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## CASE REPORT

# Island of salivary gland in adipose tissue: a report of three cases

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**Although uncommon, many variants of lipomatous lesions in or around salivary glands have been reported in the literature. We report a series of three such cases in the minor salivary gland region. The first case (oral floor) is a well-circumscribed lipocytic lesion admixed with glandular components (mucous acini, serous demilunes and ducts). The second case (alveolar mucosa) is a diffuse lipomatous proliferation with entrapped salivary glandular elements, muscles and blood vessels. The third case (palate) is similar to the first case but the gland is located at the periphery of the lesion. The purpose of the article was to report these three lesions and discuss in relation to other pertaining lipomatous lesions (sialolipoma, lipoadenoma, lipomatosis, lipometaplasia in pleomorphic adenoma and infiltrating lipoma).**
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## Case reports

*Case 1*

A 60-year-old man presented with a 2-cm pedunculated mass covered by normal appearing surface in the oral floor. Microscopically, under the cover of a thin and stretched surface epithelium, lobular proliferation of the lipomatous tissue containing clustered salivary gland elements were found (Fig. 1). The acinar cells include mucous, serous demilunes and serous. The ductal components showed proliferations and include intercalated, striated or oncocytic. Ductal dilatation and fibrosis were also evident. These glandular elements were scattered throughout the lipomatous lesion. Both the glandular and adipose components were found in almost equal proportion (Fig. 2). A diagnosis of sialolipoma was rendered. There is no recurrence 2 years after surgical excision.

*Case 2*

A 55-year-old woman presented with a painless raised diffuse lesion over the left mandibular alveolar mucosa of 1-month duration. There was no history of trauma or extraction. Her medical history was not contributory. On general examination, no abnormalities were detectable. Intra-orally, the lesion was found to extend from the first molar to third molar region, measuring 2.5–3.0 cm. Microscopically, it was characterized by diffuse proliferation of adipocytes with entrapped residual mucous salivary gland acini and ducts (Fig. 3). The ducts consisted of dilated ducts with papillary projections lined by a double row of oncocytic cells and a duct with squamous and oncocytic changes. Skeletal muscle and blood vessels were also found within the main lesional mass of adipocytes (Fig. 4). No mitotic figures or atypical cells were observed. A diagnosis of infiltrating lipoma was made. There is no recurrence 8 months after excision.

*Case 3*

A 70-year-old man presented with a painless mass measuring 2.5 × 2 cm on the right side of the junction of hard and soft palate. Microscopically, it was made of mature adipocytes bounded by a fibrous tissue under the cover of a thin surface epithelium. Normal uninvolved salivary gland tissue and a few skeletal muscle fibers were found at the periphery (Fig. 5). A diagnosis of lipoma was rendered. There is no recurrence 1.5 months after excision.

## Comments

Lipomatous lesions in the context of salivary glands have attracted interest in recent years and include lesions, such as sialolipoma, lipoadenoma, lipometaplasia in pleomorphic adenoma and lipomatosis. Sialolipoma is primarily a lipoma intimately admixed with relatively normal salivary glandular tissues (1). A circumscribed nodule of adenomatous ductal

