

Salivary Duct Cyst of the Lower Labial Mucosa: A Rare Case Report and Literature Review

Saede Atarbashi-Moghadam¹  | Ali Lotfi¹  | Sepehr Ghasemi²  | Mohammadreza Kashefi Baher^{3*} 

1. Department of Oral and Maxillofacial Pathology, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

2. School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

3. Health Research Center, Chamran Hospital, Tehran, Iran.

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*Corresponding Author

Mohammadreza Kashefi Baher,
Health Research Center, Chamran
Hospital, Tehran, Iran.

Email: rezakashefi78@gmail.com

Abstract

Salivary duct cyst (SDC) is a rare reactive lesion of the salivary gland ducts and is infrequently reported in minor intraoral glands, particularly the lower lip. We describe a 49-year-old man with a painless, well-circumscribed submucosal swelling of the lower lip. Clinical findings suggested mucocele or salivary gland tumor, and excisional biopsy revealed a cyst lined by thin cuboidal epithelium adjacent to salivary gland lobules, confirming SDC. The lesion was completely removed, and no recurrence has been observed. Furthermore, a review of English-language literature from PubMed, Scopus, and Web of Science shows that intraoral SDCs typically affect middle-aged adults, demonstrate slight female predominance, and present as soft, mucosa-colored swellings with variable histopathologic patterns, including occasional epithelial metaplasia. Surgical excision remains the preferred management, with recurrence considered rare, although follow-up is recommended. This case underscores the need to include SDC in the differential diagnosis of atypical lip swellings.

Keywords: Salivary glands, Salivary ducts, Salivary gland diseases, Mucocele.

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1. Introduction

Cystic lesions account for less than 10% of salivary gland disorders, with salivary duct cysts (SDCs) representing one of their subtypes (1). SDC, also referred to as a sialocyst or mucous retention cyst, is an acquired lesion characterized by reactive dilation of the salivary ducts. It typically develops as a consequence of intraluminal blockage and presents as a mucus-filled cavity lined by epithelium (2,3). There is considerable confusion in the terminology for this lesion, as some researchers have mistakenly equated the more common intraoral mucocele, caused by mucous extravasation into connective tissue due to trauma or salivary duct rupture, with the mucous retention cyst (mucous retention phenomenon), which arises from a different cause and shows distinct clinical features and histopathology (4). Earlier studies may have considered both mucous extravasation and mucous retention cysts as mucoceles, reporting their findings collectively, which has resulted in further confusion. SDC is the most

appropriate term, as its origin is linked to the epithelial lining of salivary gland ducts (4).

SDCs are most frequently found in the major salivary glands, particularly the parotid gland. However, in rare cases, they can also arise in the minor salivary glands of the oral cavity, with the floor of the mouth being the most common site (2,5).

Beyond the overall rarity of SDC in intraoral minor salivary glands, it is noteworthy that only approximately 15% of reported intraoral cases occur in the lower lip (2). Additionally, due to the lesion's non-specific clinical presentation and markedly variable histopathological features, we present this rare case of labial SDC accompanied by a comprehensive review of previously reported cases in minor salivary glands, emphasizing their demographic profiles, clinical features, and microscopic characteristics.

2. Case Report

A 49-year-old man with a chief complaint of painless swelling

of the lower lip with unknown duration was referred to a private oral pathology center (Tehran, Iran) in May 2025. Past medical and familial history were unremarkable. The intraoral examination showed a large, well-defined, and deep submucosal mass in the left lower labial mucosa near the buccal mucosa measuring 2×1.8 cm. It was similar in color to the surrounding mucosa, with a fluctuant-to-soft consistency, without any mucosal erosion or ulcer (Figure 1).



Figure 1. A well-defined submucosal mass in the left lower labial mucosa near the buccal mucosa.

Based on the lesion's location and clinical characteristics, the differential diagnoses included mucocele, benign mesenchymal neoplasms (lipoma), and salivary gland tumors such as pleomorphic adenoma and low-grade mucoepidermoid carcinoma (MEC).

The extra-oral examination and laboratory data were normal. There was no cervical lymphadenopathy. An excisional biopsy was performed under local anesthesia (Figure 2).



Figure 2. Excisional biopsy of the lesion.

Histopathological examination using H&E staining displayed a cystic lesion lined by thin cuboidal epithelium. Lobules of minor salivary glands were observed adjacent to the fibrous cyst wall (Figures 3 and 4), confirming the diagnosis of a salivary duct cyst. Given the lesion's well-defined margins,

limited size, and benign characteristics, the biopsy was regarded as both diagnostic and therapeutic.

