

Mucocele of the Upper Lip: Case Report of an Uncommon Presentation and Its Differential Diagnosis

(Mucocèle de la lèvre supérieure : rapport de cas sur une présentation peu courante et son diagnostic différentiel)

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S o m m a i r e

Ce rapport décrit une lésion de la lèvre supérieure qui a finalement été diagnostiquée par examen histologique comme étant une mucocèle ou un phénomène de rétention de mucus. Le siège habituel des mucocèles est la lèvre inférieure. Ce cas illustre une présentation peu courante de la mucocèle quant aux symptômes, au siège et à la durée. On aborde et on compare les caractéristiques d'une gamme de lésions buccales afin d'aider les cliniciens à établir un bon diagnostic différentiel.

Mots clés MeSH : *biopsy; diagnosis, differential; lip diseases/pathology; mucocele/pathology*

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A mucocele is a mucus retention phenomenon of the major and, more commonly, the minor salivary glands.¹ This lesion has also been called a mucus extravasation phenomenon.² Although such a lesion can occur anywhere in the oral mucosa, the term mucocele has been applied only infrequently to lesions of the upper lip.³⁻⁵ Mucoceles are usually associated with the minor salivary glands and hence are less likely to occur on the anterior hard palate and the attached gingiva, which do not typically possess minor salivary glands.

Mucoceles are usually formed secondary to rupture of an excretory duct of a salivary gland, which leads to an outpouring of saliva into the surrounding tissues.^{2,6} The resulting pool of glandular secretion is first surrounded by inflammatory cells and later by reactive granulation tissue consisting of fibroblasts. This granulation tissue reflects an immune response (i.e., to wall off the mucin). Although there is no epithelial lining surrounding the mucin, it becomes well encapsulated by this granulation tissue and is therefore categorized as a false cyst or pseudocyst.

In contrast, a mucus retention cyst is a true cyst, lined with epithelium. This type of cyst appears to be caused by epithelial

proliferation of a partially obstructed salivary duct.⁷ Complete obstruction of a salivary duct by a calcified mass is called a sialolith, also known as a salivary calculus or stone.

Parafunctional habits such as lip biting may contribute to the lower lip being the most commonly described location of mucoceles. Cohen and others⁸ observed that, of 63 mucoceles, 82% were found on the lower lip, 8% on the buccal mucosa, 3% on the retromolar area, and 1% on the palate. The Armed Forces Institute of Pathology collected data on 2,339 cases of mucocele and found that 33.0% occurred on the lower lip, 7.7% on the buccal mucosa, 6.3% on the floor of the mouth, 6.1% on the tongue and only 0.4% on the upper lip.⁷ Curtis and Hutchinson⁹ documented a single case of mucus extravasation phenomenon of the posterior hard palate after a periodontal free gingival graft procedure. A potential source of trauma to the upper lip is surgery, such as plastic surgery for lip reduction or augmentation, but no documented cases of mucocele in conjunction with surgery have been identified.

On clinical presentation, a mucocele usually appears as an asymptomatic nodule, with a normal or bluish colour. It is fluctuant and movable because of its mucinous contents.



Figure 1: Bluish nontender nodule on the inner mucosa of the upper lip.

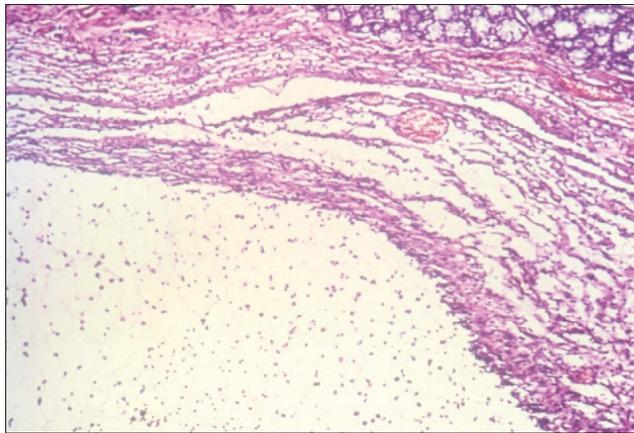


Figure 3: Magnified image of biopsy sample shows inflammatory cell infiltrate throughout the lesion and surrounding tissue. (Hematoxylin and eosin stain, original magnification $\times 100$).

