



Salivary Duct Cyst with Sialolithiasis in Upper Lip: A Rare Entity

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Abstract

Keywords

- ▶ salivary duct cyst
- ▶ upper lip
- ▶ sialolithiasis
- ▶ minor salivary gland
- ▶ sialocyst

Salivary duct cyst (SDC) is a true salivary gland pathology that rarely occurs in the upper lip. The proposed rarity of SDC is around 0.39% of all the salivary cystic lesions. It usually arises in the lower lip or the floor of the mouth and is prevalent in the sixth/eighth decade of life. We report a case of 76-year-old male patient with a chief complaint of an abnormal growth of the upper lip. The growth had increased in size from the last two months. Clinically it was a well circumscribed, solitary, nodular, painless, mobile, noncompressible, soft growth. Excisional biopsy was performed. Histology revealed a SDC and sialolithiasis in the upper lip. This article aims to include SDC in the differential diagnosis of lesions in the upper lip. Their clinical and histopathology features mimic salivary gland pathology such as mucocele, pleomorphic adenoma, basal cell adenoma, low-grade mucoepidermoid carcinoma.

Key Message

Differential diagnosis for a solitary nodular growth in the upper lip needs utmost attention as the lesion could be ranging from a simple mucocele to a low-grade mucoepidermoid carcinoma.

Introduction

Salivary duct cyst (SDC) is a true developmental salivary gland pathology caused by the obstruction of the salivary ducts and is also known as sialocyst or mucous retention cyst. They affect different sites in the oral mucosa, usually the lower lip, floor of the mouth, and rarely the upper lip.¹ They typically occur in the major salivary glands specifically in the

parotid and is extremely rare in the minor salivary glands. SDC is most commonly seen in the sixth–eighth decade of life with a male predilection. The cyst clinically presents as a slow-growing, soft, fluctuant painless swelling, which may be accompanied by bluish tinge. Size of the lesion ranges from 2 to 10 mm in dimension. In the duct mucous stasis, subsequently, acts as a nidus for calcification leading to the presence of sialoliths in such cysts, which further increases the intraluminal pressure and dilation of the duct. Histopathologically, SDCs are found lined at least focally by one to two layers of the cuboidal or columnar epithelium and cysts showed some evidence of squamous metaplasia. Treatment in the form of surgical excision is the preferred approach in such cases. Differential diagnosis of SDC includes mucocele, pleomorphic adenoma, cystadenoma, and low-grade

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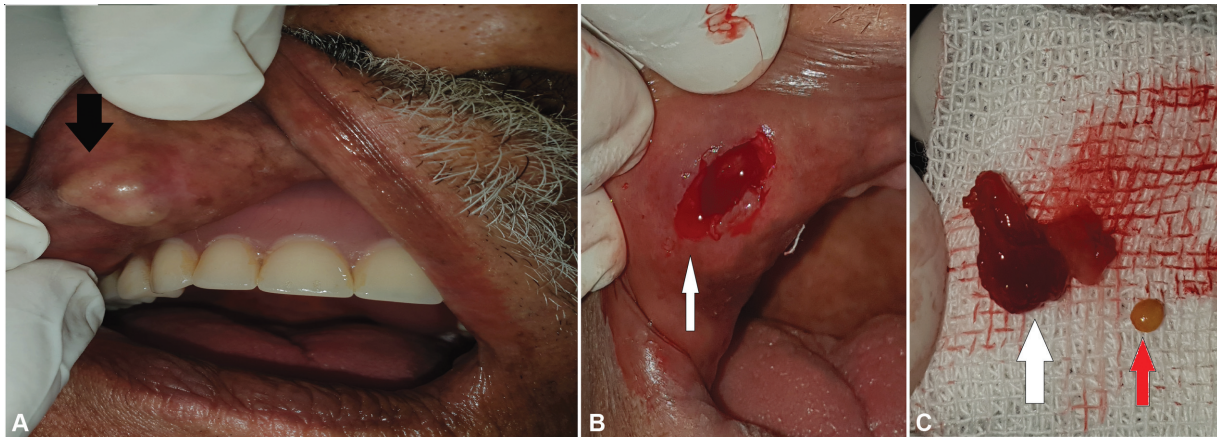


Fig. 1 (A) Intraoral image of the lesion showing solitary growth in the right upper lip (black arrow). (B) Post operative defect before suturing (white arrow). (C) Surgical specimen showing enucleated growth (white arrow) along with a sialolith (red arrow).

mucoepidermoid carcinoma. The recurrence rate of such cysts is low but still requires periodic follow-up. The proposed rarity of SDC is 0.39% of salivary cystic lesions which itself constitute 6 to 8% of all salivary gland pathology. We present a unique case of SDC in a minor salivary duct with sialolithiasis in the upper lip of a 76-year-old Indian male.