Written informed consent for the publication of this case, including all clinical details and any accompanying images, was obtained from the patient.

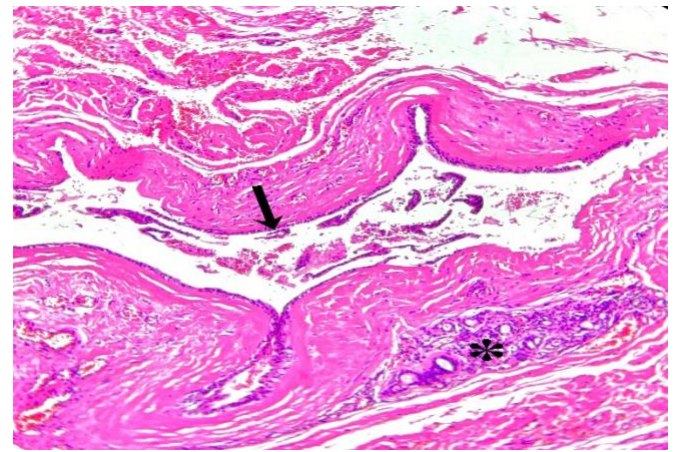


Figure 3. A cystic lesion lined by thin cuboidal epithelium (black arrow). The fibrous wall contains a minor salivary gland (asterisk) (H&E, ×100).

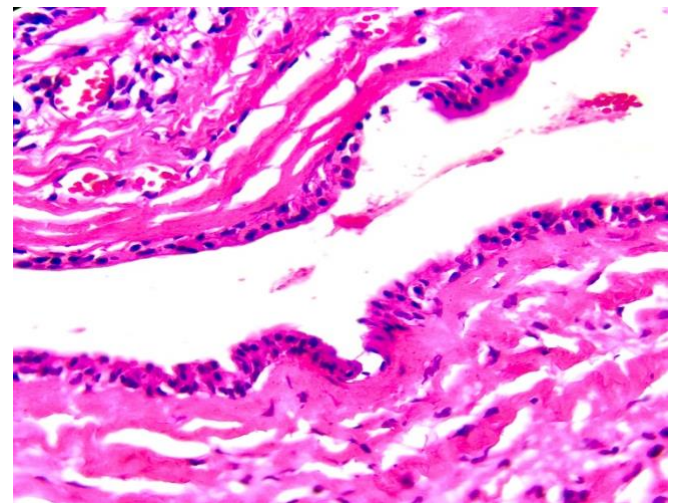


Figure 4. Simple cuboidal epithelium (H&E, ×400).

3. Discussion

The present study reports a SDC of the lower labial mucosa in a middle-aged man that clinically mimicked a mucocele, highlighting the necessity of combined clinical and histopathological evaluation for accurate diagnosis.

SDC is a rare reactive ductal ectasia, typically occurring in major salivary glands and likely resulting from obstruction (2). Given the intraoral presentation of the current case, we performed a comprehensive literature search across PubMed, Scopus, and Web of Science (WoS) using the terms (“Oral” OR “Mouth”) AND (“Salivary duct cyst” OR Sialocyst OR

Table 1. Demographic, clinical, and histopathologic characteristics of documented intraoral SDCs.

Author Year	Age (Years) Sex	Location	Size (CM)	Clinical findings	Provisional clinical diagnosis	Histopathologic finding
Shih et al. 2025 (12)	52 M	Lower Lip	0.8	Swelling	Mucocele with the fibrotic change	One to two layers of eosinophilic columnar epithelium, moreover, a dilated excretory duct lined by one to five layers of eosinophilic cuboid and columnar, oncocytic metaplasia, papillary projections, and lymphocytic infiltration.
Kang et al. 2025 (9)	54 M	Upper Lip	0.6	Swelling (sometimes disturbs the patient during mastication)	Mucocele, Fibroma, Lipoma, Sialolith, Benign minor salivary gland tumor.	Oncocytic metaplasia, lymphocytic infiltration, one to two layers of eosinophilic columnar epithelium, and papillary folds.
Ide et al. 2025 (7)	17 M	Median Mandible	1	Asymptomatic (found incidentally during a routine dental examination)	NA	Uniform layer of respiratory epithelium,
Pantham et al. 2024 (8)	87 M	Hard Palate (postolateral)	2	Swelling, surface ulceration, bleeding	Mucocele, Fibroma, Pyogenic granuloma, Benign tumor of the salivary gland	Single-lined cuboidal epithelium, squamous epithelium showed mucus metaplastic changes, papillary projections, and lymphocytic infiltration.
Jin et al. 2022 (3)	60 F	Upper Lip	0.6	Swelling	Mucocele	Oncocytic metaplasia, lymphocytic infiltration, two to three layers of eosinophilic columnar cells, papillary folds, and mucus plugs.
Nagar et al. 2019 (11)	62 M	Buccal Mucosa	1.2	Swelling	Mucocele, Salivary gland neoplasm	Mucous plug, one to two layers of cuboidal-columnar mucinous epithelium exhibiting mucous cells, squamous cells, and ciliated cells resembling maxillary sinus lining, oncocytic metaplasia, mucopapillary projections, acinic atrophy of salivary gland, and lymphocytic infiltration.
Tseng et al.	22 F	Lower Lip	1	Swelling	Mucocele, Salivary gland tumor, Fibroma, Soft-tissue	The inner layer was lined with a thick layer of connective tissue containing epithelioid histiocytes and multinucleated giant cells; the outer layer was lined with

