CASE REPORT

Salivary duct cyst on lower lip: A rare entity and literature review

Ankita Tandon, Keya Sircar, Aman Chowdhry, Deepika Bablani
Department of Oral Pathology and Microbiology, Faculty of Dentistry, Jamia Millia Islamia, New Delhi, India

ABSTRACT
Mucocele forms because of salivary gland mucous extravasation or retention and is usually related to trauma in the area of the lower lips. Salivary duct cyst, however, is a type of mucous retention cyst which is almost never located on the lower lip. The aim of this paper is to report this extremely rare salivary duct cyst present on the lower lip and to critically review the literature to build important concepts that would help clinicians in the diagnosis and treatment of this pathology.

Key words: Mucocele, mucous extravasation cyst, mucous retention cyst, salivary duct cyst

INTRODUCTION
Salivary duct cysts, of different types, clinically referred to as mucoceles comprise 6-9% of salivary gland diseases.[1,2] Mucoceles (muco meaning mucous and cele meaning cavity), by definition, are cavities filled with mucous. When they occur in the floor of the oral cavity, they are called ranulas (rana = Frog and ula = Little) because the swelling resembles the vocal or air sacs of the frog. Mucous is the exclusive secretory product of the accessory (minor) salivary glands and the more prominent product of the sublingual (major) salivary gland.[3] These lesions can be superficial (located directly under the mucosa), classic (in the upper submucosa) or deep (in the lower corium).[3]

The term mucous extravasation phenomenon (cyst) is used when mucous has been extruded into the connective tissue envelope, while the term mucous retention cyst is used to describe a cyst with retained mucin that is lined with ductal epithelium which may have undergone squamous or oncocytic metaplasia.[2]

Salivary duct cysts develop from dilatation of salivary gland ducts but are distinguished from mucous retention cysts by the fact that they do not typically contain pools of mucin.

Mucous extravasation cysts occur most commonly on the lower lip, but salivary duct cysts only rarely occur in the minor salivary glands of the lip.[4] Salivary duct cysts usually occur in people over 30 years of age and have equal predilection for males and females.[4]

CASE REPORT
A 15-year-old male patient visited the Dept of Oral Medicine Diagnosis and Radiology with chief complaint of swelling on the lower lip since 3 months. On examination, a 5 × 5 mm solitary, bluish white coloured swelling on left side of lower lip was noted [Figure 1]. The swelling was nontender and soft on palpation. Provisional diagnosis of mucocele was made and excisional biopsy of the lesion was performed. The tissue fixed in formalin was received in the department of oral pathology and microbiology. The H and E -stained sections revealed stratified squamous epithelium overlying fibrocellular connective tissue stroma. The deeper connective tissue showed a dilated minor salivary gland duct. The epithelial lining of the duct appeared bilayered and revealed oncocytic metaplasia. Dense chronic inflammatory infiltrate composed chiefly of lymphocytes was seen around the dilated duct. The predominantly mucous salivary gland acini present in deeper connective tissue showed normal architecture with no inflammatory cell infiltration around them [Figures 2-4]. The connective tissue also revealed endothelium lined blood vessels of varying caliber.

The case was diagnosed as salivary duct cyst and the patient was recalled after 6 months or earlier in case of a recurrence.

DISCUSSION AND LITERATURE REVIEW
PubMed, Google Scholar and Medline databases were searched and only six articles were retrieved using “salivary
duct cyst” as the keyword. A systematic review of literature for mucocele, mucous extravasation, mucous retention and salivary duct cyst has been summarized and discussed below.

Nomenclature

Review of the literature reveals large number of terms which have been used to refer to this group of lesions.

Hamperl, as suggested by Seifert G et al.[5] introduced the term “mucus granuloma” (“Schleimgranulom”) which is identical to what later was known as “extravasation mucocele”.

Bhaskar et al.[6] and Chaudhry et al.[7] referred mucoceles as the retention cysts.

Tal et al.[2] stated that cystic lesions of salivary glands have been collectively referred to as mucoceles. Later they suggested that based on pathogenesis, two major groups can be delineated from among these differently named lesions.

Seifert[1] described that salivary duct cysts are entities distinct from mucous retention cysts.

