10.5005/jp-journals-10015-1058

CASE REPORT

Sialolipoma of Hard Palate: A Rare Variant of Lipoma

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ABSTRACT

The designation 'sialolipoma' is given to a recently described histologic variant of lipoma characterized by well-demarcated proliferation of mature adipocytes with secondary entrapment of salivary gland elements. Nagao et al from Japan first described this new variant. These tumors have been observed in both the major and minor salivary glands with around 35 cases being reported in the English literature. The most common site for the tumor is the parotid (17 cases, 60.7%) followed by the hard palate (four cases, 14.2%). We report a similar case of sialolipoma arising in the hard palate presenting in a 45-year-old male. Diagnosed as an ordinary lipoma of the hard palate, complete surgical removal of the tumor was done. The histopathological features showed a well-encapsulated lesion exhibiting both adipose tissue and glandular epithelium, and based on the recent criteria of histological findings of sialolipoma, the tumor was diagnosed as sialolipoma of the hard palate. Because of its rarity, it was fitting to report this entity.

Keywords: Salivary gland lipoma, Sialolipoma, Histopathological.

INTRODUCTION

Lipomas in the oral cavity are relatively rare. The most common locations of lipomas in the oral cavity have been reported in the buccal mucosa, a region abundant in fatty tissue, followed by the tongue.[1] The hard palate has very little fatty tissue, and the incidence of lesions here is quite low.[2] Sialolipoma is a new variant of salivary gland lipoma, which was first reported by Nagoa et al who defined it as a well-circumscribed lesion composed of mature adipose tissue and entrapped normal salivary gland with a thin fibrous capsule.[3] The most common site for the tumor was the parotid gland (17 cases, 60.7%), followed by the palate (four cases, 14.2%).[4] Moraes M et al recently reported a case involving the hard palate.[5] We report a case which could be the sixth case involving the hard palate.

CASE REPORT

A 45-year-old male reported to the Department of Oral Medicine and Radiology, Vokkaligara Sangha Dental College and Hospital, Bengaluru, with a chief complaint of a slightly painful midline swelling in the palate. The patient had first noticed the small swelling 20 years ago, which increased over a period of time and reached the present size. Patient had pain in the region because of a recent thermal injury and discomfort while swallowing food. On physical examination intraorally, a large spherical, solid, pale pink nodule was seen in the midline region in the posterior part of hard palate. The swelling was sessile and well-circumscribed, measuring about 1.5 × 1.5 cm in size (Fig. 1). Overlying surface mucosa was intact and of normal color. Surrounding mucosa was erythematous with enlarged minor salivary gland duct orifices. On palpation, the swelling was slightly tender and the lesion was compressible. A provisional diagnosis of a lipoma was made and CT was advised. On computed tomography (CT) (Fig. 2), the mass showed low intensity with slightly compressive resorption of the surface palatal bone and the impression was that of a benign tumor, possibly lipoma. Routine blood investigations were carried out and the values were in the normal range. Complete resection was performed and the excised specimen was sent to the department of oral and maxillofacial pathology.

Grossly, the tumor was well-circumscribed and cut section resembled an ordinary lipoma measuring around 1.5 × 1.2 cm (Fig. 3). Histologically, microscopic examination of hematoxylin and eosin stained sections showed stratified squamous epithelium

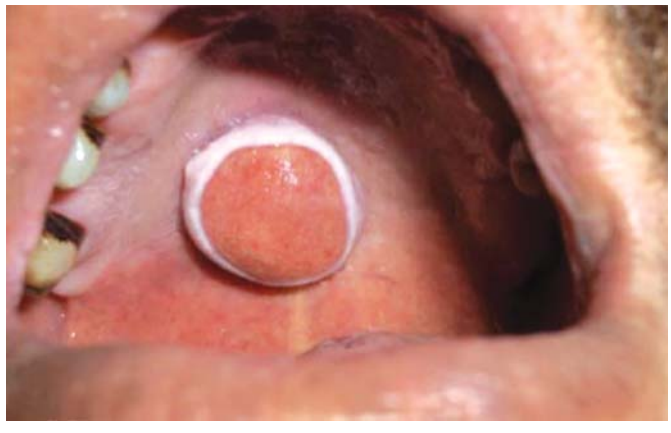

Fig. 1: A firm, pinkish nodule located in the midline region in the posterior part of hard palate