The diameter may range from a few millimetres to a few centimeters. If left without intervention, an episodic decrease and increase in size may be observed, corresponding to rupture and subsequent mucin production.¹⁰

Case Report

A 57-year-old African-American man presented for initial examination. The patient's chief complaint was that "an old filling needed to be redone." He reported no dental treatment over the past 30 years, and his medical history questionnaire yielded no findings of particular relevance to this dental examination.

Extraoral examination revealed asymmetry of the upper lip to the left of the philtrum, which was normal in colour. Intraoral examination revealed a blue, fluctuant, nontender nodule measuring 10 mm \times 10 mm on the inner labial mucosa; the lesion did not blanch under digital pressure (Fig. 1). The patient had been aware of the swelling for about a year but denied any episodic increase or reduction in size. He could not recall an episode of trauma to the maxillofacial

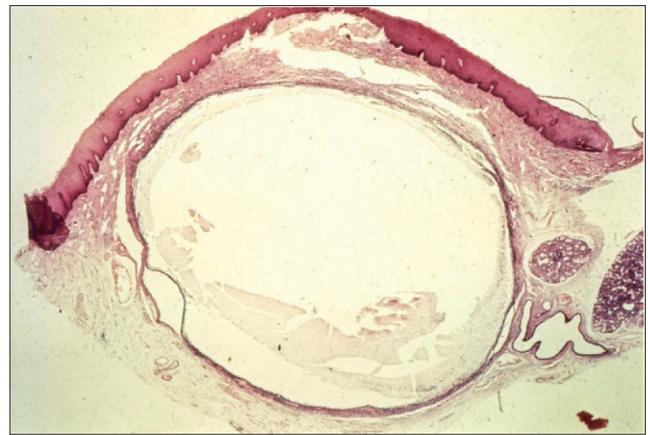


Figure 2: A pool of mucin surrounded by granulation tissue. Salivary gland tissue appears in the lower right. (Hematoxylin and eosin stain, original magnification $\times 10$.)

region. There was no evidence of calcification or retained foreign body in a radiograph of the soft tissue in this area.

Excisional biopsy was performed, and the wound was closed with 4-0 sutures (gut for deep closure and silk superficially). A mucocele was suspected. The biopsy sample was immediately fixed in 10% formalin and sent for histologic evaluation. The pathology report described the tissue as a tanned, round nodule measuring 0.8 cm in diameter, with no evidence of malignancy (Figs. 2 and 3). The definitive diagnosis was mucocele of the right upper lip. No recurrence was observed at 1-month and 3-month follow-up examinations.

Discussion

The differential diagnosis in a case such as this one should include lesions known to cause swelling of the lips. The lip contains adipose, connective tissue, blood vessels, nerves and salivary glands, so pathosis of any of these tissues is possible. Daley¹¹ 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. In patients under 20 years of age mucocele is the most common nodular swelling of the lower lip; this lesion is slightly more common in males.¹² Another common nodular lesion is fibroma, which, like the mucocele, can be initiated by trauma. Fibromas vary in consistency from soft to very firm. They are the most common intraoral soft-tissue lesion, and are seen most frequently on the lips (no distinction between upper and lower lips). Lipomas, neoplasms consisting of mature adipose tissue, are uncommon in the oral cavity, but can occur on the lips. However, many lipomas are soft and fluctuant, so when this lesion does occur, it is commonly mistaken for traumatic fibroma or mucocele.¹⁰ The lower lip is also the most common intraoral site of squamous cell carcinoma; however, unlike the previously mentioned lesions, this one presents with variations of white and red crusting and ulceration.¹⁰ Mucus retention cysts occur more commonly on the upper lip than the lower lip. The occurrence of mucus retention cysts peaks in the seventh and

eighth decades. Swelling of the upper lip is also commonly caused by sialolithiasis, mineralization that occurs in the ducts of the salivary glands. Sialoliths usually present as firm, movable nodules, most often in the fifth to seventh decades. Phleboliths, which result from calcification of intravascular thrombi, may also be considered. Both sialoliths and phleboliths, unlike mucoceles, may have an opaque appearance in radiographs.⁷

