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CASE REPORT

Sialolipoma in Minor Salivary Gland: Case Report and Review of the Literature

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Abstract Sialolipoma is a rare benign neoplasm characterized by a well-circumscribed mass composed of neoplastic mature adipose tissue and non-neoplastic salivary gland elements. A 72-year-old woman presented with a painless swelling located in the hard palate, which had been identified 15 days earlier. Microscopically, the tumor was well-circumscribed consisting of lobular proliferation of the lipomatous tissue with thin fibrous tissue septa containing clustered salivary gland elements. Both the glandular and adipose components were found in almost equal proportion. No atypia in the adipose tissue was observed. The definitive diagnosis was sialolipoma. The patient showed no signs of recurrence 8 months after surgical excision. Including the present case, 35 cases of sialolipoma have been reported in the English literature. Of these 35 cases, 16 cases were located in minor salivary glands. Gender was identified in 14 of these cases with 4 males (28.5%) and 10 females (71.5%). The age distribution was from 27 to 84 years (mean, 61.6 years) and the tumor size ranged from 0.9 to 4 cm (mean, 1.7 cm). The most frequently reported clinical presentation was of a painless swelling (56.3%).

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Introduction

Lipomatous lesions in the context of salivary glands have attracted interest in recent years and include lesions, such as sialolipoma [1, 2]. Lipomas in the oral cavity are uncommon with a reported incidence of only 1–4.4% of all benign oral lesions [3, 4]. Lipomas can be found in any oral site, especially the buccal mucosa [5–7] and tongue [3] as well as the palate, lip, and salivary glands [1, 8–11]. According to Nagao et al. [9], sialolipoma is a lipomatous proliferation containing acinar and ductal structures bounded by a very thin fibrous tissue.

The sialolipoma is believed to be a lipoma in association with entrapped normal salivary gland elements [3]. This lesion shares similar clinical features with conventional lipomas of the salivary glands including the patients' age, male predominance, and the presence of a slow-growing symptom-free mass [9]. Most reported cases of sialolipoma occur in the parotid gland followed by palate. The mean age is 51 years [3].

Upon microscopic examination, the tumors are well-circumscribed and encapsulated by thin fibrous tissue [8]. According to Nagao et al. [9], they are characterized by islands of epithelial salivary gland elements enclosed in mature adipose tissue. The epithelial islets uniformly consisted of normal duct-acinar units of the salivary gland parenchyma, without any atypia. When they occur in the parotid gland, these epithelial elements are sparsely distributed throughout the tumor. However, in the palatal lesions, the epithelial components seemed to be clustered. The amount of adipose tissue can range from 50% in the

minor salivary glands to over 90% in major salivary glands [9].

A thorough review of the English literature revealed 34 cases of oral sialolipoma, including 15 within the minor salivary glands [1, 5, 8–13]. Most patients have been treated by conservative surgical excision, and neither local recurrence nor malignant transformation has been reported [1, 3, 12]. This article reports a case of sialolipoma located in the palate of a 72-year-old woman and presents a literature review of minor salivary gland sialolipomas.

Case Report

A 72-year-old black female was referred to the Stomatology Service of the School of Dentistry for evaluation of a painless swelling located in the palate, which had been identified 15 days earlier. The patient had been a smoker for 20 years and had recently quit the habit. Her medical history was non-contributory. Clinical examination revealed an asymptomatic mass measuring 2 cm on the hard palate (Fig. 1). The mass was palpable, movable, soft and normal color with purple points. The clinical impression was that of a minor salivary gland tumor, such as pleomorphic adenoma. Occlusal and panoramic radiographs were non-contributory. An excisional biopsy was performed and the specimen "floated" in the formalin solution. The intraoperative impression was lipoma. The surgical specimen was submitted for histologic examination.

Gross examination revealed a soft, yellow mass measuring 1.7 × 1.3 × 0.6 cm. Microscopically, the tumor was well-delineated consisting of lobular proliferation of the lipomatous tissue with thin fibrous tissue septa containing clustered salivary gland elements (Fig. 2). Both the glandular and adipose components were found in almost equal proportion. The acinar cells were mucous and showed acinar atrophy and fibrosis. Duct ectasia, hyperplasia ductal focus and slightly basophilic amorphous materials compatible with mucin were also noted (Fig. 3). No atypia in the adipose tissue was observed. A scant lymphocytic infiltrate was present. These glandular elements were scattered throughout the lipomatous lesion. The definitive diagnosis was sialolipoma. The patient showed no clinical signs of recurrence 8 months after surgical excision.