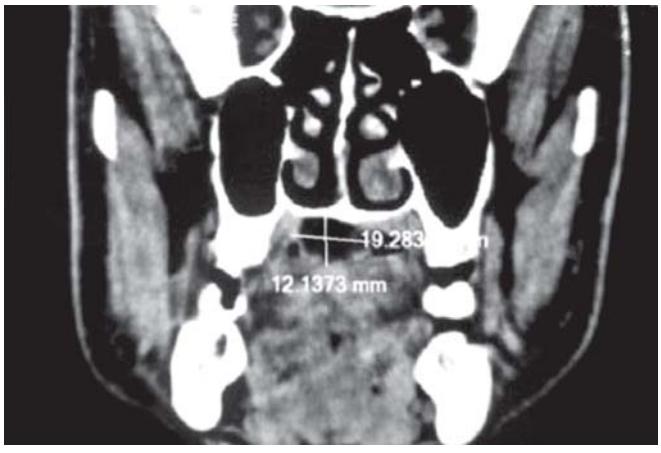

Fig. 2: Computed tomography (CT) showed a uniform spherical mass with low intensity

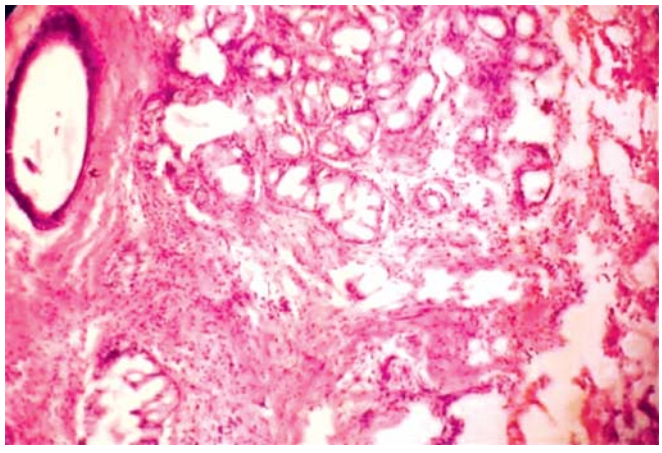

Fig. 5: H and E sections showing proliferation of mature adipocytes and the presence of entrapped salivary gland parenchyma (original magnification 10×)

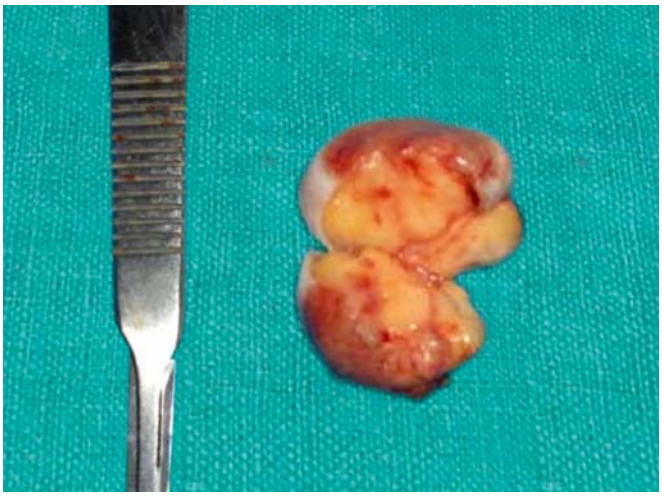

Fig. 3: Cut section of the gross specimen measuring around 1.5 × 1.2 cm

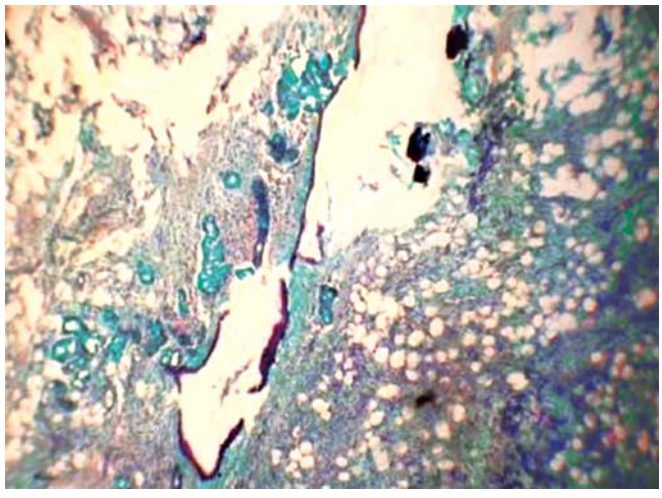

Fig. 6: Combined Alcian blue and PAS special stain showing atrophic mucous acini and marked dilation of the duct seen admixed with mature adipose tissue (original magnification 10×)

