

Case Report

Sialolipoma: a rare case of a hamartoma in the floor of the mouth

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Abstract – Sialolipomas are rare benign mature mesenchymal lipomatous proliferations that present in a wide age range. These tumors are involved in both major and minor salivary glands, and most of them are almost only seen in adults when the minor salivary glands are involved. In this article, it underlines the occurrence of a rare case of sialolipoma that manifests as a swelling in the floor of the mouth of a 53 year old female patient and interferes with speech, mastication, and tongue movement.

Introduction

Oral mucosal benign hamartomatous proliferations are comprised of fibrous tissue, adipose tissue, nerve, and muscle [1]. A lipoma is a benign well-delineated neoplasm composed of uniformly sized and shaped lobules of mature fat cells, and it frequently develops in the trunk and the proximal extremities and less in the oral cavity. It accounts for approximately 1–4% of all such tumors [2]. Sialolipoma is a rare, benign, and recent variant of a lipomatous hamartoma occurring in the salivary glands. First described in 2001 by Nagao *et al* [3]. The incidence of sialolipoma occurs more frequently in females and is present in both major and minor salivary glands. Most of them are almost only seen in adults when the minor salivary glands are involved [4]. The presence of mature adipocytes and normal salivary gland tissue represents this tumor. Clinically, this type of lipoma typically manifests as an asymptomatic, slow-growing, soft, movable tumor with no evidence of mucosal or skin abnormalities or other disorders of the salivary glands [5]. Lipomas may have a wide range of radiographical features on the CT and MRI and are useful to assess the extension and location of the lesion. The classic appearance of a lipoma on a non-contrast CT is a well-circumscribed, homogeneously low-signal (fat) radiodense mass ranging from –120 to –65 Hounsfield units [6].

In this study, we highlight an additional case of an intraoral sialolipoma developed on the floor of the mouth independently of the sublingual salivary gland.

Observation

A 53-year-old female patient presented to the oral and maxillofacial surgery department and reported a painless, non-tender, gradually increasing mass in the left floor of the mouth of six months duration, with a slight interference with speech, mastication, and tongue movement, as well as a negative history of trauma, bleeding, ulceration, a decrease in salivation, or any dysgeusia. The medical history of the patient was unremarkable. No lymphadenopathy, oral examination revealed a well-circumscribed soft, movable mass with overlying normal-looking mucosa, causing a slight medial elevation of the tongue (see Fig. 1).

The non-contrast CT demonstrated a well-delineated hypodense mass in the medial surface of the lower left mandible, above the mylohyoid muscle (see Fig. 2).

The patient was operated on under general anesthesia with oral intubation and in the supine position. Surgical access to expose the mass was achieved via an intraoral approach, and the entire lesion was excised with preservation of the sublingual gland.

On gross examination, the mass is encapsulated, yellowish, and measured (3 × 2.5 × 1.5 cm) of smooth outer surface homogenous yellow content (fatty) (see Fig. 3).

Microscopical examination of the sections taken from the submitted specimen revealed well-circumscribed, encapsulated minor salivary gland tissue admixed at the periphery of abundant mature lobular adipocytes showing mild variation in size. The adipocytic tissue is divided into lobules by thin fibrous septa containing scattered benign ductules and acini of the salivary gland (see Fig. 4).

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Fig. 1. A pre-operative clinical picture demonstrates the size and location of the sialolipoma.

