

## Mucocele of lingual glands of Blandin and Nuhn: A report of 5 cases

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الأكياس المخاطية تحدث غالباً وبشكل متكرر في الحفرة الفموية. أكثر من ٧٠% من الحالات تظهر في الشفة السفلية. ظهور الأكياس المخاطية في اللسان يعتبر نادراً وتظهر في حوالي ٢,٢٥% من الأكياس المخاطية الفموية. ولكن وجد حديثاً أن نسبة حدوث الأكياس المخاطية الفموية في اللسان قد تتراوح من ٩-١٠% من إجمالي الأكياس المخاطية الفموية وإذا حدثت فهي تحدث في الغدد اللعابية اللسانية الأمامية لغدد (Glands of Blandin & Nuhn). تعرض هذه الدراسة خمس حالات للأكياس المخاطية في اللسان للغدد اللعابية الأمامية وكذلك مراجعة للأبحاث السابقة، والمفترض هو إزالة كل هذه الأكياس مهما كان صغيراً والمتواجدة في الغدد اللعابية اللسانية الأمامية الغير رئيسية لكي نتحاشى النكس وظهورها مرة أخرى.

Although mucoceles occur frequently in the oral cavity, more than 70% of cases occur on the lower lip. The occurrence of mucoceles on the tongue is considered rare and accounted for only 2.25% of all oral mucoceles in earlier studies. However, more recent reports suggested that the incidence of mucocele of the tongue is 9% to 10% of oral mucoceles and that they occur exclusively in the anterior lingual salivary glands of Blandin and Nuhn. In this report, five additional cases of mucocele of the glands of Blandin and Nuhn are described with review of the literature. The need is stressed to excise all lesions, no matter how minor, in the anterior ventral surface of tongue together with all minor salivary glands in the surgical field to avoid a recurrence or creation of a mucocele.

### INTRODUCTION

Mucocele is a clinical term used to describe a swelling caused by the pooling of saliva from a severed or obstructed minor salivary gland duct. Although mucoceles can occur in any location where minor salivary glands are present, they are more commonly found in the lower lip but rarely reported on the tongue.<sup>1</sup>

Three main groups of minor salivary glands are found in different locations of the human tongue. They are glands of Von Ebner, glands of Weber and the glands of Blandin and Nuhn. The glands of Von Ebner are serous glands, which are found mainly in association with the circumvallate and the foliate papillae. The glands of Weber are purely mucous glands located along the lateral border of the tongue and open into the crypts of the lingual tonsils in the posterior aspect of the dorsum of the tongue. The glands of Blandin and Nuhn are mixed mucous and

serous glands found in the musculature of the anterior ventral surface of the tongue.<sup>2</sup>

Mucoceles on the tongue are rare and occur almost exclusively on the ventral surface where the glands of Blandin and Nuhn are located. So far no cases involving the glands of Von Ebner or glands of Weber have been reported.<sup>1</sup>

Harrison,<sup>1</sup> in 1975, in a review of 400 cases of mucocele of the oral cavity, found that more than 70% involved the lower lip and only 2.25% (9) arose from the tongue. In 1980, Ishida<sup>3</sup> reported that only 8 (1.9%) cases out of 425 mucoceles involved the glands of Blandin and Nuhn. But later studies have suggested higher incidence of mucocele on the tongue. In 1982, Saza *et al.*<sup>4</sup> found that mucoceles of Blandin and Nuhn represented 9.6% (37) of cases out of a total of 385 mucoceles. Kurozu<sup>5</sup> reported that 13 (10.3%) out of 126 cases of mucoceles involved the glands of Blandin and Nuhn.

In the most recent study in 2003, Jinbu *et al.*<sup>6</sup> found that out of 263 cases of

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mucocèles of the oral cavity, 205 (77.9%) involved the lower lip while 26 (9.9%) involved the glands of Blandin and Nuhn. The latest reports suggest a higher rate of occurrence. This may be due to the fact that more cases are being reported.

The ages of the patients ranged from 5 years to 36 years with an average of 17 years. The duration between when the lesion was first noticed and the first presentation to the hospital ranged from 1 week to 2 years with an average of 3.6 months.

