Case Report

Intraoral salivary duct cyst: Report of rare entity

Saurabh R Nagar[1], Nishant L Pandey[2], Shivani Bansal[3], Rajiv S Desai[3]

[1]Department of Oral Pathology and Microbiology, Government Dental College and Hospital, Mumbai, [2]Private Practitioner, Thane, [3]Department of Oral Pathology and Microbiology, Nair Hospital Dental College, Mumbai, Maharashtra, India

Abstract

Salivary duct cysts (SDCs) are true cysts caused by obstruction of the salivary ducts and are rare in minor salivary glands. A 62-year-old male reported with a painless swelling in the left buccal mucosa for 2 years. Excision of the entire lesion was performed under local anesthesia following which histopathological examination was performed. Microscopically, a dilated salivary gland duct composed of 1–2 layers of cuboidal cells with intraluminal mucous plug was observed. Cystic lumen lined by mucous cells, squamous cells and ciliated cells was seen. Oncocytic metaplasia was also present at various places. Histopathologically, it was consistent with the diagnosis of SDC. Intraoral SDCs and mucoceles clinicopathologically mimic salivary gland neoplasms, making diagnosis difficult and subject to errors in treatment. It is important for oral and maxillofacial surgeons to include SDC in the differential diagnosis of swelling affecting buccal mucosa.

Keywords: Buccal mucosa, metaplasia, salivary duct cyst

Address for correspondence: Dr. Saurabh R Nagar, Assistant Professor, Department of Oral Pathology and Microbiology, Government Dental College and Hospital, St. George Hospital Campus, Near Chhatrapati Shivaji Terminus Area, Mumbai - 400 001, Maharashtra, India.
E-mail: saurabh.nagar90@gmail.com

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INTRODUCTION

Cystic lesions represent around 6%–9% of the total salivary gland disorders, particularly in the major salivary glands.[1] The salivary duct cyst (SDC), also known as sialocyst, is an acquired cystic dilatation of salivary ducts that are thought to arise secondary to ductal obstruction, which may be transient or persistent.[2,3] Various terminologies may be used to denominate these lesions; however, "SDC" is the most adequate by virtue of its origin being related to the epithelial lining of salivary gland ducts.[4] SDCs typically occur in the major salivary glands mainly in the parotid, where they present as unilateral, asymptomatic, compressible nodules in adult patients. Its occurrence in minor salivary gland is rare and occurs more commonly in the sixth decade of life.[5] We report a rare case of SDC of the buccal mucosa in a 62-year-old male.

CASE REPORT

A 62-year-old healthy man was reported with a gradually increasing swelling in the left buccal mucosa of 2 years duration. While eating, there was no pain or increase in the size of swelling. Intraoral examination revealed painless, mobile, noncompressible, soft, nodular, solitary swelling measuring approximately 10 mm × 12 mm on bimanual palpation. The differential diagnosis of mucocele and salivary gland neoplasm were considered. A provisional diagnosis of mucocele was made, and excisional biopsy of the lesion was performed under local anesthesia followed by

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histopathological evaluation. Gross examination revealed soft brownish cystic sac filled by slimy gel-like material [Figure 1].

H and E-stained sections revealed dilated salivary gland duct with intraluminal mucous plug [Figure 2]. The cystic lumen was lined by one to two layers of cuboidal-columnar mucinous epithelium exhibiting mucous cells, squamous cells and ciliated cells resembling maxillary sinus lining and oncocytic metaplasia at various places [Figure 3]. Dense chronic inflammatory infiltrate composed chiefly of lymphocytes was seen around the dilated duct. Cystic lining also showed mucopapillary projections in some areas and acini of the salivary gland mucosa exhibiting discrete atrophy. Postoperative healing was uneventful.

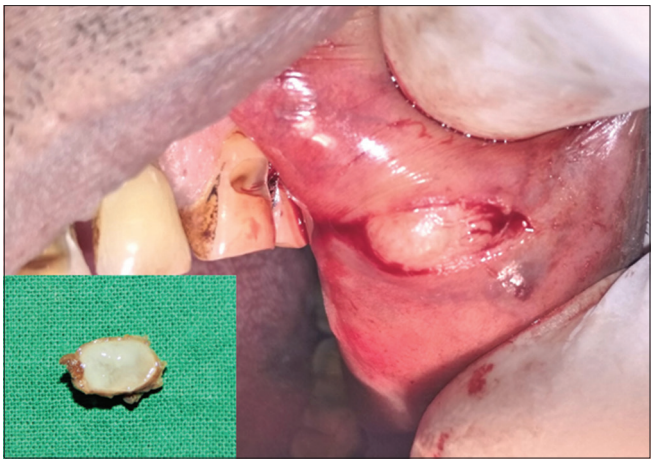

Figure 1: Clinical image showing surgical exposure of salivary duct cyst in the left buccal mucosa (Inset: gross specimen showing cystic sac filled with slimy gel-like material)