Case Presentation

A 76-year-old Indian male patient reported to the dental clinic with a chief complaint of a painless growth in the right upper lip since 2 months, which gradually increased to the present size and he feared that he may injure it during denture wearing or trimming the moustache (►Fig. 1A). The patient gave a medical history of antihypertensive medication (tab Azilsartan Medoxomil 40 mg once a day for 15 years) along with symptoms of the dry mouth. The patient gave a positive history of

smoking 10 cigarettes/day for 20 years. Clinical examination revealed a growth on the right upper lip bulging on the inner surface of the vermilion border, measuring nearly 1 cm × 1 cm. On bi-manual palpation, it was found to be a well-circumscribed, solitary, nodular, painless, mobile, non-compressible, soft growth. The overlying mucous membrane was normal. The possibility of trauma caused by denture or sharp tooth was ruled out by comprehensive intraoral examination of the denture in function.

Diagnostic Assessment

Based on clinical findings a provisional diagnosis of salivary duct cyst of upper lip, excisional biopsy was performed (►Fig. 1B). Clinical differential diagnosis included mucocele, fibroma, lipoma, pleomorphic adenoma, and rarely cystadenoma and low-grade mucoepidermoid carcinoma. A white hard calcified spherical body came out of the mass measuring 1.5 cm × 1.5 cm.

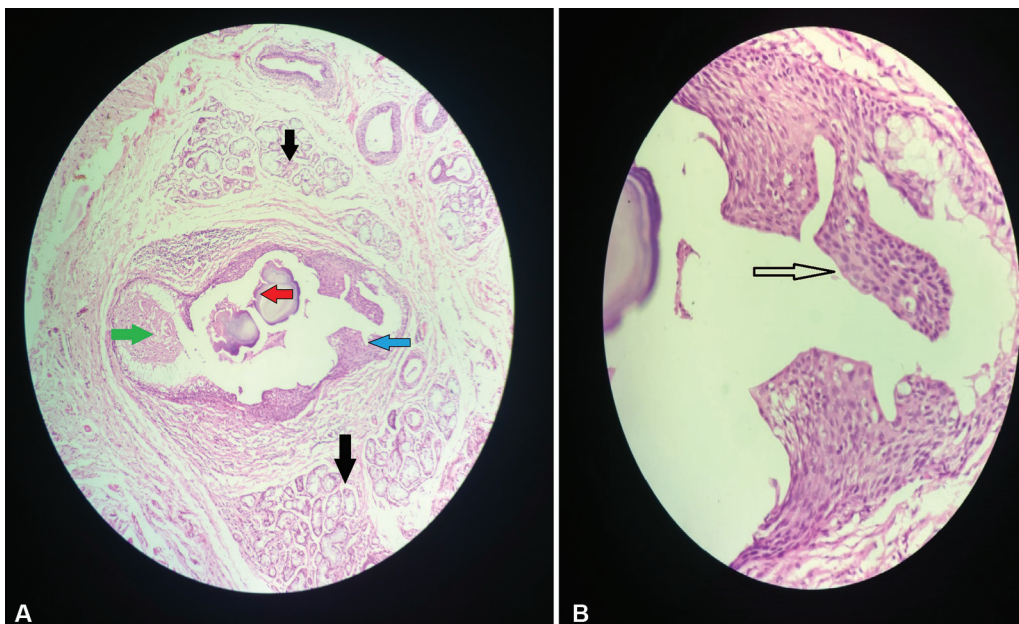


Fig. 2 (A) Photomicrograph showing acinar structure with ductal system (black arrow). Calcified material inside excretory duct which is showing ectasia (red arrow). Squamous metaplasia visible (blue arrow). Mucous plug seen (green arrow). Hematoxylin and eosin (H&E) staining 100X. (B) Photomicrograph showing ductal squamous metaplastic changes (black arrow). H&E Staining. 400X.

The growth was completely excised (►Fig. 1C). Ultrasonography images were inconclusive and size location of the lesion lead to the decision for excisional biopsy.