Andiran *et al.*,<sup>7</sup> in their literature review, found only 24 reported cases of mucocele of Blandin and Nuhn and added one more case to bring the total to 25 cases. In 2003, Jinbu *et al.*<sup>6</sup> reported 26 additional cases. Therefore, 51 cases of mucoceles of Blandin and Nuhn have been reported up to date in the English literature.

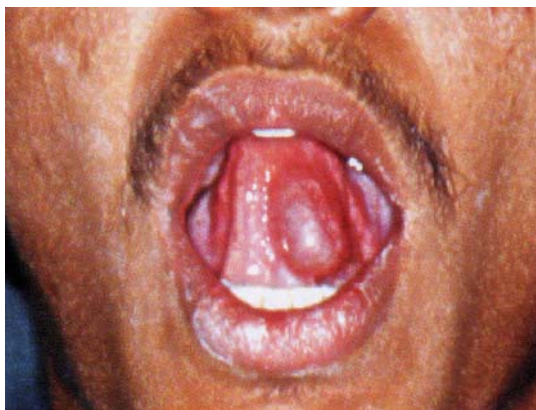
We report additional 5 cases of mucoceles of Blandin and Nuhn as well as a review of the literature.

## CASE REPORTS

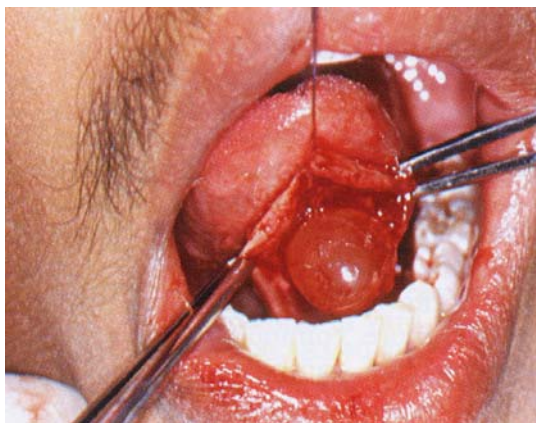
### Case 1

A 17-year old male patient was referred to our clinic with painless swelling in the ventral surface of the tongue, left of the lingual frenum, which was noticed four months earlier. There was no history of trauma. Past medical history was unremarkable.

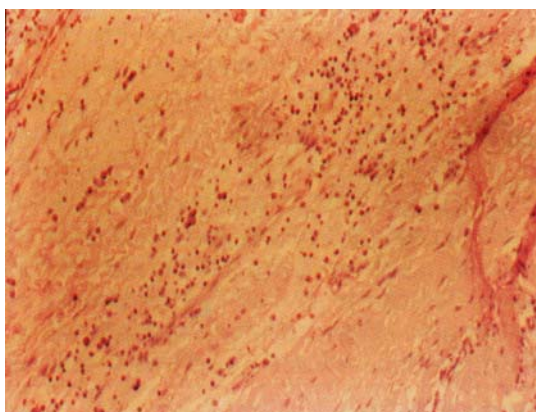
On examination, there was a soft translucent swelling on the ventral surface of the tongue, left of the midline, measuring about 20mm x 30mm, which did not empty on application of pressure. The clinical appearance was that of a mucocele. Under local anaesthesia, the lesion was excised by dissection down to the muscle layer and removed together with the associated normal minor salivary glands to avoid recurrence. The wound was closed with loose sutures (Fig. 1a and Fig. 1b).



**Fig. 1a.** Photograph of the tongue showing a large swelling on the ventral surface of tongue.



**Fig. 1b.** Intra-operative photograph showing deep dissection of the mucocele.



**Fig. 1c.** Histology of the lesion. Haematoxylin & Eosin stain. Magnification x 4.

The histopathology report was as follows: "Cystic soft tissue measuring 10mm in diameter. Cut section showed

mucus material. Microscopically, the section studied does not show cystic wall lining epithelium. However, it has a fibrous wall with few inflammatory cells and mucin collection. There is no evidence of malignancy. This is confirmed as mucocele” (Fig. 1c). There has been no recurrence 4 years postoperatively.