2015 (14)					tumors	focally keratinized stratified squamous epithelium, exfoliated keratin debris, and chronic inflammatory cell infiltration.
Misra et al. 2014 (6)	7 (present at birth) F	Hard Palate (anterior midline)	1	Swelling	Congenital epulis, Irritational fibroma, Pyogenic granuloma,	Pseudoepitheliomatous-like hyperplasia, inflammatory cell infiltration is not seen, and parakeratinized stratified squamous epithelium.
Chaves et al. 2013 (13)	78 M	Upper Lip	2	Swelling	NA	Two to three layers of cuboid cells, eosinophilic material, compatible with mucin.
Antoniades et al. 2009 (1)	68 M	Upper Lip	1.5	Swelling	Mucocele, Salivary gland tumor	Basal cell adenoma arising from the lining epithelium of the cyst, single or bilayered lining of flattened to cuboidal cells, trabecular pattern projections, and chronic inflammatory infiltration.
	67 M	Upper Lip	0.5	Swelling	Mucocele, Salivary gland tumor	Basal cell adenoma arising from the lining epithelium of the cyst, focal palisading, single or bilayered lining of flattened to cuboidal cells, and trabecular pattern projections.
De Las Casas et al. 2000 (17)	44 F	Tongue	1	Swelling	Mucinous salivary gland neoplasm (low-grade mucoepidermoid carcinoma)	Simple cuboidal epithelium, luminal pseudopapillary projections lined by stratified epithelium.
Tal et al. 1984 (10)	58 F	Lower Lip, Buccal Mucosa	1	Painful swelling, enlarged and tender submandibular lymph nodes	NA	Most of the small, dilated ducts were lined with cuboidal and columnar oncocytes, while the larger ones were lined with mucus-secreting pseudostratified columnar epithelium, oncocytic metaplasia, and minor salivary gland elements were replaced by fibrous tissue, chronic inflammatory cell infiltration.
	83 M	Upper Lip, Buccal Mucosa	NA	Swelling	NA	Two to three layers of flattened cuboidal epithelium, pseudostratified columnar epithelium, oncocytic metaplasia, and absence of inflammatory cell infiltration.

Present Case	49 M	Lower Lip	2	Swelling	Mucocele, Benign mesenchymal neoplasms, Pleomorphic adenoma, Low-grade MEC	Thin cuboidal epithelium, lobules of minor salivary glands in the cyst wall.
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M: Male, F: Female, NA: Not Available

Note that the size of the lesion has been reported according to its greatest dimension.

“Mucous retention cyst”) to characterize demographic, clinical, and histopathological features associated with this rare site of involvement. The initial search identified 78 records in PubMed, 67 in Scopus, and 28 in Web of Science. After removing duplicates, excluding review articles, and restricting to English-language full-text publications without a publication date filter, 14 eligible studies were included. The final search was completed on 10 December 2025. Data extraction was performed independently by two reviewers (M.K.B. and S.A.M.), and discrepancies were resolved through consensus with a third reviewer (A.L.).

Including the present case, a total of 13 case reports and two case series (2,5) with larger sample sizes have been documented. The key details of the case reports are summarized in Table 1.

Intraoral SDCs most commonly present in individuals in their sixth decade of life, with a slight female predominance (2). However, a single congenital case has also been reported (6). This case involved a male patient in his fifth decade. Intraoral SDCs are more evenly distributed across the floor of the mouth and adjacent areas, such as the buccal mucosa, lower lip, mandibular vestibule, and hard or soft palate, sites that are less susceptible to trauma (2). Interestingly, intraosseous SDC has also been reported in the mandible (7). This case involved the lower labial mucosa.