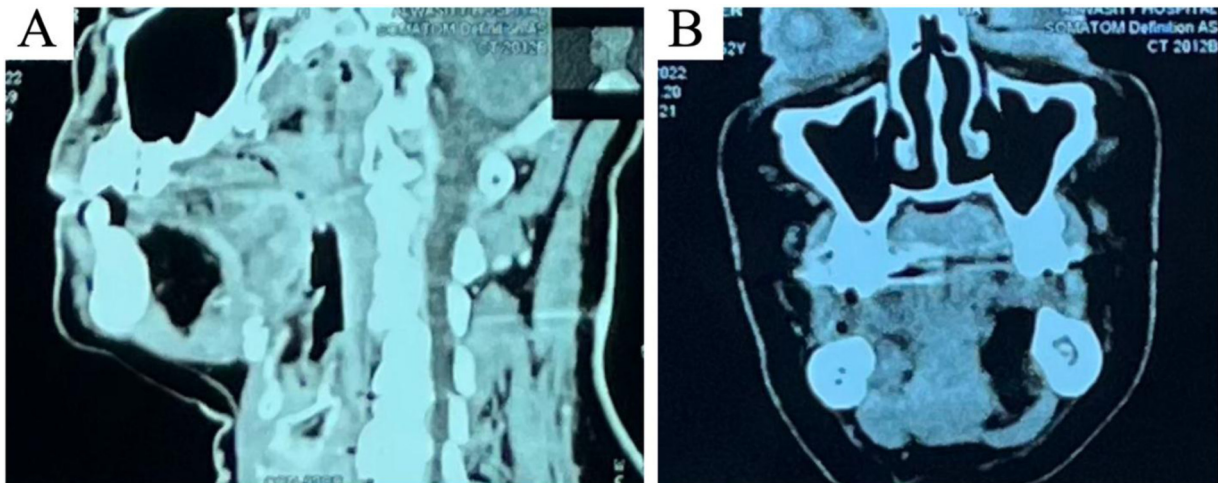


Fig. 2. Non-contrast CT scan views. (A) and (B) sagittal and coronal views, the white arrows demonstrate the extension of the sialolipoma.

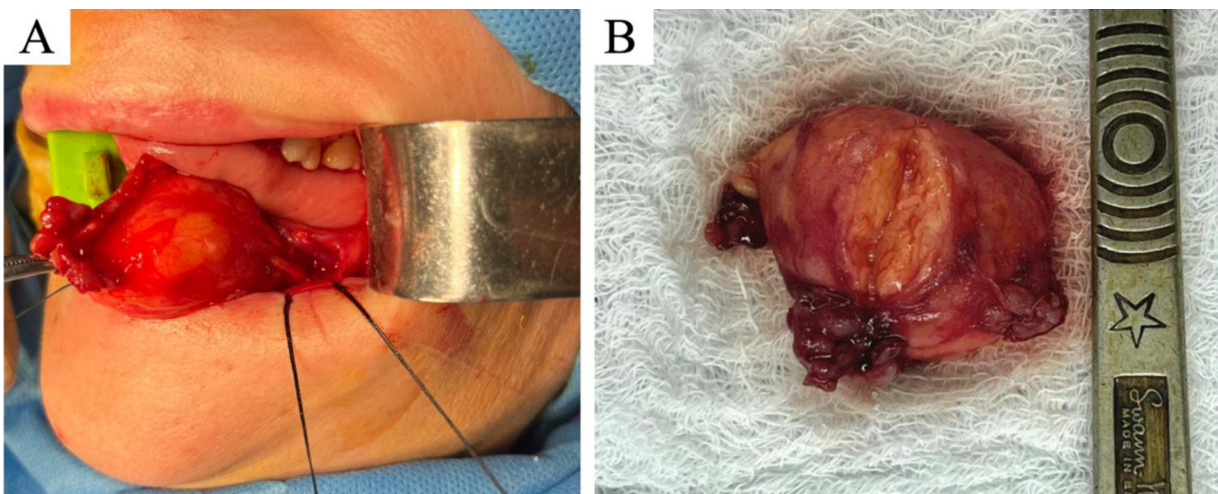


Fig. 3. Intraoperative picture. (A). Demonstrates the surgical excision of the intraoral mass, (B). Demonstrates yellowish, soft, well-circumscribed mass.

