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SIALOLIPOMA OF PALATE: A RARE HISTOLOGICAL DIAGNOSIS WITH REVIEW OF LITERATURE

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ABSTRACT: Lipomatous lesions coexisting in the major and minor salivary glands have attracted interest in recent years and include lesions, such as sialolipoma. Sialolipoma is a newer variant of salivary gland lipoma, first described by Nagao et al in 2001. As the hard palate has very little fatty tissue, the incidence of lipomas here is very low. Till date 35 cases of sialolipoma have been found to be reported on review of English scientific literature [PubMed], of which 18 cases involved major salivary glands and rest 17 cases occurred in minor salivary glands. On thorough literature survey, eight cases have been found to involve palate (including hard and soft palate) and our case is likely to be the ninth case of sialolipoma involving the palate.

KEYWORDS: Minor salivary glands, Oral pathology, Palate, Sialolipoma.

INTRODUCTION: Lipomatous lesions coexisting in the major and minor 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, most commonly in the buccal mucosa, a region with abundant fatty tissue⁵⁻⁷ followed by tongue³ and less commonly in the palate, lip, and salivary glands.^{1,8-11} As the hard palate has very little fatty tissue, the incidence of lipomas here is very low.⁸ Sialolipoma is a newer variant of salivary gland lipoma, first described by Nagao et al in 2001.⁹ He described it as well circumscribed lesion having lipomatous proliferation containing acinar and ductal structures bounded by a very thin fibrous tissue as capsule.

The sialolipoma is believed to be a lipoma showing mature adipocytes in association with entrapped normal salivary gland elements.^{3,9} Till date 35 cases of sialolipoma have been found to be reported in English scientific literature [PubMed], of which 18 cases involved major salivary glands and rest 17 cases occurred in minor salivary glands.¹² On thorough literature survey, eight cases have been found to involve palate (Including hard & soft palate) and our case is likely to be the ninth case of sialolipoma involving the palate.

CASE REPORT: A 53 year old male patient was presented in the ENT OPD of a tertiary care hospital with painless swelling over left half of palate for last 6 months, insidious in onset and gradually progressive in nature. The patient was a non-smoker and his medical history had nothing significant to be mentioned. Although painless the swelling caused some difficulty while swallowing foods. On intra-oral examination a normal mucosa covered irregular swelling was found, about 3cm x 2.5cm, involving left half of posterior hard palate, ipsilateral soft palate and anterior pillar of left tonsil and

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extending medially close to the midline (Figure 1). The lesion was non-tender and soft on palpation, devoid of any fluctuation or pulsation.

Intra-oral fine needle aspiration cytology (FNAC) was performed which indicated towards benign tumour arising from minor salivary gland, such as pleomorphic adenoma. Computed tomographic (CT) scan of skull base to clavicle was ordered, which revealed an irregular heterogenous non-enhancing soft tissue mass involving left half of the palate, extending posteriorly and laterally upto anterior pillar of left tonsil. There was thin fibrous capsule around the lesion, as it merged with the palatal tissue and it was about 3.5 x 2.4 x 1.9 cm in dimension without any evidence of infiltration or palatal bone destruction (Figure 2). Radiographs like dental occlusal view and orthopantomogram were unremarkable. There was no associated cervical lymphadenopathy noted either clinically or radiologically.

His blood parameters were all within normal limits and after obtaining fitness for general anesthesia, we embarked on surgical resection of the palatal mass with an impression of a benign minor salivary gland tumour. General anesthesia was given via trans-nasal endotracheal intubation and the exposure was obtained using a Boyle-Davis mouth gag. An incision was made on the mucosa over the mass and it was dissected out from the overlying uninvolved palatal mucosa. Haemostasis was obtained using bipolar diathermy and branches of greater palatine artery were cauterised. The mass was removed en bloc and palatal incision was closed in two layers.

The post-operative period was uneventful. Intraoperatively the specimen was found to be "floating" in the formalin solution and so the impression was lipoma. The excised specimen was ovoid in shape, 3.6 x 2.5 x 2 cm in size, lobulated, with thin but definite fibrous capsule. Surface was greyish with yellowish areas scattered in between (Figure 3). On histological examination the mass was found to be composed of islands of neoplastic mature adipose tissue (Black arrow) entrapping salivary gland acini showing focal atrophic changes (Green arrow) along with dilated ducts showing secretions (Red arrow). Focal areas show chronic inflammatory infiltrates (Blue arrow). No malignant changes are seen (H&E, 100X) (figure 4). The mature adipose tissue showing lobular proliferation wholly entrapping small island of normal salivary gland acini along with dilated ductular system (H&E, 100X) (Figure 5).