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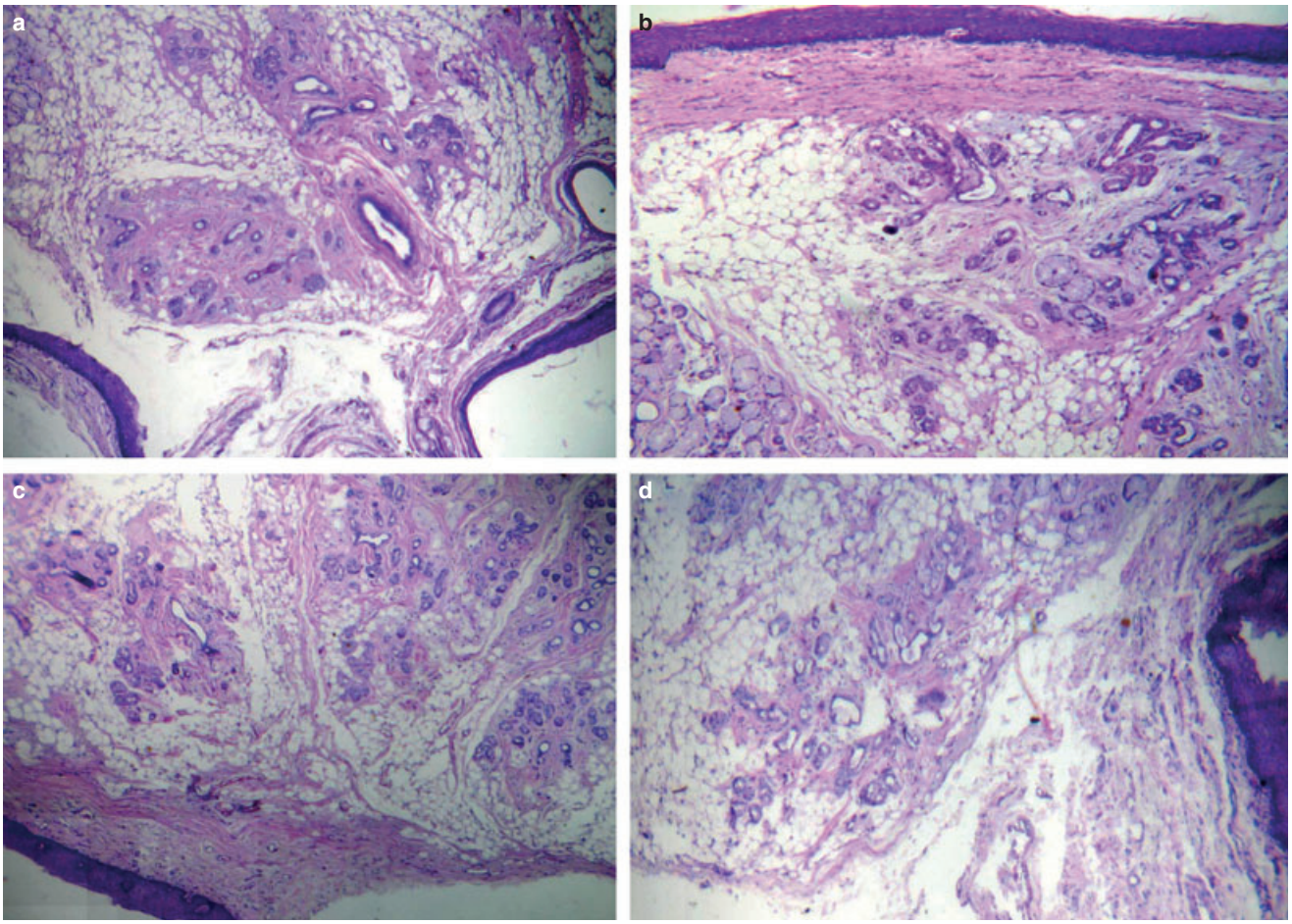


**Figure 1** Case 1: shows greater part of the well-circumscribed lesion containing adipose tissue, acini, ducts in (a–d) (Original magnification ×20, ×24, ×24, ×20, H&E).

proliferation devoid of acinar differentiation and myoepithelial component associated with adipocytes is referred to as lipoadenoma (2, 3). Occasionally, lipocytic component constitutes over 60–70% of the total tumor volume in pleomorphic adenoma as a result of lipid accumulation in myoepithelial cells. This phenomenon is more descriptively referred to as lipometaplasia (4). Unlike the three encapsulated lesions, lipomatosis is a non-neoplastic lesion that lacks circumscription but occurs in the clinical setting of diabetes, liver cirrhosis, chronic alcoholism and old age (5). The purpose of the present article was to report three cases of lipomatous lesion in association with minor salivary gland elements and discuss in relation to the other pertaining lipomatous lesions reported in the literature.

Oral lipomas developing in association with salivary glandular elements have occasionally been recognized in surgical specimens and have been designated as sialolipoma by Nagao *et al*. in 2001 (1). It was considered as a distinct variant of salivary gland lipoma that can occur in both major and minor salivary glands (1).

According to Nagao *et al*. (1) sialolipoma is a lipomatous proliferation containing acinar and ductal structures bounded by a very thin fibrous tissue. The amount of adipose tissue ranged from 50% in minor salivary glands to over 90% in major salivary glands.

The glandular elements may either be sparsely distributed or compressed peripherally in major salivary glands. On the other hand, it was clustered and evenly distributed in minor salivary glands.

Although it was not stated explicitly (1), the interpretation of the term sialolipoma implied definite origin within the salivary gland connective tissue stroma. While such a conclusion can be supported in major salivary glands, there may be argument over the use of the term in the context of minor salivary glands as there are no distinct demarcations to assess the origin more precisely. Yet the terminology is accepted because they still have exactly the same histomorphology regardless of the anatomic difference in the structure of these glands.

In 2003, Fregnani *et al*. (6) in their series of 46 lipomas found two salivary gland lipomas. While Lin *et al*. (7) in 2004 and Sakai *et al*. (8) in 2006, reported a case of intra-oral lipoma in association with salivary glands under the term sialolipoma. Their case appears to represent a lipoma with peripherally adherent salivary gland tissue. This is at variance with the sialolipoma of