The last category of upper lip lesions mentioned by Daley,¹¹ salivary gland neoplasms, comprises a variety of conditions. Salivary duct cysts occur in the minor salivary glands of the lip, but only rarely. This type of cyst develops from dilatation of a salivary gland duct but is distinguished from a mucus retention cyst by the fact that it does not typically contain pools of mucin. Salivary duct cysts usually occur in people over 30 years of age, and there is no difference in frequency between men and women.⁵ Nasolabial cysts, which derive from epithelial remnants of the nasolacrimal duct, are nonodontogenic and are found in the upper lip or in the nasolabial fold. Cases of nasolabial cyst have been documented in people over 30 years of age, with a slight preponderance among women.¹³

The upper lip is also a common location for benign salivary gland tumours. Canalicular adenoma almost always occurs in the upper lip, most often at the midline. Its histologic appearance is dominated by double rows of columnar epithelial cells, which branch and interconnect; peak incidence is in the seventh decade of life.¹⁴ Benign mixed tumour, or pleomorphic adenoma, is the second most common benign tumour of the upper lip; it is usually seen in women less than 40 years old. It is also the most common tumour of the major and minor salivary glands.¹⁵ As its name suggests, benign mixed tumour displays an equal ratio of epithelial and mesenchymal cells.

The most common malignant lesion of the salivary glands of the lower lip is mucoepidermoid carcinoma. This tumour occurs over a wide age range, with equal frequency among men and women. Low-grade mucoepidermoid carcinoma may resemble a mucocele on clinical examination, because the predominant cell type in this tumour produces mucin.⁷ Acinic cell adenocarcinoma is the most common malignant lesion of the salivary glands of the upper lip. It, too, can occur over a wide age range but appears predominantly among women.¹⁶

Almost all benign and malignant salivary gland tumours that have been documented in the lips have a similar clinical presentation. Lesions superficial to the mucosa usually present with a bluish colour, whereas the overlying tissue of deeper lesions can have normal coloration.⁷

The differential diagnosis of swelling of the lips in children should also include vascular malformations such as hemangiomas and varices. Usually blue in colour, these blanch under digital pressure, which distinguishes them from other pigmented lesions such as nevi, mucoceles, hematomas and melanomas.¹⁷ Benign mixed tumour, discussed previously, is the most common salivary gland tumour in children and adolescents. Neuroectodermal tumour of infancy is a rare benign

tumour that may occur in the anterior maxilla; in this situation it can elevate the infant's upper lip and appear as a pigmented swelling in this area. Because of the derivation of this tumour from neural crest cells, urine levels of vanillylmandelic acid may be elevated.¹⁸

Palpation of the lesion may aid in developing the differential diagnosis. Cysts, mucoceles, abscesses, hematomas, lipomas and salivary gland tumours may exhibit fluctuance. However, a mucocele that has ruptured would not be fluctuant, and a chronic mucocele that has developed fibrosis may lose some degree of fluctuance. The patient described in this case report had had the lesion for about a year, and the lesion was nontender on palpation.

All of the lesions that have been discussed here may present asymptotically, as in this patient. Some lesions, such as mucoceles, vascular malformations, fibromas and lipomas, may not require treatment. However, because of the possibility of salivary gland tumours in the upper lip, biopsy is necessary to establish the definitive diagnosis, which is paramount for appropriate treatment.

Conclusions

The lesion in the case reported here was diagnosed as a mucocele of the left upper lip, an uncommon location for this lesion. The patient denied trauma to the lips, but a parafunctional habit such as lip biting may go unnoticed. The patient also denied any change in the size of the lesion over the previous year. A mucocele may persist for as long as a year without rupturing, but many patients present within a few weeks of onset for evaluation. A mucocele of uniform size of such long-standing duration is rare.

Because of the possibility that a lesion in this location might be a tumour, 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.¹³ If a benign or malignant tumour is diagnosed, then referral to an appropriate specialist is warranted, as further surgery (to obtain clear margins), radical neck dissection, radiation therapy or chemotherapy may be indicated. This review suggests that the clinician should have a high index of suspicion for any lesion occurring in the upper lip. ♦



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