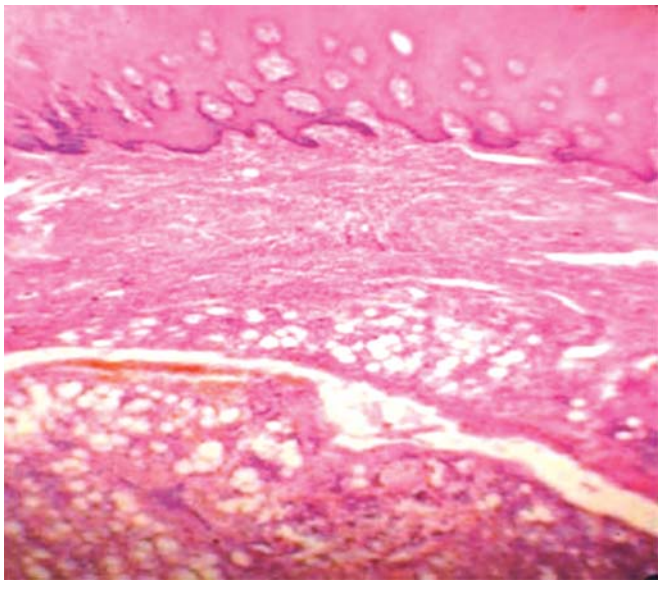

Fig. 4: H and E sections showing stratified squamous epithelium overlying the lesion proper and a demarcation of the tumor from the surrounding connective tissue (original magnification 4×)

overlying the lesion proper which was well circumscribed (Fig. 4). The lesion showed an admixture of adipose cells and glandular components consisting of effaced acinar units (Fig. 5). Some sections showed salivary duct dilatation and few areas showed normal salivary gland tissue. All these features were suggestive of sialolipoma. A combined special stain of Alcian blue and PAS demonstrated the mucous salivary glands and dilated duct (Fig. 6). A final diagnosis of sialolipoma was made and reported after analyzing the literature reviews.

DISCUSSION

Although lipoma is a common soft tissue tumor, oral lipoma, especially salivary gland lipoma, is rare. It is well known that major salivary glands are the most common anatomical location of the lipomas in the oral and maxillofacial region. Several histological variants, but not ordinary lipomas are found in salivary glands, e.g. angiolipoma, pleomorphic lipoma, spindle cell lipoma and lipoadenoma.[1] Fregnani et al in their analysis of 46 cases of intraoral lipomas showed that the most common

histopathological subtype is the ordinary lipoma followed by the fibrolipoma.[2] Lipomas in the hard palate are rare owing to the very little adipose tissue.[1,2] In fact, very few cases, only around eight cases of conventional lipomas have been reported till date in the hard palate.[1] Davis and Flaggert et al in their reviews of lipomas have reported three cases of angiolipoma, two cases of fibrolipoma and one case of ordinary lipoma in the hard palate.[6,7]

Nagoa et al retrospectively reviewed 2,051 cases of surgical specimens of salivary gland tumors and found seven cases, five in the parotid gland, one in the soft palate and one in the hard palate, showing features, i.e. the tumors were well circumscribed and encapsulated by thin fibrous tissue and were composed of mature adipose tissue and salivary gland; and tissue that was clustered or peripherally located within the tumor. They also came up with the concept of a new variant of lipomatous tumor, which they termed "sialolipoma", occurring in the salivary glands and showing the histologic features defined above.[3] In our present case, the tumor was situated in the hard palate showing salivary gland tissue within mature adipose tissue with fibrous encapsulation, which lead us to diagnose it as sialolipoma after clinical and histopathological correlation.

