterally to the lingual frenulum; each gland is 8 mm wide and 12 to 25 mm deep [3]. Despite the small size of the BN glands, BNMC can sometimes be large, with a long axis measuring over 5 cm, even in children. In such cases, BNMC can cause suffocation [4] or dysphagia. BNMC are more common in female children [3]; but in the current study there were 4 males and 1 female. Their clinical appearance is unique, with longitudinal swelling in the ventral surface of the tongue parallel to the lingual frenulum. Paraclinical exams (ultrasound, CT, MRI, or needle aspiration) can confirm the diagnosis but are not always necessary, as the diagnosis is clinically obvious. The BN glands of the affected side must be completely removed (Fig. 7). This requires a longitudinal incision parallel to the frenulum and 5 mm away from it. After the injection of saline solution with epinephrine, submucosal dissection removes the BN glands on the affected side [2,5,6], preserving the lingual nerve, the lingual veins, and the Wharton duct. The sublingual gland that is not concerned must be respected.

The most frequent location of MC is at the lower lip (77.9%), followed by the floor of the mouth and tongue (15.6%) [2]. BNMC are rare (9.9% of all oral mucocèles in the Jinbu study [7] and 15.4% in the de Camargo Moraes study [8]). Generally, these lesions are benign and small, measuring less than 20 mm in diameter. Because of their clinical characteristics and

location, these lesions can be confused with vascular lesions, botriomycomas, and squamous papillomas, among others [2,3]. The differential diagnosis of mucocoeles is mucoepidermoid carcinoma, whose clinical appearance is similar. It is therefore important to assess the location of the lesion, the presence of trauma, any rapid changes, an increase or decrease in the lesion size, a bluish colour, and fluid consistency.

There are 2 types of lingual mucocoeles:

- An exudative form, with the presence of granulation tissue containing leukocytes and phagocytes without epithelial lining of the lesion
- A cystic form, with a cavity lined with a squamous epithelium with inflammatory cells and mucus acini [3]

The cystic form (CF) is more alarming because it can be suffocating while the CF evolves over several weeks (one week to 6 months) [4]. The CF appears bluish, non-ulcerated, and is painless. The average age is 20 years [7,8], with a female predominance [7].

In the exudative form, we often find a trauma history, which causes a transection of the duct and therefore mucus leakage in the connective tissues. The mucus then causes inflammation and granulation tissue. The retentive cystic form occurs when debris, stones, or inflammation block the duct, causing lesion expansion. Out of 400 cases, Harrison showed that the exudative form affected mostly young subjects (20-30 years of age) at the lower lip while the cystic form instead affected the elderly and in locations other than the lower lip [5].

The recommended treatment is a complete en bloc surgical excision of the lesion [2], even in asymptomatic patients, to prevent increasing volume and complications. Surgery can be performed under local anesthesia (case 1) if the patient is compliant and if the lesion is small [1,6]; in other cases, general anesthesia is necessary (cases 2, 3, 4, and 5) [2]. A vertical incision is made in the ventral surface of the tongue, and the mucus is discharged. Because of

the presence of adhesion due to inflammation, the lesion walls can be difficult to debride; this can explain the recurrence in some cases (case 4 in the present series). The only patient herein who presented a recurrence was initially treated by marsupialisation and then sublingual gland excision. In this inflammatory context, complete resection of the cystic membrane is difficult and can explain a new recurrence. Some clinicians inject a mixture of alginate into the lesion to better discern the lesion walls once the content has been evacuated. The ventral surface of the tongue is then sutured with absorbable 4.0 stitches [6]. A histological analysis of the excised lesion is performed. The histological analysis is systematic. It shows a well-circumscribed lesion on top of mucous filled with a colourless mucoid material containing proteins and large histiocytes [4,6].

The postoperative time is often uneventful. Other treatments have been described for large lesions: IV injection of corticosteroids, cryotherapy, laser, “micromarsupialisation,” and intralesional steroid injection [4,6]. In his study [9], Zhang treated BN mucocles by absolute ethanol injection under local anesthesia in 14 patients. Mucocles disappeared in all patients after 1 to 3 injections. There were no complications. The recurrence was not observed during a 2 years follow-up. The management of Blandin and Nuhn mucocles is unclear because they are rare and poorly understood. There is no spontaneous regression of the MC, and the common treatment is surgical and performed under local or general anaesthesia. Studies are needed to elucidate the success or failure rates of these treatments before the management of BNMC can be standardised.

Disclosure of interest

The author declares that he has no disclosure.

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Figures

Figure 1: Small Blandin and Nuhn mucocoele in an 8 years old child (case 1)

Figure 2: Left Blandin and Nuhn mucocoele in a 16 years old male, progressive volume increase (case 2)

Figure 3: Multimodal imaging of mucocoele of the glands of Blandin-Nuhn (case 2)

a, b, d, d: MRI sequences, respectively axial T2SE with suppression of fat, coronal T2SE, axial T1SE and post gadolinium coronal T1SE ; e: contrast-enhanced CT; f: ultrasound.

Ultrasound and contrast enhanced CT show an oblong cyst (arrow heads) situated on the left anterior ventral surface of the tongue, with liquid density but not strictly anechogenic. MRI confirms the liquid content of this cyst (low T1signal, high T2signal and no postcontrast enhanced) with a thin wall enhancement.

Figure 4: Left Blandin and Nuhn mucocoele in a 39 years old male (case 3)

Figure 5: Left Blandin and Nuhn mucocoele (red arrow) previously confused with a ranula and first treated by sublingual excision (case 4)

Figure 6: Left Blandin and Nuhn mucocoele in a 7 years old female after cyst puncture failure (case 5).

Figure 7: Left Blandin and Nuhn excision after a longitudinal incision parallel to the frenulum and dissection respecting lingual nerve and sublingual gland (case 4).