Dent et al.[8] believed that those mucoceles which resulted from mechanical trauma to the excretory duct of the salivary glands, causing duct transection or rupture, with consequent extravasation of mucin to the connective tissue stroma must be referred to as mucus extravasation phenomenon. In addition, when mucus is retained in the duct and/or acinus as a result of duct obstruction it must be referred to as mucus retention phenomenon.

Shear and Speight[9] clarified that the term “mucus extravasation cyst” is reserved for those lesions in which...

---

**Figure 1:** 5 × 5 mm solitary, bluish white coloured swelling on left side of lower lip

**Figure 2:** Dilated minor salivary gland duct surrounded by predominantly mucous acini with normal architecture (H & E stain ×50)

**Figure 3:** Dilated ducts with papillary projections towards the duct lumen (H & E stain, ×100)

**Figure 4:** Papillary projection showing bilayered lining of dilated duct with eosinophilic granularity of cells (H & E stain, ×400)
mucus has extravasated into the connective tissues and in which there is no epithelial lining. The term “mucous retention cyst” is employed to describe mucocoeles that result from dilatation of the ducts and which are lined by epithelium. Salivary duct cysts on the contrary are similar in pathogenesis and histopathological features to retention cysts of minor salivary glands.

**Etiopathogenesis**

Two mechanisms have been proposed to distinguish between mucoceles and mucous retention cysts. Partial obstruction of a salivary duct due to inflammation, calculi, or tumor growth,[10,11] with subsequent duct dilatation (but not rupture) will give rise to a mucous retention cyst, with epithelial lining of the cyst wall.

The presence or absence of epithelium in a mucous cyst (extravasation/retention) appears to be related to the pathogenesis. Robinson L.[12] concluded that ductal dilatation occurs consequent to ductal obstruction. The dilated ducts may then fuse together to form an epithelium lined cystic cavity.

Mucoceles may develop as a result of a traumatic defect or severance of salivary duct and escape of saliva into the tissue. Therefore, Standish and Shafer expressed the opinion that rupture of an excretory duct allows for the escape of mucus into adjacent tissues, but they also considered the possibility of “aneurysmal dilatation of a partially occluded duct”.[12]

Mucoceles which do not undergo spontaneous resolution are those in which constant trauma and the accumulation of mucus did not lead to the destruction of the adjacent glandular parenchyma. The autolysis of mucoceles may be related to the degeneration and breakdown of the adjacent acini due to enzymatic digestion, not only because of the action of polymorphonuclear cells and macrophages but also due to the accumulated salivary secretion.[13]

A number of authors have attempted to clarify the pathogenesis of mucous retention cysts, relating them to spontaneous changes in the eosinophilic-oncocytic epithelium, or describing them as a cystic form of papillary cyst adenoma. Other authors consider oncocytic metaplasia in retention cysts to be the response to partial duct obstruction.[10]

Mucoceles may develop as a result of spontaneous secretion producing a small but permanent increase in luminal pressure which leads to duct dilatation. Southam suggested that salivary ducts lined by oncocyte-like cells may undergo spontaneous cystic change to produce retention mucoceles. However, strongly eosinophilic cells which could be oncocyes have been reported.[14] It has also been suggested that oncocytic metaplasia in salivary gland ducts may occur as a response to partial obstruction.[14]

The etiopathogenesis of our case may be partial obstruction of the minor salivary gland duct as indicated by the oncocytic metaplasia seen histopathologically. However, the cause for the obstruction such as stricture, mucous plug, or a sialolith was not confirmed surgically.

**Clinical appearance**

Mucus retention cysts are rarely found on the lower lip and are more common on the upper lip, whereas mucous extravasation cysts are common on the lower lip followed by buccal mucosa, floor of the mouth, tongue, retromolar area, palate and upper lip.[4] The occurrence of mucous retention cysts peaks in the 7th and 8th decades,[4,16,14] whereas mucous extravasation cysts affect both genders in all age groups, with peak incidence between 10 and 29 years.[11] On the contrary, our case was unique in this aspect as it presented in a 15-year-old male patient with the lesion on the lower lip.