The mature adipose tissue encroaching salivary glandular structure along with inflammatory infiltrates, focal fibrosis and collagenisation (Capsular remnant) (Blue arrow) (H&E, 100X) (Figure 6). Both the glandular and adipose tissue components were found in almost equal proportion. Duct ectasia and hyperplastic ductal foci were also noted. The adipose tissue was free from any atypia. Thus the definitive diagnosis was sialolipoma. On special staining with PAS-Alcian blue, proliferation of neoplastic adipose tissues interspersed with atrophic salivary gland acini and dilated ducts was noted (PAS-Alcian blue 100X) (Figure 7). PAS staining shows atrophic salivary gland acini and markedly dilated ectatic Salivary duct (PAS-400X) (Figure 8). The patient was followed up 1 week and 1 month post-operatively and now is on monthly follow-up without having any clinical evidence of recurrence 8 months after resection of the lesion.

DISCUSSION: Although lipoma is a common soft tissue tumour, it rarely occurs in the oral cavity, especially in salivary gland.^{13,14} Histological variants of lipomatous lesions, such as sialolipoma, lipoadenoma, lipometaplasia in pleomorphic adenoma and lipomatosis affecting salivary glands, have attracted interest in recent years.² Lipomas of hard palate are very rare due to the presence of very

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little amount of adipose tissue.^{7,8} Oral lipomas developing in association with salivary glandular elements have been recognized in surgical specimens and have been designated as sialolipoma.³

Sialolipoma is a newer variant of salivary gland lipoma, first described by Nagao et al in 2001, who described these lesions microscopically as 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.⁹ The entrapped epithelial islets consist of normal duct-acinar units of the salivary gland parenchyma without any atypia.⁹ The amount of adipose tissue can range from 50% in the minor salivary glands to over 90% in major salivary glands.⁹

Our case also showed similar histological features with almost equal proportions of adipose tissue and glandular elements. In sialolipoma the glandular components show acinar atrophy. Usually the ductal structures exhibit dilatation, hyperplasia and thin fibrous tissue around them.¹⁵ Some authors have reported an intense lymphoid infiltrate, marked ductal dilatation, fibrosis and myxoid change in adipose tissue.^{1,5,9} We observed acinar atrophy, ductal dilatation and scarce chronic inflammatory cells but no myxoid areas.

Since the histological description of sialolipoma by Nagao et al, 35 cases of sialolipoma have been reported in English literature, of which 17 cases involved minor salivary glands. Of these only eight cases till date have been reported to involve the palate (Including hard & soft palate) and these cases are summarized below:

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.Sakai et al. ^[5]	60/F	Hard palate	Painless swelling	10 years	1.8	NED
4.Ramer et al. ^[10]	43/F	Soft palate	Painless swelling	NA	2	NA
5.Okada et al. ^[13]	66/F	Hard palate	Painless swelling	10 years	1.2	NA
6. Moraes et al. ^[15]	72/F	Hard palate	Painless swelling	15 days	1.7	NED at 8 months
7. Arai Y et al ^[16]	75/M	Palate	Painless swelling	3 years	1.2 x 1.2	NED at 21 months
8. Smitha T et al ^[14]	45/M	Palate	Painful swelling (Thermal burn)	20 years	1.5 x 1.2	NA
9. Present case	53/ M	Palate	Painless swelling	6 months	3.6 x 2.5 x 2	NED at 8 months

TABLE 1

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

Table 1: Showing the summary of clinic-pathologic features of sialolipoma involving the palate.

Thus our case is likely to be the ninth case being reported in English literature with sialolipoma involving the palate (Table 1). These lesions share similar clinical features with conventional lipomas of the salivary glands including the patients' age, gender predominance, and the presence of a slow-growing symptom-free mass.⁹ Among the cases of sialolipomas involving the palate, age range was 43-75 years (mean 61.7 years) (Table 1) and the mean age is close to the mean age of the cases of minor salivary gland sialolipomas (i.e. 61.6 years).¹⁵ 5 cases were males & 4 cases were female, thus without any distinct sex predilection (Table 1), which is again in accordance with the fact that no direct sex predilection can be found in the cases affecting minor salivary glands.¹⁴