Histopathology revealed salivary glandular tissue along with supporting stroma. The glandular tissue showed predominantly mucous acini along with the ductal system. There was the presence of calcified material along with a mucous plug in the lumen of the excretory duct showing ectasia (►Fig. 2A). The ductal lining showed squamous metaplasia with papillary projections into the lumen (►Fig. 2B). Acinar structures were normal. No dysplasia was noted. A histological diagnosis of salivary duct cyst with sialolithiasis was established.

Therapeutic Intervention

The patient was prescribed tab. ketorolac 10 mg (twice daily) for 5 days.

Follow-up and Outcome

Because the patient was out stationed, follow-up was done after 6 months and showed uneventful healing (►Fig. 3). No recurrence was reported. The patient was satisfied with treatment outcome and relieved of his fear of injury to the lip while wearing dentures, trimming of moustache. The timeline of events is explained in ►Table 1.

Discussion

Cystic lesions represent around 6 to 9% of the total salivary gland disorders, particularly in the major salivary glands.² According to Ivan, Umer, Sook, out of 177 cases of SDCs examined only 5.6% of SDCs occurred in the upper lip³ making them a rare entity. SDCs present clinical and histopathological characteristics similar to those of other salivary gland pathologies and differential diagnosis includes such as mucocele, pleomorphic adenoma, cystadenoma, and low-grade mucoepidermoid carcinoma, making diagnosis difficult and hence subject to errors in treatment.⁴ Other diseases that can develop at the same site are calcified lymph nodes, phlebolith, fibroma, myxoma, vascular malformation, salivary gland tumors, non-specific sialadenitis, and malignant tumor.⁵⁻⁷ History of trauma can lead us to diagnosis of fibroma or a mucocele. Vascular malformations or hemangiomas have a history of growth present at infancy, which



Fig. 3 Postoperative follow-up with uneventful healing (white arrow).

may grow rapidly in size and have a characteristic reddish-blue tint with normal overlying mucosa.

A mobile growth with superficial ulceration should point out toward the possibility of pleomorphic adenoma of the upper lip. Basal cell adenoma of the salivary glands is usually located in the upper lip, normally firm to palpation, and present in fifth to sixth decades of life and can be diagnosed through trans-surgical macroscopic effects. Warthin tumor (papillary cyst adenoma lymphomatosum) and low-grade mucoepidermoid carcinoma are the other tumors, which need clinical and histopathological correlation in the differential diagnosis of a hard palpable mass. Possibly SDCs develop secondary to idiopathic stasis in secretion due to age-related narrowing of the duct of the minor salivary glands as in this case. Only 4.5% of SDCs show focal intraluminal calcifications, suggesting that this stasis resolves or provides a nidus for sialolith formation,⁸ as must be the case in this patient where multiple calcific bodies could be elucidated. Nidus formation has been linked to smoking and aging as was seen in our case. Sialolithiasis in the minor salivary glands lacks distinctive clinical features and the incidence in minor salivary glands is less than 2%. Histopathological evaluation is important to reach a correct diagnosis. As reported with reactive conditions the histopathological picture of SDC is diverse. 85.3% SDCs are found lined at least focally by one to three layers of cuboidal or columnar

Table 1 Timeline of events

History given by patient	Growth in the right upper lip since 2 months which gradually increased to the present size and he feared that he may injure it during denture wearing or trimming the moustache
Clinical Examination	Growth on the right upper lip bulging on the inner surface of the vermillion border. well circumscribed, solitary, nodular, painless, mobile, non-compressible, soft growth
Diagnosis (Clinical and Histopathological)	Salivary Duct Cyst with Sialolithiasis
Therapeutic Intervention	Tablet Ketorolac dt 10mg (twice a day) for 5 days
Follow-up and Outcome	Follow up after 6 months showed uneventful healing and with no recurrences. Patient condition satisfactory.

epithelium and 68.4% of cysts showed some evidence of metaplastic change.^{9,10} Squamous metaplasia was seen in our case. The strength and uniqueness of this case is the rarity of the lesion in upper lip with concomitant presence of sialolithiasis. The limitation was a regular follow-up due to the pandemic (COVID-19) and difficulty in obtaining a radiographic diagnosis due to the variable appearance of such lesions. Prognosis of such entity is good and recurrence rate is low if completely excised to base.

Conclusion

We report a rare case of salivary duct cyst in the upper lip of a 76-year-old Indian male with multiple sialoliths. Diagnosis in such cases is challenging as the sialoliths are small and not discovered on imaging. Surgical excision and histopathological confirmation is the optimal approach. The goal is to include SDC in the differential diagnosis of a solitary nodular growth of the upper lip.

Conflict of Interest

None declared.

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