Since Nagao et al presented seven cases of the new variant of lipoma, sialolipoma, many new cases are reported till date. Sialolipomas share similar clinical features with conventional lipomas of the salivary glands. A literature search by Okada in 2009 revealed a total of 24 cases of sialolipoma, affecting the minor and major salivary glands in 11 and 13 cases. The data for sialolipomas indicate that all of the cases affecting the minor salivary glands occurred in adults (average age 64 years) whereas among cases affecting the major salivary glands three were pediatric (newborn, 3 and 11 years), and the average patient age was 41 years. Sialolipoma of the major salivary glands showed a male predilection, whereas no distinct sex predilection was evident for cases affecting the minor salivary glands. Minor salivary gland sialolipomas tended to be smaller (average greatest dimension 17 mm, compared with major salivary gland sialolipomas, 39 mm), despite having a longer duration time (all being present for more than a year).[8]

Our case presented in the hard palate involving the minor salivary glands with a history of swelling for almost 20 years and measured around 15 mm. In the latest report, Moraes M et al analyzed 35 cases of sialolipoma 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 four 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%).[5] Our case was in a 45-year-old male and had developed pain in the region of swelling because of a recent thermal injury.

Some reports have documented the CT and/or MRI findings of sialolipoma and stated that these imaging modalities may be diagnostically useful because sialolipoma appears as a well-circumscribed tumor with a low-intensity CT signal and high MRI intensity.[8,9] In our case, CT showed a low-intensity CT signal, and MRI was not carried out. Okada et al in their review of sialolipomas have compared the clinical and histopathogical features. They found both minor and major salivary gland sialolipomas showed atrophy of the glandular component, duct dilatation, lymphoid infiltration and oncocytic metaplasia, whereas periductal fibrosis was evident only in cases involving the minor salivary glands. They analyzed that proliferation of adipose tissue in the minor salivary glands would cause saliva retention in the duct and microcirculation disturbance, resulting in common histological changes, such as duct dilatation and glandular atrophy. Histopathologically, they categorized the pathological findings which included duct dilatation (9 cases), atrophy of the glandular component (7 cases), periductal fibrosis (5 cases), lymphoid infiltration (3 cases), enlarged congested vessels (3 cases), oncocytic (2 cases) and squamous ductal metaplasia (1 case), myxoid degeneration (1 case) and peripheral nerve involvement (1 case). Excision was performed in all cases they had reviewed, and none of them recurred. They concluded that there were some clinical and histopathological differences between sialolipomas affecting the minor and major salivary glands.[8] Histologically, in our case, there was atrophy of the glandular components and few areas showed effaced acinar units. Abnormal ductal dilatation was seen in one area and there was no lymphoid infiltration or oncocytic metaplasia seen in our case. Excision was performed in our case and it has not recurred till date.

The differential diagnosis, histopathologically, for sialolipoma is lipoadenoma,[10] pleomorphic adenoma with extensive adipose tissue[11] and lipomatosis.[12] Yau reported a case of lipoadenoma which was composed of neoplastic adipose tissue and neoplastic ductal components without normal acinar cells. Yau and Lin et al concluded that the presence of acinar cells allowed the differentiation of sialolipoma from lipoadenoma.[8,13] Lin et al reported a rare case of pleomorphic adenoma with extensive adipose tissue. They concluded that the presence of normal salivary gland tissue with ductal dilatation and fibrosis were the features that could distinguish sialolipoma from pleomorphic adenoma.[13] Okada and Lin et al have distinguished lipomatosis from sialolipoma by the presence of a fibrous capsule which occurs in sialolipoma but is absent in lipomatosis.[8,13] Okada from their immunohistochemical study observed that the glandular components of the tumor showed the same reactivity as the existing normal palatal gland, and had low proliferative activity, suggesting secondary entrapment. Furthermore, in their review they noticed the duct dilatation was recognized in all cases. They concluded that the low proliferative activity of the glandular component suggests that the entrapment is not attributable to true neoplasia, but occurred secondarily.[8]

In conclusion, the presence of residual salivary gland units in amid lipomatous proliferation is a rare event in oral cavity. Sialolipoma, the new variant of lipoma, can occur in any location

with salivary gland and adipose tissue. After its first description as a new variant of lipoma many reports have surfaced. Our case, which was diagnosed as lipoma with salivary gland elements, was rediagnosed based on the new reports. This could be the sixth case involving the hard palate. More retrospective analysis may reveal a higher incidence of this new variant, because some cases diagnosed as ordinary lipoma with salivary gland elements may be rediagnosed as sialolipoma.

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