Case Report

Mucocele of the tongue: A case report and review of literature

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Abstract

Mucoceles are common cystic lesions in the oral cavity. It may occur in different locations in the oral mucosa due to trauma or obstruction of minor salivary gland ducts with the lower lip as the predominant site. However, mucoceles located on the ventral surface of the tongue originating from the anterior lingual salivary glands are extremely rare and often overlooked during screening procedures because of their asymptomatic nature. Here, we report an interesting case of mucocele on the anterior ventral surface of the tongue in an 11-year-old female based on the clinical and histopathological diagnosis. Moreover, mucoceles should be considered as one of the differential diagnoses while evaluating a growth involving the ventral surface of the tongue in young female children.

Keywords: Histopathology, mucocele, tongue

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INTRODUCTION

Mucoceles are formed in the oral mucosa owing to the accumulation of saliva, thereby leading to the swelling of the involved area.[1] They generally appear as soft, asymptomatic swellings with a color that can range from deep blue to the color of the oral mucosa.[2] A characteristic finding of the lesion involves its alternate regression and recurrence due to the cystic cavity being subjected to the rupture and re-aggregation of saliva.[3] Post rupture, they create painful ulcerations that heal within days. They can develop anywhere in the human body[4-8] but are particularly very common in the oral mucosa and the frequency of progression of their occurrence in the oral cavity surpasses other areas in the body. However, it is difficult to estimate the incidence of this lesion owing to the vast number of cases that do not get referred for a histopathological examination.[9]

Mucoceles are classified into two subcategories: (a) mucus extravasation cyst reports an incidence of 90% and resembles a pseudocyst due to the absence of the epithelial lining.[3] It occurs post trauma to the salivary gland, thus leading to leakage and pooling of saliva into the surrounding tissues, whereas (b) mucus retention cyst[10]

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is a true cyst that is lined by epithelium and is formed due to the obstruction of the salivary duct from insisted debris or calculi or from kinking of a tortuous duct and reports an incidence of 10%.[3] The mucin spillage is often in conjunction with trauma; however, most cases do not report any history of trauma. Mucoceles show no particular gender predilection, but studies have reported a higher incidence in the second and third decades of life. Moreover, mucus extravasation cyst tends to occur at a younger age in comparison to its counterpart, mucus retention cyst.

Mucoceles most frequently appear on the lip, especially the lower lip, since it can get injured easily, followed by the floor of the mouth, the ventral tongue and the buccal mucosa.[1] Standish and Shafer reported that nearly 45% of the mucoceles occurred on the lower lip and they are less frequently observed on the anterior ventral surface of the tongue where the Blandin–Nuhn seromucous salivary glands exist, in close proximity to the lingual tonsils, the glands of Weber and finally in the periphery of circumvallate papillae and the base of the clefts between the foliate papillae and the serous salivary glands of von Ebner. Pathologies of the oral cavity tend to show the greatest predilection in children, of which salivary gland pathology is most commonly occurring with a percentage of 87.5% of mucous extravasation cysts. Despite numerous reports of mucoceles originating on the ventral surface of the tongue, there have been only scarce reports of such a cyst forming on the dorsal surface. This article describes a mucous extravasation cyst developed on the ventral tongue of a female.

CASE REPORT

A 11-year-old healthy female patient presented with a painless swelling under her tongue for 1 month with the difficulty of eating and speaking. Intraoral examination revealed a painless, fluid-filled, soft, solitary flaccid growth measuring about 10 mm × 8 mm on the anterior ventral surface of the tongue with an intact overlying mucosa [Figure 1]. She had an unremarkable medical history. There was no history of trauma. Extraoral examination did not show any swelling or lymphadenopathy. Excision of the lesion was performed under local anesthesia followed by histopathological evaluation. Histopathological examination revealed an amorphous material surrounded by granular connective tissue with and without an epithelial lining on the periphery [Figure 2a and b]. There were numerous foamy histiocytes and some monomorphonuclear leukocytes accompanying the mucus in the cavity [Figure 2c]. Based on the clinical and histopathological findings, the final diagnosis of mucocele extravasation cyst was established. The postoperative healing was uneventful and the patient was examined on a regular follow-up basis, exhibiting no signs of recurrence during the entire period of a year.

DISCUSSION

Here, we present a case of a mucocele occurring along the midline on the ventral surface of the tongue in an 11-year-old female. The human tongue consists of three groups of minor salivary glands such as the glands of Weber located along the border of the lateral tongue, the glands of von Ebner surrounding the circumvallated papillae and glands of Blandin and Nuhn. These glands are about 8 mm in width and 12–25 mm in depth and consist of five to seven small ducts that open in the oral cavity medial to the plica fimbriate on the undersurface of the tongue, laterally to the lingual frenum. Moreover, they extend laterally and posteriorly from the midline of the tongue, forming a mass resembling a horseshoe with its opening pointing toward the root of the tongue, which enabled our diagnosis. The nature of secretory products of these glands has been histologically described as consisting of seromucous acini in their anterior portion and of mucous acini capped seromucous demilunes in their posterior portion. There have been eight previously identified cases of the mucocele of the tongue, of which four cases[11-14] have been similar to ours (mucus extravasation cyst) and four were mucus retention cyst[15-18] [Table 1].

