Mucocele of the Glands of Blandin-Nuhn in Children: A Clinical, Histopathologic, and Retrospective Study

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Abstract

Background: The anterior lingual salivary glands (glands of Blandin–Nuhn) are mixed mucous and serous glands that are embedded within the musculature of anterior tongue ventrum. **Aim:** The present study was designed to describe the clinical and histopathological features. **Material and Methods:** We investigated the clinical and histopathologic features of 30 cases of mucocele of glands of Blandin–Nuhn. All the cases were seen in the department of pedodontics. **Results:** All the lesions were located on the ventral surface of the tongue. Lesions were situated at the midline in 24 patients and laterally in 6 patients. All the lesions were surgically treated. There was female predominance, and most patients were younger than 15 years. **Conclusion:** Histopathological examination showed extravasation type of mucocele, suggesting that trauma may be a frequent initiating factor. All the patients were younger than 15 years.

Keywords: Children, glands of Blandin-Nuhn, mucocele

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Introduction

A mucocele is a benign, common, mucus-containing cystic lesion of the minor salivary glands in the oral cavity. [1] It is nodular, and/or vesiculobullous lesion, bluish red in color and flabby in consistency. [1] Two types of mucoceles occur based on the histologic features of the cyst wall: a extravasation mucocele formed by mucous pools surrounded by granulation tissue (92%) and a retention mucocele with an epithelial lining (8%). [2,3] These two forms of oral mucocele show some differences in the age of occurrence and anatomic distribution. Although minor salivary glands are found in most parts of the oral cavity except the gingival, the prevalence of mucocele varies depending on the specific location. The lower lip is the most common site for mucocele, and

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mucocele of glands of Blandin-Nuhn has been reported as unusual.^[4]

Mucoceles represent the 15th most common oral mucosal lesion, with a prevalence of 2.4 cases per 1000 people. Mucoceles appear with higher frequency in children than in adults and are associated with traumatic injuries. [4,5] Mucoceles of the anterior lingual salivary glands (glands of Blandin and Nuhn) are relatively uncommon, with only isolated case reports and case series in the literature. This type of mucocele represents an estimated 2–8% of all mucoceles. [5]

Mucocele of glands of Blandin–Nuhn was considered an uncommon lesion. [6-8] Out of the 400 cases of mucocele reviewed by Harrison, [8] only 9 cases were on the tongue. Because the mucocele of glands of Blandin–Nuhn have not been fully characterized and conflicting results were obtained from the previous studies, the current study of 30 cases was compiled to describe the patient characteristics, and clinical and histopathological features.

Materials and Methods

Oral and written consents were obtained from the patients or their parents. The study was approved by

the Ethical Committee of Rural Dental College.

Study population comprised all the patients diagnosed with mucocele of glands of Blandin and Nuhn reporting to the Department of Pediatric Dentistry, Rural Dental College, Loni, Maharashtra, India. Cross-sectional retrospective study of 164 cases of mucocele was carried out from 2005 to 2010 and 30 cases of glands of Blandin and Nuhn were selected for the analysis. The clinical and histopathological records were compiled at the department of Pediatric Dentistry and checked by routine data search. The data collection proforma was designed to include variables such as socio-demographic characters of patients, etiologic factors, oral habits, etc. The purposive random sampling method was applied to collect the data under this study.

Patients included in the study were clinically diagnosed cases of mucocele of glands of Blandin and Nuhn and were aged between 1 and 15 years. The lesions located on the tip and ventral surface of tongue were part of the study. Cystic swellings on the bilateral sides of the ventral surface of the tongue were considered. All the patients included were systemically healthy without any underlying disease.

All other mucoceles other than those on the tip and ventral surface of the tongue were excluded from the study. Any lesions on the dorsal surface of tongue were not included as well. Patients who were systemically ill were also excluded from the study.

Statistical analysis

Statistical analysis software SYSTAT for Windows version 12 was used to calculate or determine the absolute and relative frequencies of the qualitative variables, proportions/percentage.

Results

All patients with mucocele (164 cases) were treated at the Department of Pediatric Dentistry, Rural Dental College, Loni during the 5-year study period. The most frequent site of occurrence was the lower lip (92 cases, 56.1%), followed by the tongue [30 cases, 18.3%; Table 1]. All the mucoceles of the tongue arose on the ventral surface and were diagnosed as mucocele of glands of Blandin and Nuhn.

Of the 30 cases, 23 cases were females and 7 cases were males, for a female-to-male ratio of 3:1 [Table 2]. The age range was from 1 to 15 years, with an average age of 8.5 years. In 28 patients (93.3%), lesions were clinically diagnosed as mucocele and two lesions were suspected as papilloma. All the patients were examined in our department by the Pedodontist.

The interval from the time the patient first noticed the lesion to professional evaluation ranged from 1 week to 12 months, with an average interval of 2 months.

Lesion dimension ranged from 0.2×0.2 to 1.1×1.2 cm, with average dimension of 0.6×0.6 cm.