## Case 2

A 6-year old female patient was referred with a painless swelling of 10 days duration on the ventral surface of tongue. There was no history of trauma. Past medical history was non-contributory.

Physical examination revealed a fit young girl who was very apprehensive even for clinical examination. There was a large non-tender and non-fluctuant translucent swelling on the ventral surface of the tongue, left of the lingual frenum, measuring about 10mm x 3.5mm, which extended from almost the tip of the tongue to the papilla of the submandibular duct. The clinical diagnosis was mucocele (Fig. 2a).



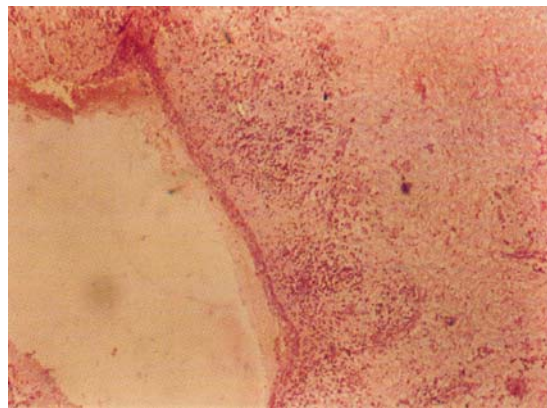
**Fig. 2a.** Photograph of the swelling of the ventral surface of tongue.

Since the child was young and uncooperative, it was decided to excise the lesion under general anaesthesia. Preoperative investigations showed haemoglobin of 10.5gm/dl, white cell count of  $8.6 \times 10^9/l$ , platelets  $237 \times 10^9/l$ .

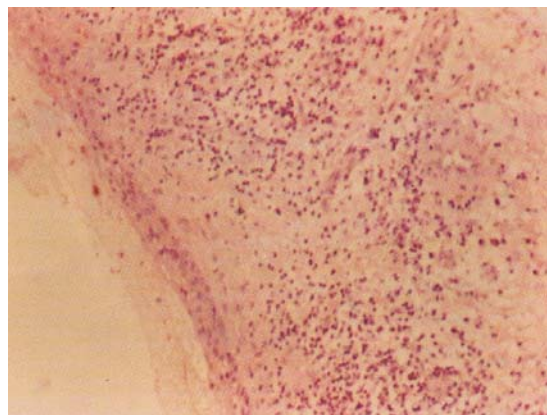
1. Urea and electrolytes and chest x-ray were all within normal parameters.

Under general anaesthesia with nasotracheal intubation, the mucocele was excised together with the associated minor salivary glands by sharp and blunt dissection down to the muscle layer. The mucocele ruptured during final stage of excision to reveal the mucus content.

The microscopic findings were stratified squamous epithelium with subepithelial hypertrophic striated muscle bundles and haemorrhage. Focal areas showed cyst wall lined by flattened lining epithelial cells, with minor salivary glands tissue showing acini and ducts. Few inflammatory cell collection, lymphocytes, neutrophils, RBC's and



**Fig. 2b.** Histology of the lesion showing epithelial lining. Haematoxylin & eosin stain. Magnification x 4.



**Fig. 2c.** Histology of the lesion showing epithelial lining. Haematoxylin & eosin stain. Magnification x 10.

muscle bundles. No malignancy noted. The diagnosis was "mucocele with non-specific inflammation and haemorrhage" (Fig. 2b and Fig. 2c). There has been no recurrence after 3 years.

### Case 3

An 8-year old female patient was referred to the clinic complaining of painless swelling of the left ventral surface of the tongue of one-month duration. She looked healthy and had no medical problems.