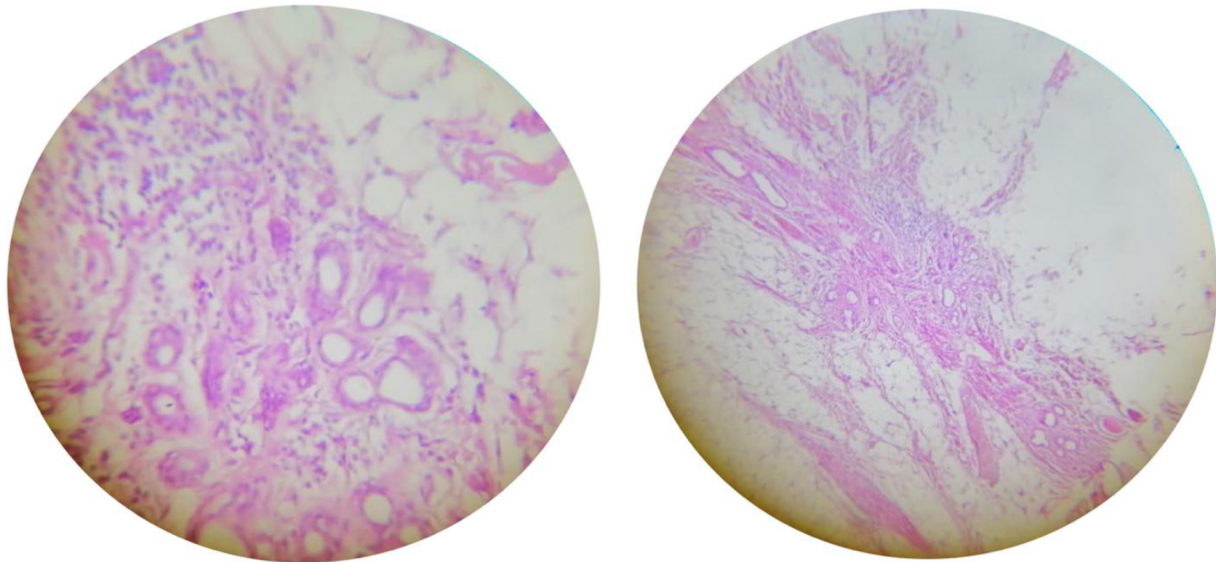


Fig. 4. Histopathological examination demonstrates the adipocytic tissue entrapment inside a minor salivary gland, hematoxylin and eosin stains were used. (A). Magnification power 10 $\times$. (B). Magnification power 4 $\times$.

The patient showed no evidence of recurrence at the 9-month follow-up.

Discussion

The neoplastic nature of lipomatous proliferations within major and minor salivary glands occur with either nononcocytic (sialolipoma) or oncocytic (oncocytic lipoadenoma) epithelial components [7]. Sialolipoma has represented less than 0.3% of all adult salivary gland neoplasms since it was first identified in 2001 [8], sialolipomas are benign hamartomas that develop in the minor salivary glands, and lipomatous tissue accounts for 50–80% of their volume [9]. It was identified in the 2005 WHO classification and included as a distinct entity for the first time in the 2017 WHO classification of Head and Neck tumors [10]. In respect to anatomical site, any location of the oral cavity that contains salivary glands may be affected. The most affected minor salivary glands are the palate, followed by the buccal mucosa, floor of the mouth, tongue, lip, retromolar area, and alveolar ridge, respectively [4].

Such cases manifested clinically as submucosal, well-circumscribed nodules with a soft to firm consistency, normal to yellowish [5]. The lesion is well-circumscribed on gross examination, and it has a uniform yellowish color, a soft to firm consistency, and a smooth or irregular surface. In our case, all these features were present. There is no radiographic evidence distinguishing it from a typical head-and-neck fatty lesion when viewed using high-resolution CT or high-intensity MRI, as it appears as a well-circumscribed hypodense lesion [11].

Nevertheless, our patient was sent for CT because it's informative in narrowing the differential diagnosis due to the lipomatous tumor contents.

Fine needle aspiration was ruled out in this case because of the tumor's clinical and radiographical features and because of the FNA's low accuracy in diagnosing minor salivary gland lipomatous tumors, which is less than 50%. An excision biopsy and histopathological examination confirmed the definitive diagnosis of sialolipoma [12].

The most widely proposed theory for the pathogenesis of the sialolipoma is that it is a histological variant of the conventional lipoma with salivary gland components entrapped in the stroma, which is consistent with the current case [3].

One of the histologic criteria for a sialolipoma diagnosis is the presence of a fibrous capsule that surrounds the tumor and possesses equal amounts of adipose tissue and glandular components [13]. Since the precise pathophysiology of this tumor is unclear, it has been correlated to smoking, diabetes, liver cirrhosis, chronic alcoholism, malnutrition, and hormone imbalances [9].

As with other sialolipomas, the treatment of choice is total surgical excision with the involved gland. In this case, the tumor developed in relation to a minor salivary gland tissue in the floor of the mouth; however, it was not reported to originate from the sublingual gland or to be attached to it upon surgery, therefore it was enucleated without the gland's removal. The case demonstrated no recurrence during a 9-month period of follow-up. Such tumors have no evidence of recurrence except in one case due to incomplete removal [14].

Conclusion

Lipomatosis is a benign proliferation of adipose tissue in the salivary gland parenchyma that causes prominent enlargement of the latter. The fibrous capsule that surrounds sialolipomas is a diagnostic hallmark of this benign tumor. Although the diagnostic criteria and microscopic characteristics of sialolipomas have been relatively well established, many aspects of their histopathogenesis remain poorly understood. Therefore, additional study should be conducted on this newly recognized histologic variant of lipoma.

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Conflicts of Interest

The author reports no potential conflicts of interest.

Data availability statement

The data that support the findings of this study are openly available in journal of oral medicine and oral surgery at <https://doi.org/10.1051/mbcb/2024011>, reference number mbcb230201.

Informed consent

Written informed permission was obtained from the patient to publish this case report and any accompanying images.

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