Of the 30 patients, mucocele were observed at the ventral tip of the tongue in 22 patients (73.3%), and the lesion occurred midway between the tip and the root in 8 patients (26.7%). Lesions were located at the midline in 24 patients (80%), with all the lesions situated near the tip located at the midline. Six lesions occurred laterally to the midline [Table 3].

Twenty-three lesions (76.6%) clinically presented as a polypoid mass, and these lesions were exophytic, nonulcerated, and fluid filled. Five lesions (16.7%) demonstrated a smooth, raised, bluish appearance. Laterally positioned lesions tended to show a smooth, raised appearance, whereas midline lesions were commonly polypoid. Only two lesions (6.7%) appeared papillary. After obtaining submucosal injection of 2% lignocaine with 1:100000 epinephrine, all the lesions were surgically unroofed and the entire glandular tissue was removed, leaving a firm muscular base. Recurrence was observed in three cases. However, healing was uneventful, with no further recurrence after repeat excision in these three patients.

On histopathologic examination, all lesions were diagnosed as extravasation type of mucocele. The walls of most of the specimens were composed of granulation tissue [Figure 1].

Table 1: Anatomic distribution of mucocele					
Location	No. of Cases				
Lower lip	92 (56.1%)				
Tongue	30 (18.3%)				
Floor of the mouth	25 (15.2%)				
Buccal mucosa	13 (7.9%)				
Upper lip	3 (1.8%)				
Palate	1 (0.7%)				
Total	164 (100%)				

 Table 2: Age distribution of patients with mucocele of glands of Blandin–Nuhn

 Age (years)
 No. of cases

 1-5
 4 (13.3%)

 6-10
 12 (40%)

 11-15
 14 (46.7%)

 Total
 30 (100%)

Table 3:Mucocele of glands of Blandin-Nuhn: Patient features							
Case	Sex	Age(years)	Dimensions	Location	Features		
1	Female	8	3×3	Tip	Polypoid		
2	Female	4	2×3	Lateral	Polypoid		
3	Female	12	4×5	Lateral	Raised		
4	Female	13	12×11	Tip	Polypoid		
5	Male	10	5×5	Lateral	Polypoid		
6	Female	9	4×5	Tip	Polypoid		
7	Female	12	6×4	Lateral	Raised		
8	Female	5	3×5	Tip	Raised		
9	Female	11	10×8	Tip	Polypoid		
10	Female	8	5×6	Tip	Polypoid		
11	Female	4	5×4	Tip	Polypoid		
12	Female	13	7×8	Tip	Polypoid		
13	Male	11	8×7	Tip	Raised		
14	Female	10	5×5	Tip	Polypoid		
15	Female	11	6×6	Tip	Polypoid		
16	Male	10	2×2	Tip	Polypoid		
17	Female	5	3×5	Tip	Polypoid		
18	Male	8	4×2	Tip	Polypoid		
19	Female	12	3×3	Lateral	Raised		
20	Female	7	2×3	Tip	Papillary		
21	Male	13	3×3	Lateral	Polypoid		
22	Male	13	2×4	Tip	Polypoid		
23	Female	11	5×5	Tip	Papillary		
24	Female	11	5×6	Tip	Polypoid		
25	Female	8	4×4	Tip	Polypoid		
26	Female	12	5×5	Tip	Polypoid		
27	Female	7	3×4	Tip	Polypoid		
28	Female	7	3×3	Tip	Polypoid		
29	Female	12	4×4	Tip	Polypoid		
30	Female	6	2×2	Tip	Polypoid		

Discussion

There are three distinct sets of lingual, oral minor salivary glands, namely the glands of von Ebner, glands of Weber, and glands of Blandin-Nuhn. [6] The glands of von Ebner open at the base of the grooves that surround the circumvalate papillae and the base of clefts of foliate papillae. [7] The glands of Weber are pure mucous glands. They are abundant along the lateral border of the tongue and open into the crypts of the lingual tonsils. [7] The Blandin-Nuhn glands are a compact group of small mixed mucous and serous salivary glands, situated on both sides of the midline of the ventral tongue surface, embedded in the muscles of the ventral aspect, and recovered by a thin layer of mucosa.^[7] Five to seven small ducts open in the oral cavity medial to the plica fimbriate on the undersurface of the tongue, laterally to the lingual frenum.[7] Each gland was about 8 mm in width and 12-25 mm in depth. The nature of secretory products of these glands have not been precisely determined, but the glands have been histologically described as consisting of seromucous acini in their anterior portion and of mucous acini capped seromucous demilunes in their posterior portion. [7]

Mucoceles of the glands of Blandin-Nuhn have been considered to be uncommon. [1,2,7,8] Out of the 400 cases of mucocele reviewed by Harrison,[8] only 9 cases were on the tongue. Even though it is described as a rare lesion, some authors have several cases reported. According to Jinbu et al., [4] Blandin-Nuhn mucoceles comprise 9.9% of all oral mucoceles studied by them. In a Brazilian study with pediatric patients up to 15 years old, mucoceles in the ventral aspect of the tip of the tongue counted for 8.3% of the cases. [9] According to Saza et al., mucoceles of glands of Blandin-Nuhn comprised 9.6% of all the mucoceles.[10] Kurozu reported the proportion of oral mucoceles that were of glands of Blandin-Nuhn as 10.3%.[11] In our study, mucocele of glands of Blandin-Nuhn made up 18.3% of all oral mucoceles. Therefore, mucoceles of glands of Blandin-Nuhn should not be considered rare.