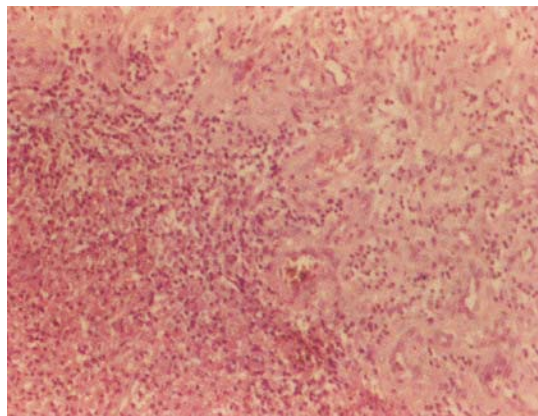
On examination, there was a firm swelling on the ventral surface of tongue, left of the lingual frenum. The clinical diagnosis was fibroepithelial polyp. It was excised under local anaesthesia with great difficulty, as the patient was not very cooperative. The histopathological diagnosis was squamous polyp.

Two weeks post operatively, the patient reported with a large non-tender translucent swelling in the area of operation extending from the tip of the tongue to just above the sublingual papilla. Because the patient did not tolerate the initial surgery under local anaesthesia very well, it was decided to excise the mucocele under general anaesthesia. The mucocele together with the adjacent minor salivary glands were



**Fig. 3a.** Photograph of the ruptured swelling of the ventral surface of tongue.

excised to avoid recurrence. The histology report was mucocele (Fig. 3a and Fig. 3b). There has been no recurrence 3 years post operatively.

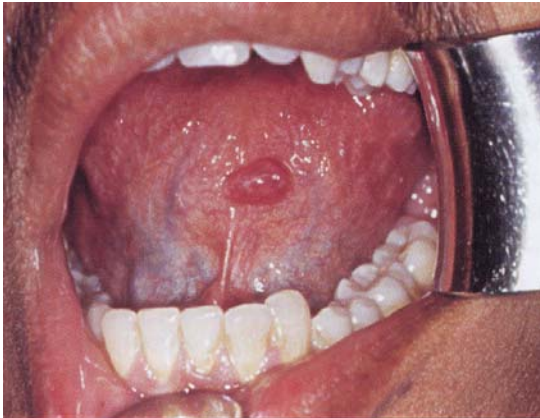


**Fig.3b.** Histology of the lesion. Haematoxylin & Eosin stain. Magnification x 10.

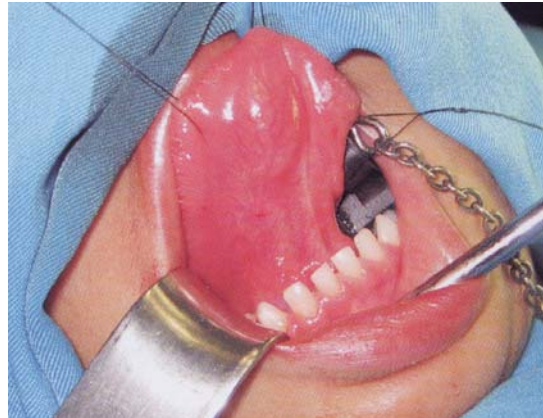
### Case 4

A 22-year old male patient was referred with a recurrent swelling on the ventral surface of tongue of about one and half month duration. He had no recollection of any trauma but revealed that the swelling usually ruptured after meals only to fill up again. His past medical history was unremarkable.

On examination, there was a pedunculated swelling on the ventral surface of tongue to the right side of the lingual frenum. The clinical diagnosis was mucocele of the anterior lingual gland (Fig. 4a and Fig. 4b). Under local anaesthesia, the lesion was excised up to the muscle layer and the surgical defect was closed with loose vicryl sutures. The histology report was as follows; " the section shows mildly hyperplastic epidermis. There is a cavity lined by granulation tissue and infiltrated by chronic inflammatory cells in continuity with salivary gland tissue. Few muscle tissues are also seen. There is no evidence of malignancy. The diagnosis is mucocele." There has been no recurrence after 2 years.