Mucoceles usually appear as an asymptomatic nodule, with a normal or bluish color.[4,17] The deep blue color results from tissue cyanosis and vascular congestion associated with the stretched overlying tissue and the translucent character of the accumulated fluid beneath.[15] It is fluctuant and movable because of its mucinous contents. The diameter may range from a few millimetres to a few centimeters.[16,17] The consistency is typically soft and fluctuant in response to palpation. Mucoceles are usually asymptomatic, though in some patients they can cause discomfort by interfering with speech, chewing, or swallowing.[17] If left without intervention, an episodic decrease and increase in size may be observed, corresponding to rupture and subsequent mucin production.[4,15]

**Diagnosis**

The case history and an objective examination of the lesion are crucial for diagnosing mucoceles correctly. In particular cases, the diagnosis may require traditional radiography, ultrasonography, or advanced diagnostic methods like computed tomography and magnetic resonance imaging. Ultrasonography shows mucoceles as cystic masses that sometimes contain fibrillar processes produced by fibroblasts seen in minimal numbers within the mucinous area (septa). Fine-needle aspiration is a useful diagnostic technique for evaluating patients with salivary gland nodules and enlargement. Differentiating between mucoceles and vascular lesions preoperatively is very important, because large
angiomas mistaken for mucoceles can result in major bleeding if removed.\textsuperscript{[15]}

A simple technique known as fine-needle aspiration biopsy (FNAB) is very helpful, especially when differential diagnosis of angiomatous lesions is involved. FNAB of mucocele often reveals abundant minor salivary gland acini with many inflammatory cells, especially histiocytes but without any epithelial component.\textsuperscript{[11]}

The clinical presentation of our case conformed to the characteristic features of mucoceles; hence, lesion was surgically excised and submitted for histopathological evaluation.

**Histopathologic spectrum**

Robinson and Hjorting-Hansen\textsuperscript{[12]} suggested three morphological patterns of mucous (extravasation and retention) cysts which although not commonly used in present times have been listed in Table 1. They also proposed that in a well-defined cyst with only a partial epithelial lining, the loss of portions of the lining epithelium could be due to atrophy resulting from increased intraluminal pressure.

Seifert et al.,\textsuperscript{[5]} analyzed 360 salivary glands cysts and concluded that the extravasation mucocele is the most frequent type of salivary cyst. As suggested by Hamprel, three stages of development can be distinguished in the pathogenesis of the mucus granulomas: An initial stage (interstitial mucus lakes), a resorption stage (mucus granulomas with macrophages, foam cells and foreign body giant cells) and a terminal stage with the development of a pseudocyst (capsule of collagen tissue, no epithelial demarcation). On the contrary, the retention mucoceles contain viscous mucous material, always possess an epithelial demarcation and as a rule, show no inflammatory reaction compared with the extravasation mucoceles.\textsuperscript{[5]}

Mucous retention cysts are lined by epithelium whose structure corresponds with the epithelium of the different segments of the salivary duct system. The epithelial lining may consist of flat duct cells similar to intercalated duct cells or of bilayered duct cells similar to striated ducts or the surrounding excretory ducts. In some areas, plump papillary projections may be seen. In other areas, epithelial transformations can be observed with squamous cell metaplasia, goblet-like cells, or clear reserve cells.\textsuperscript{[15]} Eversole has categorized these lesions as mucous retention cysts, reactive oncocyctoid cysts and mucopapillary cysts.\textsuperscript{[18]}

Mucous retention cysts which have been specified as salivary duct cysts are mostly limited by a bilayered duct epithelium, which may include some oncocytic cells. Analogous with mucous retention cysts, focal papillary projections and epithelial transformation with squamous metaplasia or goblet-like cells may be observed\textsuperscript{[1]} and the connective tissue wall of the cyst may be slightly inflamed.\textsuperscript{[15]} Seifert G (1996)\textsuperscript{[5]} reported epithelial alterations especially in salivary duct cysts of parotid gland. Characteristic cellular changes seen were epithelial metaplasia (goblet cells/clear cells/squamous cells) and focal epithelial proliferations with plump or papillary plaques projecting into the cyst lumen.

Takeda and Yamamoto (2001)\textsuperscript{[19]} reported three cases of salivary duct cyst and suggested that two cases appeared as a unicocular lesion lined by double- and multi-layered epithelium as is also seen in our case.