The lesion size ranged from 1 cm to 3.6 cm (mean 1.8cm) (Table 1) and on review of available literature it has been found that minor salivary gland sialolipomas tend to be smaller than their major salivary gland counterparts, with average greatest dimension being 1.7 cm.¹⁴ Most of the lesions were of long duration (> 1 year duration, the longest duration was 20 years & shortest was 15 days) and the case under discussion was present for 6 months. (Table 1) Minor salivary gland sialolipomas are common to have long duration of presentation, all being present for more than one year as found in reported literature.¹⁴ Most of the cases presented as slow growing, asymptomatic, painless mass (Table 1) and literature review also supports the same presentation in most of the cases (56.3%).¹⁵

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.¹⁵ Although no preference for an anatomic site has been described for sialolipomas the palate is the most common site with eight (i.e. 47%) cases reported^{1,5,8-11} and the present case is an addition to the tally. Okada et al reviewed the histological features of sialolipoma and they found glandular atrophy, periductal fibrosis, ductal dilatation in all the cases¹³ with oncocytic metaplasia with mild to moderate lymphocytic infiltrate in some of the cases.^{9,13,17} Peripheral nerve involvement has been found in cases involving major salivary glands¹³ whereas periductal fibrosis & myxoid degeneration have been reported in minor salivary gland sialolipomas.¹³ Okada et al analyzed that proliferation of adipose tissue in minor salivary glands causes saliva retention in the ducts which causes microcirculatory disturbances resulting in ductal dilatation and periductal fibrosis, thus accounting for the common histological features.¹³

Microscopically sialolipoma needs to be differentiated from lesions with extensive adipose tissue, such as lipomatosis and pleomorphic adenoma with extensive adipose content.^{3,10,13} Lipomatosis is histologically characterized by nonmalignant unencapsulated overgrowth of adipose tissue throughout the salivary gland parenchyma, resulting in an intensive shrinkage of acinar cells, while sialolipoma is well-capsulated and exhibits ductal dilatation.⁹ Pleomorphic adenoma has distinct histological features with ducts and sheets or strands of dark-staining epithelial cells¹⁸ and can easily be distinguished from sialolipoma, where these are absent and instead there are ductal dilatation and periductal fibrosis. Okada reported after immunohistochemical study, that glandular component of sialolipoma has same reactivity pattern as the rest of normal palatal gland and has low proliferative index, suggesting secondary entrapment.

Thus the entrapped glandular tissue within the adipose tissue is involved secondarily and it is not attributable to true neoplasia, rather it is a benign lesion.¹³ On the basis of benign nature of

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sialolipoma, conservative surgical excision has been regarded to suffice as treatment, and neither local recurrence nor malignant transformation has been reported.^{1,3,13,18} In the present case no evidence of recurrence has been noted 8 months after surgical excision.

CONCLUSION: Sialolipoma, which is a newer variant of lipoma, is a rare lesion in the oral cavity although it should be kept in mind during differential diagnosis of lesions showing lipomatous proliferation containing residual salivary gland units intra-orally. Our case of sialolipoma involving the palate, which is likely to be the ninth case, was initially presumed to be benign salivary gland tumour involving minor salivary glands and later was re-diagnosed as sialolipoma based on final histological study. Thus only a high index of suspicion and study of histological details can only lead us to the definitive diagnosis of this rare benign tumour.

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- FIGURE 1:** Clinical photograph showing the intra-oral lesion over left side of palate.

FIGURE 2: CT scan figure (axial cut) showing irregular heterogeneous non-enhancing soft tissue mass involving left half of the palate.

Fig. 2

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FIGURE 3: Showing the excised specimen, lobulated, capsulated with scattered yellowish areas.

Fig. 3

FIGURE 4: Photomicrograph shows neoplastic mature adipose tissue (black arrow) entrapping salivary gland acini showing focal atrophic changes (green arrow) along with dilated ducts showing secretions (red arrow). Focal areas show inflammatory infiltrates (blue arrow). No malignant changes are seen. (H&E, 100X).

Fig. 4

FIGURE 5: Photomicrograph shows predominantly neoplastic mature adipose tissue wholly entrapping small island of normal salivary gland acini along with dilated ductular system. (H&E, 100X).

Fig. 5

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FIGURE 6: Photomicrograph shows mature adipose tissue encroaching salivary glandular structure along with inflammatory infiltrates& focal fibrosis & collagenisation (capsular remnant)(blue arrow). (H&E, 100X).

Fig. 6

FIGURE 7: Photomicrograph shows proliferation of neoplastic adipose tissues interspersed with atrophic salivary gland acini and dilated ducts. (PAS-Alcian blue 100X).

Fig. 7

FIGURE 8: Photomicrograph shows atrophic salivary gland acini and markedly dilated ectatic Salivary duct. (PAS-400X).

Fig. 8

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