**Fig. 4a.** Photograph of the swelling of the ventral surface of tongue.



**Fig. 5a.** Photograph mucocele of the right ventral surface of tongue.



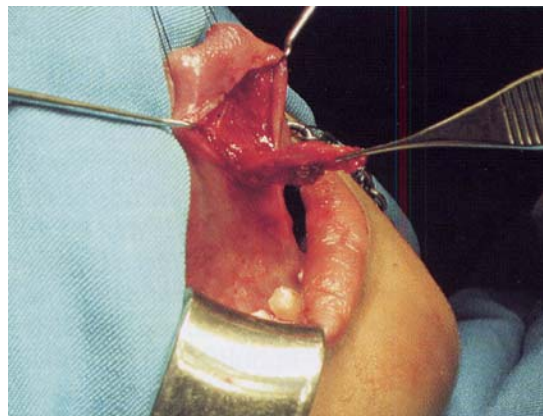
**Fig. 4b.** Photograph showing the pedunculated nature of the swelling.

### Case 5

A 4-year old boy was referred by a pedodontist with a painless swelling on the ventral surface of the tongue which the parents noticed 2 days earlier when they were brushing his teeth. The parents could not recollect any trauma to the tongue. The patient's medical history was non-contributory. On examination, there was a fluctuant translucent swelling of ventral surface of the tongue, right of the midline near the tip. The clinical diagnosis was mucocele (Fig. 5a).

Under general anaesthesia, the mucocele was excised together with the adjacent minor salivary glands up to the

underling muscle layer. The wound was closed with 4/0 vicryl and the specimen sent for histopathological examination. The patient was discharged one day post-operatively (Fig. 5b).



**Fig. 5b.** Intra-operative picture showing dissection of the mucocele.

The microscopic examination was reported as follows, "striated muscle bundles with lobules of mucin containing glandular acini and ducts, fibrous wall with collections of lymphocytes, plasma cells, few polymorphs and haemorrhage. Diagnosis is mucocele with haemorrhage and inflammation" (Fig. 5c). There has been no recurrence two years post-operatively.



Fig. 5c. Photograph of the tongue 6 months post operatively

## DISCUSSION

Mucoceles of the oral cavity are classified as extravasation or retention types. The extravasation type consists of extravasated mucus in the connective tissue and the retention mucocele results from mucus retained in an epithelial-lined cavity usually a dilated duct of minor salivary glands.<sup>1</sup>

The extravasation type, which is the more common mucocele, usually results from local trauma such as biting and is found most commonly in the lower lip and in younger age groups while retention mucocele occurs in the older age group as a result of dilatation of the duct due to blockage by a sialolith or a mucus plug and is usually found in the palate and floor of mouth. The extravasated saliva elicits inflammatory reaction and results in formation of granulation tissue whereas the retention variety is enclosed in the dilated minor salivary gland duct and may be confused with true cysts.<sup>8</sup>

Harrison,<sup>1</sup> in a review of 400 cases of mucoceles, reported that the majority was of the extravasation type and most commonly occur in the lower lip of younger patients, usually during the second and third decades of life while the retention mucoceles occur in older patients and in places other than the lower lip.

Heimansohn<sup>9</sup> first reported a case of mucocele of Blandin and Nuhn in a 14-year old female (his daughter). Since then more cases have been reported, one each by Ellis *et al.*,<sup>10</sup> Mandel and Kaynar,<sup>11</sup> and 5 cases by Sugerman *et al.*<sup>12</sup> In their literature review, Andiran *et al.*<sup>13</sup> reported that the age of occurrence of mucoceles of Blandin and Nuhn ranged from 10 weeks to 26 years with a peak in the second decade and a female predominance without stating the ratio between the sexes. However Jinbu *et al.*,<sup>7</sup> in their report of 26 cases, found that females were more affected than males by a ratio of 4:1 (21:5) and an age range of occurrence from 5 years to 36 years. So far all the patients have been younger than 40 years.

Mucoceles of Blandin and Nuhn are usually asymptomatic and relatively small in size ranging from 2mm in diameter to 20mm x 12mm even though a case measuring 35mm x 30mm in 6 weeks following trauma,<sup>7</sup> and another large case measuring 74mm x 30mm<sup>2</sup> in 2 weeks with no history of trauma,<sup>11</sup> have been reported. The lesions presented here seem to be bigger than the average reported cases including the series by Sugerman *et al.*<sup>12</sup>