**Differential diagnosis**

The lip contains adipose, connective tissue, blood vessels, nerves and salivary glands, so pathosis of any of these tissues is possible. Daley reviewed the clinical differential diagnosis of a swelling of the upper lip, listing mucocele, fibroma, lipoma, mucus retention cyst, sialolith, phlebolith and salivary gland neoplasm as possibilities.\textsuperscript{[4,20]}

Palpation can be helpful for a correct differential diagnosis. Lipomas and tumors of minor salivary glands present no fluctuation, while cysts, mucoceles, abcess and

| Epithelium lining | Connective tissue | Inflammatory infiltration | Communication between duct and cystic area |
|------------------|------------------|--------------------------|------------------------------------------|
| Poorly defined cysts | No epithelium | Poorly defined laking and puddling of faintly eosinophilic material, with separation of the collagen fibers of lamina propria of mucous membrane and of the submucosa | Macrophages within lakes and many scattered eosinophils and few plasma cells | + |
| Well defined cysts (1) | No epithelium | Sharply circumscribed cavity containing a faintly eosinophilic amorphous material | Numerous vacuolated macrophages and occasional eosinophils | +++ |
| Well defined cyst (2) | Partial (several layers of flattened or polygonal epithelial cells or of stratified squamous variety) or complete (simple or pseudostratified columnar) | Well-delineated cavity | - |

---

**Table 1: Morphological patterns of mucous (extravasation and retention) cysts**

---

Journal of Oral and Maxillofacial Pathology: Vol. 18 Supplement 1 September 2014
hemangiomas do. However, cystic degeneration may also occur in neoplasms such as pleomorphic adenoma, Warthin’s tumor, mucoepidermoid carcinoma, acinic cell carcinoma and the adenoid cystic carcinoma.

The clinical and microscopic features of superficial mucoceles may be mistaken for other conditions, such as pemphigoid, bullous lichen planus, or recurrent herpes simplex virus infection. Concurrent lichenoid disorders have been reported with superficial mucoceles. Such cases may require biopsy, in addition to direct immunofluorescence studies for immunoglobulins and complement.

Mucoceles are mobile lesions with soft and elastic consistency depending on how much tissue is present over the lesion. However, both a drained mucocele and a chronic mucocele would not show significant fluctuation. Perhaps the most striking significant differential diagnosis of mucocele is from the low-grade mucoepidermoid carcinoma as both will have a bluish color and both will fluctuate on palpation.

Mucoepidermoid carcinoma as well as mucous retention cyst is more common on the upper lip, whereas mucous extravasation cyst is common on the lower lip. However, in our case, histopathological examination of surgically excised tissue revealed dilated minor salivary gland duct lined by bilayered epithelium with oncocytic metaplasia and absence of proliferating epidermoid cells thereby favoring the diagnosis of salivary duct cyst.

Treatment

The literature describes different treatment options, including cryosurgery, intralabial corticosteroid injection, micromarsupialization, marsupialization of the mucocele, conventional surgical removal of the lesion and laser ablation. As regards to treatment, resection is carried out when the lesions are multiple, recurrent, or cause patient discomfort. According to Yagüe-García et al., it must be taken into account that typical minor salivary gland mucoceles rarely resolve on their own, that is, surgical removal is required in most cases.

Other modalities of treatment have included the use of cryosurgery and sclerosing agents. A single intralabial injection with a sclerosing agent like OK-432 preceded by aspiration of the cyst fluid, causes the pseudocyst, or cyst wall to collapse and triggers a severe inflammatory reaction of the wall that results in marked fibrosis, which seals the leak in the perforated gland capsule and prevents further mucus extravasation. The result is acinar atrophy and consequent healing. However, adverse reactions to OK-432 like shock (0.05%), persistent fever (21.9%) and local inflammatory symptoms have been reported.

CONCLUSION

The clarification of terminologies regarding this group of lesions is necessary. The term oral mucocele must be referred only to mucus extravasation cysts. The salivary duct cyst represents a rare form of mucous retention cyst in which pools of mucin and extent of inflammatory infiltrate are less as compared with the conventional retention variety. The lesion in the case reported was diagnosed as a salivary duct cyst of the left lower lip, an uncommon location for this lesion. Because of the possibility that a lesion in this location might be a tumor, excision is warranted for definitive diagnosis. When possible, it is beneficial to identify and remove the glands associated with the lesion to reduce the rate of recurrence. This review also suggests that the clinician should keep salivary duct cyst as a differential diagnosis for lesion occurring in the lower lip.