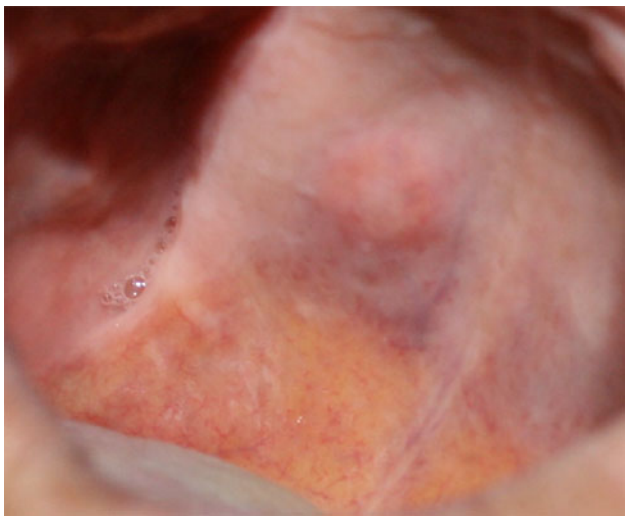

Fig. 1 Clinical appearance of the lesion in palate

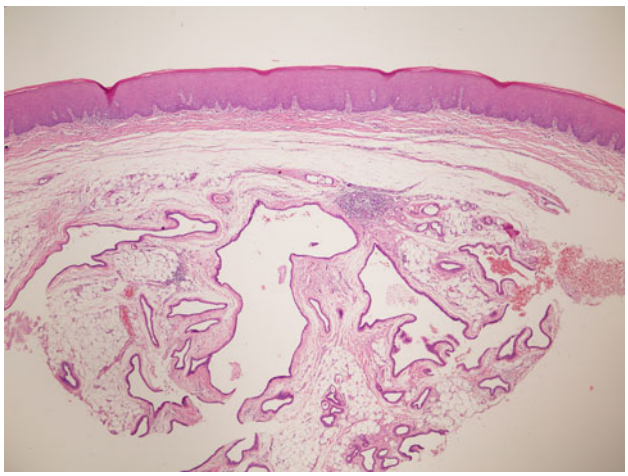

Fig. 2 Microscopic aspect of the tumor well-delineated H.E. (100×)

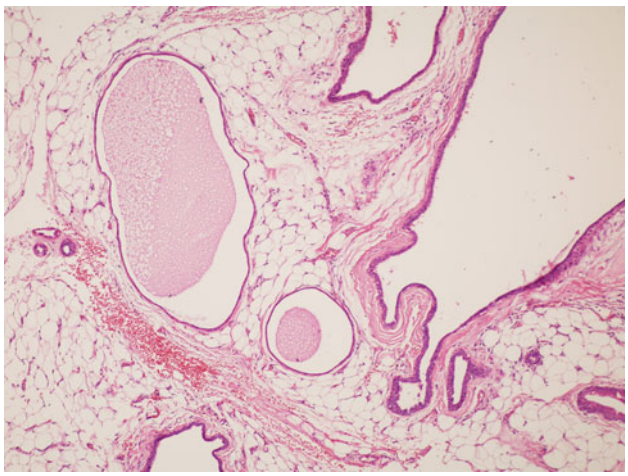

Fig. 3 Microscopic aspect of ectasia of the duct with slightly basophilic amorphous material compatible with mucin H.E. (200×)

Review of the Literature

The present case is 35th reported sialolipoma in the English literature and the 16th case presenting in minor salivary glands (Table 1). The age distribution was 27–84 years (mean, 61.6 years), and 4 cases were in males (28.5%) and

Table 1 Summary of clinicopathological features of minor salivary gland sialolipoma