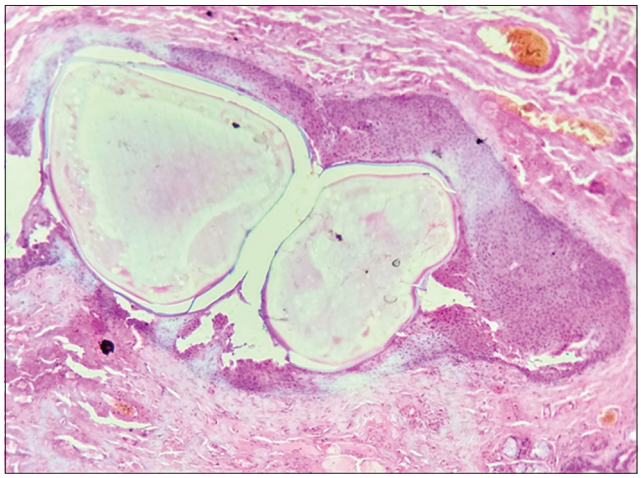

Figure 2: Histopathological image showing cystically dilated salivary gland duct with intraluminal mucous plug and squamous metaplasia of the lining epithelium (H & E, ×100)

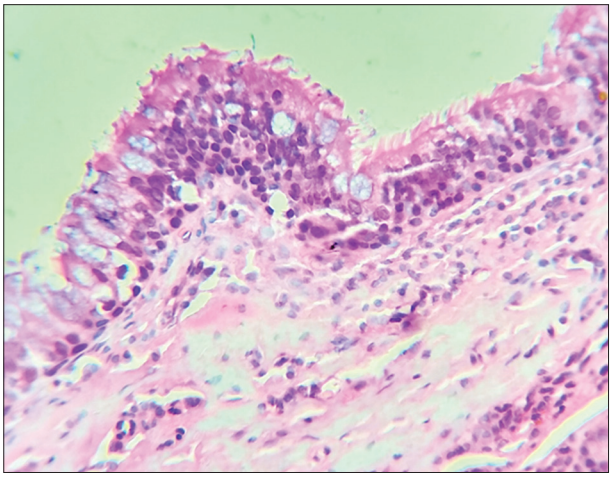

Figure 3: Histopathological image revealing cystic lining exhibiting ciliated, mucous and oncocytic metaplasia (H & E, ×400)

DISCUSSION

The pathogenesis of the SDC is still uncertain. It is believed that with the increase in age, there is a natural reduction in salivary secretion, promoting the formation of a mucous plug that causes partial or total obstruction of the salivary gland duct, resulting in dilation of the duct and increase in intraluminal pressure.[4,6] Although various terminologies such as mucoceles and mucous retention cyst were used to denominate these lesions, the term "SDC" is the most preferable for several reasons.[4] Mucous retention cyst does not adequately describe the phenotypic variation seen in these cysts, whereas SDCs are already commonly used to describe these cysts in the major salivary glands associated with the salivary duct.[7]

SDCs may present clinical-pathological characteristics similar to those of salivary gland pathologies such as mucocele, pleomorphic adenoma, cystadenoma and low-grade mucoepidermoid carcinoma, making diagnosis difficult and subject to errors in treatment.[8]

Clinically, both mucocele and SDC represents as an asymptomatic nodule, but later one is rare in occurrence. Mucoceles can be clinically differentiated from SDCs, having the site of predilection more common on lower lip mucosa (59%–82%), which is a prone site for trauma. SDCs occur on mucosa less prone to trauma include floor of the mouth, soft and hard palate, mandibular vestibule and contiguous areas of the buccal mucosa. Mucoceles occur in younger age group belong to first to fourth decades of life against SDCs having common occurrence in the sixth decade.[4,6,9]

Histologically, SDCs show cystic cavity lined by 1–2 layers of cuboidal or columnar epithelium, consistent with normal salivary duct architecture. Metaplastic changes occur often in the cystic epithelium of the SDC, wherein most commonly reported are oncocytic (50.8%) followed

by mucous cell (36.2%), squamous (23.7%), ciliated cell (16.4%) and apocrine-like (14.1%) metaplasia.[9] The present case demonstrated oncocytic, mucous, squamous and ciliated metaplasia at various places. Intraoral SDCs are reactive ductal ectasia that develops secondary to intraluminal obstruction. Intraluminal calcification foci are rare as approximately half of SDCs show patent lumens and stasis resolving transiently.[9]

Although the occurrence of SDC is rare in the oral cavity, it is important to include it in the differential diagnosis of a cystic lesion affecting buccal mucosa, especially in geriatric patients.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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