REFERENCES

1. Seifert G. Mucoepidermoid carcinoma in a salivary duct cyst of the parotid gland. Contribution to the development of tumours in salivary gland cysts. Pathol Res Pract 1996;192:1211-7.
2. Tal H, Altini M, Lemmer J. Multiple mucous retention cysts of the oral mucosa. Oral Surg Oral Med Oral Pathol 1984;58:692-5.
3. Jani DR, Chawda J, Sundaragiri SK, Parmar G. Mucocele: A study of 36 cases. Indian J Dent Res 2010;21:337-40.
4. Mustapha IZ, Boucree SA Jr. Mucocele of the upper lip: Case report of an uncommon presentation and its differential diagnosis. J Can Dent Assoc 2004;70:318-21.
5. Seifert G, Donath K, von Gunther C. Mucoceles of the minor salivary glands. Extravasation mucoceles (mucus granulomas) and retention mucoceles (mucus retention cysts. HNO 1981;29:179-91.
6. Bhaskar SN, Bolden TE, Weinmann JP. Pathogenesis of mucoceles J Dent Res 1956;35:863-74
7. Chaudhry AP, Reynolds DH, Lachapelle CF, Vickers RA. A clinical and experimental study of mucocele (retention cyst). J Dent Res 1960;39:1253-62.
8. Dent CD, Svirsky JA, Kenny KF. Large mucous retention phenomenon (mucocele) of the upper lip. Case report and review of the literature. Va Dent J 1997;74:8-9.
9. Shear M, Speight P. Cysts of the Oral and Maxillofacial Regions. 4th ed. Blackwell Munksgaard, 2007.
10. Boneu-Bonet F, Vidal-Homs E, Maizurrena-Tornil A, González-Lagunas J. Submaxillary gland mucocele: Presentation of a case. Med Oral Patol Oral Cir Bucal 2005;10:180-4.
11. Ata-Ali J, Carrillo C, Bonet C, J Balaguer, M Peñarrocha, M Peñarrocha. Oral mucocoele: Review of the literature. J Clin Exp Dent 2010;2:e18-21.
12. Robinson L, Hjorting-Hansen E. Pathologic changes associated with mucous retention cysts of minor salivary glands. Oral Surg Oral Med Oral Pathol 1964;18:191-205.
13. Oliveira DT, Consolaro A, Freitas FJ. Histopathologic spectrum of 112 cases of mucocele. Braz Dent J 1993;4:29-36.
14. Harrison JD. Salivary mucocoeles. Oral Surg Oral Med Oral Pathol 1975;39:268-78.
15. Re Cecconi D, Achilli A, Tarozzi M, Lodi G, Demarosi F, Sardella A, et al. Mucoceles of the oral cavity: A large case series (1994-2008) and a literature review. Med Oral Patol Oral Cir Bucal 2010;15:e551-6.

16. Kakarantza-Angelopoulou E, Triantaphyllou A. Mucous retention cysts of the minor salivary glands. A specific type of mucocele. Odontostomatol Proodos 1989;43:373-9.

17. Yagüe-García J, España-Tost AJ, Berini-Aytés L, Gay-Escoda C. Treatment of oral mucocele-scalpel versus CO₂ laser. Med Oral Patol Oral Cir Bucal 2009;14:e469-74.

18. Carlson ER, Ord RA. Textbook and Color Atlas of Salivary Gland Pathology: Diagnosis And Management. Blackwell Munksgaard. p. 91-108.

19. Takeda Y, Yamamoto H. Salivary duct cyst: Its frequency in a certain Japanese population group (Tohoku districts), with special reference to adenomatous proliferation of the epithelial lining. J Oral Sci 2001;43:9-13.

20. Rangeeth BN, Moses J, Reddy VK. A rare presentation of mucocele and irritation fibroma of the lower lip. Contemp Clin Dent 2010;1:111-4.

21. Khandelwal S, Patil S. Oral mucoceles-review of the literature. Minerva Stomatol 2012;61:91-9.

How to cite this article: Tandon A, Sircar K, Chowdhry A, Bablani D. Salivary duct cyst on lower lip: A rare entity and literature review. J Oral Maxillofac Pathol 2014;18:151-6.

Source of Support: Nil. Conflict of Interest: None declared.