Mucoceles of the glands of Blandin–Nuhn have been considered to be very rare; in fact, Harrison[27] reported only nine cases of mucocele of the tongue out of a total of 400 mucoceles in the oral cavity, whereas Jinbu *et al*.[28] reported a total of 9.9% Blandin–Nuhn mucoceles, while Saza *et al*.[29] reported 9.6% of mucoceles of glands of Blandin–Nuhn and Kurozu[30] reported the proportion of oral mucoceles that were of glands of Blandin–Nuhn as 10.3%. This implies that tongue mucoceles are not

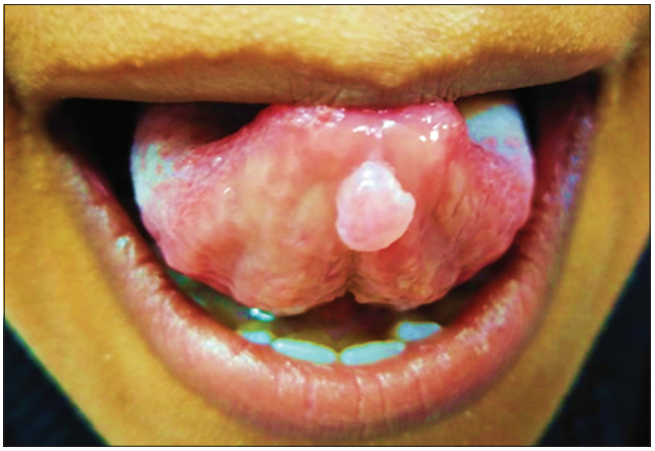

Figure 1: Intraoral examination showing fluid-filled, soft, solitary flaccidgrowth on the anterior ventral surface of the tongue

rare but in fact ignored due to nonreferral or skipped diagnosis.

The diagnosis of mucoceles of the Blandin–Nuhn glands stems from their clinical characteristics, although they may resemble a vascular lesion, pyogenic granulomas, polyps, or squamous papillomas. For this reason, it is important to raise awareness among the medical population of this pathology since medical practitioners are unaware of it and can often be misdiagnosed as a bullous lichen planus and mucous membrane pemphigoid.

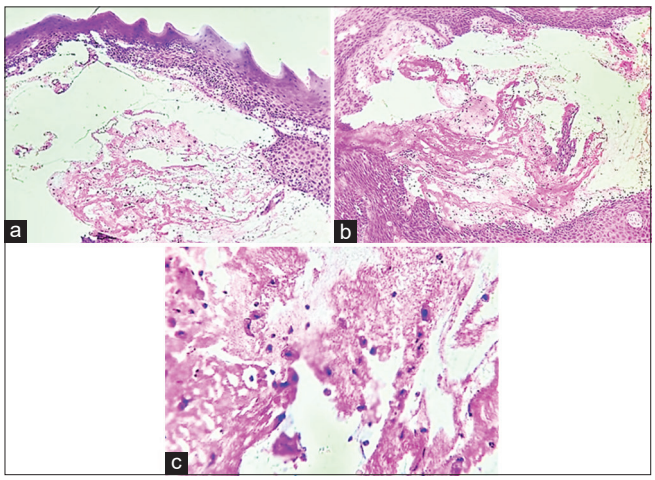

Figure 2: (a and b) revealed an amorphous material surrounded by granular connective tissue with and without an epithelial lining on the periphery, respectively (H & E*, ×100). (c) Shows foamy histiocytes and some monomorphonuclear leucocytes accompanying the mucus in the cavity (H & E*, ×400). *Hematoxylin and eosin stain

Menta *et al*.,[31] Yamasoba *et al*.,[32] and Oliveira *et al*.[19] reported that more than 65% of their patients with mucoceles were <20 years of age, which was in conjunction with our patient who was 11 years old. Oral mucoceles rarely present symptoms but in extreme cases can cause discomfort, interference with speech, mastication, swallowing and external swelling in case of large mucoceles like our case.[20] The color of mucoceles ranged from deep blue to the normal color of the oral mucosa (pink). The deep blue color results from tissue cyanosis, vascular congestion associated with the stretched overlying tissue and the translucency of the accumulated fluid beneath which was in accordance with the studies of Jani *et al*.[3] Joshi *et al*.[21] reported that mucoceles of the glands of Blandin and Nuhn show a predilection toward females in the second decade of life, with the midline of the tongue being the most frequently affected site which was in accordance with our case where we diagnosed a mucocele in an 11-year-old female on the midline of the tongue.