Clinically, it usually appears as a painless, well-circumscribed submucosal mass with a soft-to-fluctuant consistency, exhibiting a coloration similar to that of the surrounding mucosa (3,8,9). The duration of the lesion can influence its firmness, as longer-standing lesions may undergo sufficient calcification, resulting in a firmer consistency (2). While most cases present as unifocal lesions, multifocal involvement of the buccal mucosa and both lips have also been reported (10). The most common clinical differential diagnoses include mucocele, salivary gland tumor, sialadenitis, and irritation fibroma (2, 11). In this case, an excisional biopsy was carried out under the provisional diagnosis of mucocele, benign mesenchymal neoplasm, and salivary gland tumor.

Mucous extravasation cyst may be distinguished by a history of trauma, younger onset age, and its typical site of involvement (2,4). However, a diagnostic challenge in this case was the occurrence of intraoral SDC on the lower lip, where mucoceles most often occur (4). Notably, a case of concurrent mucocele and SDC has also been reported, posing an additional diagnostic challenge (12).

The histopathological appearance of SDCs is highly variable due to their reactive nature. It most often manifests as an

epithelium-lined cyst, sometimes showing papillary projections, along with a dilated excretory duct filled with eosinophilic material consistent with mucin (3,8,9,13). The lining epithelium is most often composed of one or more layers of cuboidal or columnar cells (1,9,11). Moreover, various types of metaplasia may be present, such as oncocytic, mucous, squamous, ciliated, and apocrine-like metaplasia (2, 3, 11). The coexistence of SDC and basal cell adenoma arising from the lining epithelium of the cyst and the presence of Keratin debris, have also been described (1, 14). It should be noted that the diagnosis of SDCs does not need the assistance of immunostaining (3).

The histopathologic differential diagnosis comprises cystadenoma and low-grade MEC (2). Cystadenoma is a well-circumscribed neoplasm characterized by a proliferation of ducts that most commonly occurs in the parotid gland. Histopathologic variants include papillary cystadenoma, papillary oncocytic cystadenoma, and papillary mucinous cystadenoma. Although true papillae with fibrovascular cores, adenomatous plaques, or encapsulation are not seen (2).

MEC can be cystic, but a mixture of epidermoid, mucous, and intermediate cells must be present, and at least one focus of invasion into the underlying connective tissue must be evident. In addition, MEC shows diffuse expression of p63, while positivity is limited to the basal/abluminal cells of SDC and cystadenoma (15).

The treatment of choice includes cryosurgery, carbon dioxide laser surgery, and conservative surgical excision (13). However, SDCs of the parotid may need superficial parotidectomy or marsupialization (16). To the best of our knowledge, recurrence has not been reported. However, long-term monitoring is crucial due to the lack of data for this rare lesion.

4. Limitations

The main limitation of this study is its reliance on a single case and a small number of published cases, limiting generalizability. Additionally, historical misclassification between mucous extravasation and retention cysts confounds the existing evidence. Long-term follow-up data on recurrence and outcomes also remain lacking for this rare lesion.

5. Conclusion

Intraoral salivary duct cysts represent an uncommon presentation of salivary gland pathology, particularly within minor salivary glands, and exceptionally, in the lower lip. The current case reinforces the importance of including SDC in the differential diagnosis of submucosal swellings in salivary gland-rich sites, where the clinical appearance may overlap with more frequent entities such as mucoceles or benign salivary tumors. Accurate diagnosis, therefore, relies on a combination of clinical assessment and histopathological evaluation, given the lesion's variable microscopic profile and non-specific presentation.

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Ethics

Written informed consent for the publication of this case, including all clinical details and any accompanying images, was obtained from the patient.

Using artificial intelligence (AI)

It is declared by the authors of this manuscript that no generative artificial intelligence (AI) or AI-assisted technologies were used to generate content, ideas, or theories during the writing process of this work.

Author contributions

Saede Atarbashi-Moghadam: Conceptualization, Writing - Review & editing, Visualization

Ali Lotfi: Methodology, Investigation, Supervision

Sepehr Ghasemi: Writing - Original Draft, Formal analysis, Validation

Mohammadreza Kashefi Baher: Investigation, Data curation, Writing – Original draft, Writing - Review & editing, Supervision

Conflict of interest

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