	Author	Age/sex	Site	Clinical presentation	Duration	Size of specimen (cm)	Follow-up
1	Nagao et al. [9]	75/M	Hard palate	NA	3 years	1	NA
2	Nagao et al. [9]	66/M	Soft palate	NA	6 years	2.2	NED at 11 months
3	Fregnani et al. [8]	NA	Tongue	NA	NA	NA	NED
4	Fregnani et al. [8]	NA	Buccal sulcus	NA	NA	NA	NED
5	Lin et al. [1]	67/F	Floor of mouth	Painless swelling	1 year	3	2 years
6	Sakai et al. [5]	60/F	Hard palate	Painless swelling	10 years	1.8	NED
7	Ramer et al. [10]	84/F	Buccal mucosa	NA	NA	1	11 months
8	Ramer et al. [10]	43/F	Soft palate	Painless swelling	NA	2	NA
9	Ponniah et al. [2]	60/M	Floor of mouth	NA	NA	2	2 years
10	De Freitas et al. [11]	38/M	Lower lip	NA	NA	1	NA
11	Okada et al. [12]	66/F	Hard palate	Painless swelling	10 years	1.2	NA
12	Nonaka et al. [13]	27/F	Tongue	Painless swelling	5 years	1	1.5 month
13	Nonaka et al. [13]	73/F	Floor of mouth	Painless swelling	NA	4	NA
14	Nonaka et al. [13]	64/F	Buccal mucosa	Painless swelling	2 years	2	NA
15	Nonaka et al. [13]	68/F	Retromolar pad	Painless swelling	NA	0.9	14 month
16	Present case	72/F	Hard palate	Painless swelling	15 days	1.7	NED at 8 months

F Female, *M* male, *NA* not available, *NED* no evidence of disease

10 (71.5%) cases in female (gender not available in two cases). The tumor size ranged from 0.9 to 4 cm (mean, 1.7 cm) (Table 1). The clinical presentation most frequently reported was a painless swelling (56.3%) (Table 1). These findings are in accordance with the findings of Okada et al. [12] and Nonaka et al. [13], who report that the lesions present as a slow growing and asymptomatic swelling.

Anatomic sites of minor salivary gland sialolipomas include the soft palate, hard palate, tongue, buccal mucosa, floor of the mouth, buccal sulcus, retromolar pad and lower lip (Table 1). Although no preference for an anatomic site has been described for sialolipomas the palate is the most common site with 6 (37.5%) cases reported [1, 5, 8–11]. In contrast, the buccal mucosa is the site most commonly affected by oral lipomas, corresponding to 30.5–45.7% of cases [6–8].

Clinically, the localization of a nodular mass in the palate suggests a salivary gland tumor. Ramer et al. [10] reported that 4 of the 6 minor salivary gland sialolipomas reviewed, have similar clinical impressions of a benign salivary gland tumor such as pleomorphic adenoma. The yellow color of the gross specimen and its ability to float in formalin suggested a diagnosis of lipoma.

Discussion

Although lipoma is a common soft tissue tumor, it rarely occurs in the salivary gland [12]. Lipomatous lesions, such sialolipoma, lipoadenoma, lipometaplasia in pleomorphic adenoma and lipomatosis, have attracted interest in recent years [2]. Oral lipomas developing in association with salivary glandular elements have been recognized in surgical specimens and have been designated as sialolipoma [3]. The sialolipoma is believed to be a lipoma with secondary normal salivary gland elements. It was considered as a distinct variant of salivary gland lipoma that can occur in both major and minor salivary glands. There have been few literature reports for this kind of tumor [3, 5, 10].

Upon microscopic examination, sialolipomas are characterized by a well-circumscribed, often encapsulated mass, composed of benign, neoplastic, adipose tissue with scattered foci of generally atrophic, nonneoplastic, salivary gland acini and ducts contained within the lipomatous proliferation. Adipose tissue constitutes 90% of parotid gland sialolipomas and 50% of palate cases [9]. We noted a well-delineated tumor consisting of a proliferation of adipose tissue and entrapped normal salivary gland elements with equal tissue distribution.

The entrapped epithelial islets consist of normal ductacinar units of the salivary gland parenchyma [9]. In this case, the epithelial components were clustered and the glandular components showed acinar atrophy. Yet, the ductal structures exhibited dilatation, hyperplasia and thin fibrous tissue around them. Some authors have reported an intense lymphoid infiltrate, marked ductal dilatation, fibrosis and myxoid change in adipose tissue [1, 5, 9]. We observed scarce chronic inflammatory cells and no myxoid areas.

The microscopic differential diagnosis of sialolipoma includes lesions with extensive adipose tissue, such as lipomatosis and pleomorphic adenoma with extensive adipose content [3, 10, 12]. Lipomatosis is nonmalignant overgrowth of adipose tissue throughout the salivary gland parenchyma, resulting in an intensive shrinkage of acinar cells, while sialolipoma is well-capsulate and exhibits ductal dilatation [9]. Pleomorphic adenoma has distinct histological features to sialolipoma, including ducts and sheets or strands of dark-staining epithelial cells [13].

The treatment of the sialolipoma consists of conservative surgical excision, and neither local recurrence nor malignant transformation has been reported [1, 3, 12, 13]. In the present case no evidence of recurrence has been noted 8 months after surgical excision.

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