Table 1: Previously reported mucocele of the tongue in the English-language literature

Author	Year	Age/sex	Chief complaint	Location	Clinical findings	Investigations	Histological diagnosis
Yoshikawa[18]	1994	22/female	Swelling on the tongue	Dorsal midline	Elastic, no fluctuation 32 mm	MRI; HPE	MRC
Zancope[14]	2009	6/female	Lump in the throat, difficulty in swallowing	Posterior midline	Nodule/1.5 cm	Videolaryngoscopy; HPE	MEC
Acar[15]	2010	37/female	Progressive dysphagia	Tongue root extending to plica	Painless swelling 2 cm	Cystic fluid aspiration; HPE	MRC
Wong/Chung[17]	2014	Newborn male	Breastfeeding problems	Anterior dorsal midline	Bluish mass 2 cm	Cystic fluid aspiration; HPE	MRC
Hur 2[11]	2016	32/male	No symptoms	Tongue midline	2.8×2.5×2.8	Laryngoscopy; CT; MRI; Cystic fluid aspiration; HPE	MEC
Titsinides[13]	2018	74/female	Painless swelling	Middle dorsal midline	Soft mass covered by mucosa 1.5 cm	Thyroid hormone tests; Ultrasonography; Cystic fluid aspiration; HPE	MEC
Jose[16]	2018	7/female	Swelling in relation to the ventral surface of the tongue for 3 months	Ventral surface of the tongue	pedunculated growth of size 10 mm×5 mm	HPE	MRC
Pandey[12]	2018	13/female	Swelling on the lower surface of the tongue, which appear about one month back	Ventral surface of the tongue	Bluish-red, nontender fluid-filled mass measuring about 2 cm×1 cm in size, cystic, sessile and progressive	Cystic fluid aspiration; HPE	MEC
Our case	2020	11/female	Painless swelling under the tongue for 1 month	Ventral surface of the tongue	Painless, fluid-filled, soft, solitary flaccid- growth measuring about 10 mm×8 mm on the anterior ventral surface of the tongue with an intact overlying mucosa	HPE	MEC

HPE: Histopathological examination, MEC: Mucous extravasation cyst, MRC: Mucous retention cyst

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. The histopathological examination of the mucoceles of the glands of Blandin–Nuhn[21-23] reported in the literature, 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, similar to our case. Furthermore, extravasation mucoceles seen in children may be difficult to distinguish visually from vascular lesions, pyogenic granuloma, polyps, or squamous cell papillomas. Retention mucocele, especially in older patients, must be distinguished from neoplasms of minor salivary gland origin such as mucoepidermoid carcinoma, inverted ductal papilloma, sialadenoma papilliferum, intraductal papilloma and cystadenoma.

The first study on pediatric mucoceles was published in Pediatric Dermatology in 2008.[31] Chen *et al*.[24] reported that 28.6% of all biopsies from pediatric patients showed mucoceles, which is higher than most studies. This makes it imperative for the attending pediatrician to be well informed about it and to remind the pediatric physician to devote much attention to lesions of the oral cavity in children, remind the physician that oral health is an integral part of a child's overall health. Our patient was an 11-year-old female who was a pediatric patient and was similar to cases[12,14,16,17] previously reported of the mucocele of the tongue. Often, such patients tend to visit the pediatrician first, and hence, it is necessary to educate the pediatricians about these cases to allow them to make an appropriate diagnosis or at least refer them to a dentist. As discussed previously, several studies in literature are in favor of the fact that salivary mucoceles should be considered as the most frequent oral benign lesion encountered in children. Often, as stated, diagnosis can be made during the routine pediatric intraoral examination; thus, it is essential to expand the medical and dental personnel's awareness about salivary mucoceles. To avoid mucoceles on the tongue, it is necessary for parental attention to be given to children to prevent traumatic injuries and older children can wear mouthguards as a protective appliance to prevent injuries during sports.

For superficial and miniscule mucoceles,[23] generally, no treatment is advised since they heal spontaneously within 2–3 weeks; however, mucoceles present on the tongue should be surgically excised since they can be a source of strong source of irritation and annoyance to the patient. Baurmash[25,26] proposed three possible approaches to the management of mucoceles, first being the complete excision, along with the removal of the associated salivary gland tissue, second, an unroofing procedure (marsupialization) for larger mucoceles. The third procedure involves the dissection of the mucocele along with the servicing mucous glands, which is usually done for moderate-sized lesions.[25,33] For those lesions that persist or are larger like our case, the treatment of choice is surgical excision of the mucocele as well as removal of the immediate surrounding minor salivary glands. The mucocele may recur if the involved glands are not completely resected. However, recurrence can occur and a new surgical intervention is necessary. Alternative treatment options include marsupialization, dissection, cryosurgery, carbon dioxide lasers, electrocautery, intralesional injection of sclerosing agent OK-432 corticosteroid injection and cryotherapy.[23]

CONCLUSION

Since mucoceles are often overlooked or skipped, it is imperative to include mucoceles as a differential diagnosis for any growth observed on the midline of the ventral surface of the tongue in females during the second decade of life. If ignored, it can lead to further complications like excessive growth that may impede mastication and interfere with swallowing. Superficial mucoceles may regress by themselves and generally do not require treatment; however, larger mucoceles need to be surgically excised.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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