In this study, 23 of 30 patients were females; most of the patients were aged between 11 and 15 years. This is in

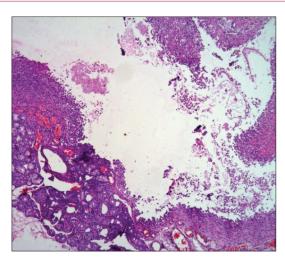


Figure 1: Histo pathologic examination of mucocele of glands of Blandin–Nuhn. Pooling of mucus with absence of epithelial lining. (Hematoxylin-Eosin Stain, 10× magnification)

agreement with the study carried out by Jinbu *et al.*,^[4] where adolescent females showed most of the lesions on the tongue. Harrison^[8] reported that in younger patients, most mucoceles were of extravasation type and the lower lip was the most commonly affected site. Traumatic injury to a duct or ducts with partition of this structure was the most likely etiologic factor leading to the development of these lesions,^[7] most probably by the frequent oscillation of the tongue.

In contrast to extravasation mucoceles, retention mucocele are frequently found in older patients and only rarely in the lower lip. It arises as a result of partial obstruction of the salivary duct by a sialolith or mucous plug. A continuous flow of gland secretions increases luminal pressure, resulting in the dilatation of duct.^[8]

Of the 30 cases, 28 lesions were clinically diagnosed as a mucocele, with 2 lesions diagnosed as papilloma. The lesions arising at the midline were typically polypoid, whereas those arising lateral to the midline presented as a raised mass. In the present study, diagnosis of the mucocele of the glands of Blandin and Nuhn was made by the anatomical site, size, flaccid nature, and asymptomatic nature of the lesion. Based on these findings we ruled out the varied differential diagnoses. Sugerman *et al.*,^[8] reported that mucoceles of the glands Blandin and Nuhn may clinically resemble a vascular lesion, pyogenic granuloma, polyp, or squamous papilloma. These mucoceles, if left untreated, the lesions may grow in size, can cause discomfort, and interfere with speech and mastication.

When mucoceles are present on the tongue, they can be easily traumatized and become a strong source of irritation and annoyance to the patient. Therefore, they should be surgically removed. Usually, the surgical excision includes the servicing mucous glands with evacuation of its contents. Baurmash^[1] showed that there are three possible approaches to the management of mucoceles. The small lesion can be completely excised, making sure to include the associated salivary gland tissue as well as any marginal glands before primary closure.[1] Large mucoceles are best treated with an unroofing procedure (marsupialization).[1] The third procedure involves the dissection of the mucocele along with the servicing mucous glands, which is usually done for moderate-sized lesions.[1] In addition, larger lesion may also be managed by marsupialization, [1,12] cryosurgery, [2] laser ablation, [13] and micro marsupialization. [14] Use of steroid injections is an alternate to surgery. [15] Some authors have suggested filling of the cystic cavity with rubber impression material presurgically, improving the visual access for surgical excision.[16]

In our study, recurrence was seen in three cases after the surgical excision. Hence we recommend that mucoceles affecting the glands of Blandin and Nuhn should be removed up to the muscle plane, including the small glands found in the surgical field, to avoid recurrence. Therefore, parental attention is necessary for the young children to prevent the traumatic injuries to tongue. The older children can wear mouthguards as a preventive appliance therapy to prevent injuries during sports. The lesion, which showed recurrence in our study, can be prevented by removing the glands of Blandin–Nuhn and other minor glands up to the muscle plane.

The histopatholocial examination of the mucoceles of the glands of Blandin–Nuhn reported in the literature, [1,17] as well as in our cases, revealed that they consist in mucus extravasation phenomenon with no epithelium lining the mucin collection. Mucocele walls consisted of densely packed granulation tissue, which contains a variable number of cells, most of them being leukocytes and phagocytes. This feature is strongly related to the fact that the extravasation-type lesion is more common in young patients.

Conclusion

In this study, mucoceles of glands of Blandin and Nuhn predominate the female subjects in the second decade of life, with the midline of the tongue being the most frequently affected site. In our series, we used surgical excision only because we treated primarily small to medium-sized lesions. In the present study, all mucocele were of extravasation type and most patients were younger than 15 years. This suggests that traumatic injury to the duct is the most likely etiologic factor leading to development of mucocele of glands of Blandin–Nuhn.

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