The time interval between when the lesion was noticed and the first visit to the clinic range from one week to six months,<sup>6,7</sup> perhaps an indication that these lesions are usually asymptomatic. Sometimes however, mucoceles of Blandin and Nuhn can grow relatively large enough to cause feeding difficulties especially in babies<sup>13</sup> or difficulty in speech and mastication.<sup>11</sup>

Even though not every case had been attributed to trauma, the majority of the histopathology reports suggest extravasation phenomenon, which is normally preceded by trauma.<sup>7</sup>

The clinical diagnosis of mucocele of Blandin and Nuhn may not be difficult since most appear as normal mucoceles. The superficial lesions appear as thin-

walled, bluish swellings that rupture easily while the deeper lesions are well circumscribed swellings usually covered by normal appearing oral mucosa.

In the review by Jinbu *et al.*,<sup>6</sup> 24 out of 26 cases were diagnosed as mucocele with only one case diagnosed clinically as papilloma. However, Sugerma *et al.*<sup>12</sup> reported that mucocele of Blandin and Nuhn may clinically present as a vascular lesion, pyogenic granuloma, polyp or squamous papilloma. They suggested that a history of trauma and aspiration of mucus from the lesion are helpful in the diagnosis of mucocele of Blandin and Nuhn.

Mucocele of Blandin and Nuhn may be located anywhere on the ventral surface of the tongue. Jinbu *et al.*<sup>6</sup> reported that, of the 26 cases patients in their study, 17 (65.4%) of lesions were located in the ventral tip of tongue while 9 (34.6%) occurred midway between the tip and the root of tongue. They also noted that 19 cases (73%) occurred in the midline while 7 occurred lateral to the midline. The majority of cases, 19 (73%), presented as polypoid, fluid filled, masses. The laterally positioned lesions presented a smooth raised appearance while the midline lesions were more commonly polypoid.<sup>6</sup>

The current series reports 5 cases of mucocele of the glands of Blandin and Nuhn. Four presented clinically as mucocele and one case (case 3) as a fibroepithelial polyp. Of these, four presented as sessile lesions and 1 (case 4) presented as a pedunculated lesion. There was male to female ratio of 3:2 and none of the patients had any history of trauma at the initial presentation. However, in case 3, the mucocele occurred after excision of the fibroepithelial polyp and therefore can be considered as traumatic in origin, albeit, iatrogenically.

The histology reports of the cases indicated that four of the five cases were extravasation mucocèles since there

was no epithelial lining while one case, involving a 6-year old female patient (case 2), was reported as true cyst lined by flattened epithelial cells with minor salivary gland tissues showing acini and duct suggesting that it may have been a retention mucocele. This is unusual since it has been reported that retention mucocèles are usually found in the palate and the floor of the mouth, and occur in much older patients compared to extravasation mucocèles.<sup>8</sup>

Treatment of choice is surgical excision together with the associated glands to avoid recurrence. Marsupialisation of these mucocèles is very likely to result in recurrence and should be avoided. Ellis *et al.*<sup>10</sup> reported a case of mucocele of Blandin and Nuhn, which recurred twice after marsupialisation, and biopsy after one week and 11 days, respectively and was only satisfactorily treated by complete excision.

Sugerma *et al.*<sup>12</sup> reported that one of their 5 cases, recurred and had to be re-excised. Jinbu *et al.*<sup>6</sup> reported 2 recurrences out of 26 cases, which were successfully treated by a deeper excision. So far none of the cases of mucocele reported here has recurred following excision. Alternative modes of treatment include intralesional corticosteroid injections and cryotherapy.<sup>8, 14, 15</sup>

In our report, one case (case 3) had initially presented with a fibroepithelial polyp but recurred, iatrogenically, as a mucocele and was excised together with the surrounding minor salivary glands with no further recurrence. It is therefore our opinion that during excision of all benign lesions in the ventral surface of the tongue, all the minor salivary gland tissues in the surgical field may be electively excised to avoid an iatrogenic occurrence of a mucocele by salivary gland tissue left behind.

We also believe that since most of the histological features of the mucocèles of

Blandin and Nuhn suggests extravasation phenomenon even though trauma is not reported in all cases, it is possible that the occurrence of such trauma was so minor as to escape the patients' attention.

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