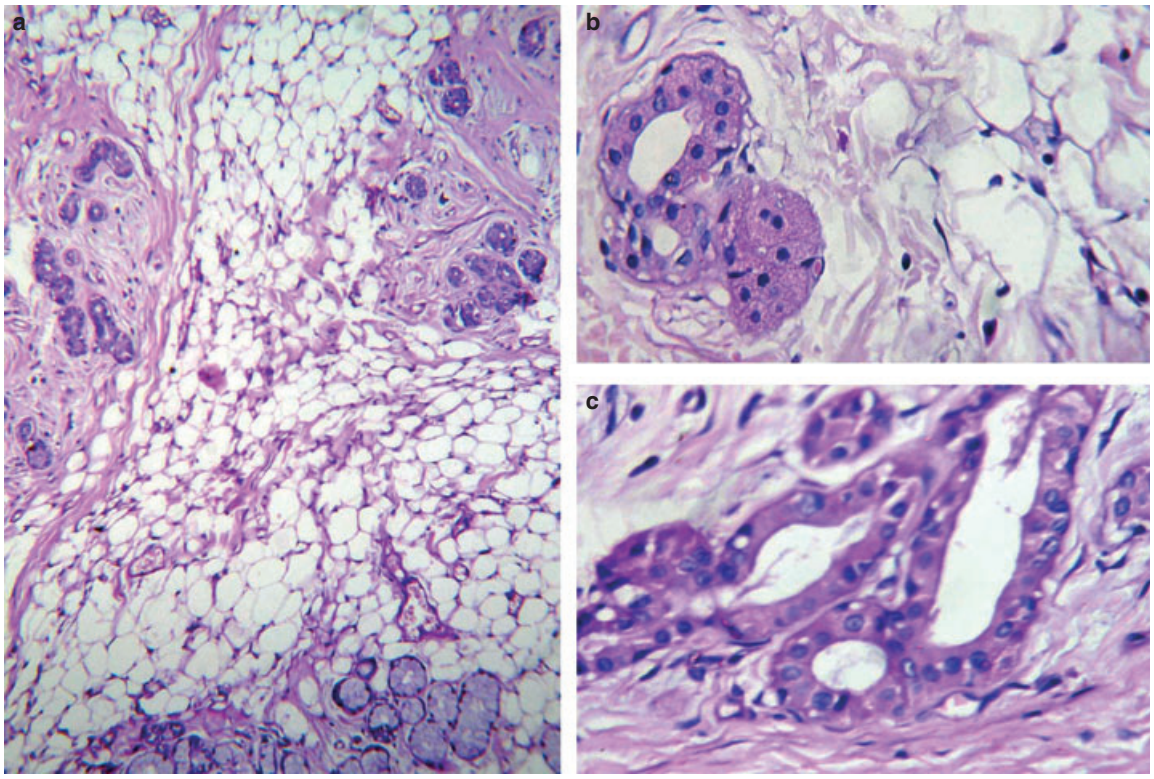


**Figure 2** Case 1: in this field clustering of glandular elements with fibrosis intermixed with adipocytes are evident in (a). Note: serous demilunes. Oncocytic and striated ducts are evident in (b,c) (Original magnification ×100, ×240, ×200, H&E].

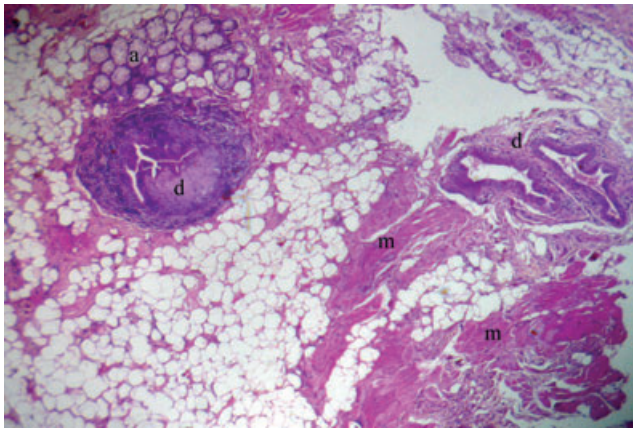


**Figure 3** Case 2: shows an island of salivary gland acini (a) and ducts (d), and skeletal muscle (m) surrounded by a lipomatous proliferation (Original magnification ×24, H&E).

Nagao *et al.* (1) which contains entrapped salivary glandular elements within the main lesion. In this sense, sialolipoma therefore more specifically refers to a proliferation of adipocytes around salivary gland elements bounded by a fibrous capsule. The salivary gland lipoma reported by Fregnani *et al.* (6) meets this requirement for a sialolipoma.

In the cases presented here, the first case is a well-circumscribed pedunculated lesion that contains both adipocytes and salivary glandular elements in almost equal proportions, whereas in the second case, the residual salivary gland and mesenchymal elements were virtually marooned by a lipomatous proliferation but without circumscription. This lesion also showed undulating ducts with papillary projections lined by a double layer of oncocytic epithelium. The third case, unlike the other two, was characterized by a circumscribed lipomatous proliferation with the normal salivary gland at the periphery.

From the preceding it would appear that the first case meets the criteria for a sialolipoma (1). But unlike the sialolipoma reported by Nagao *et al.* (1) this case is composed of relatively normal mucous acini, some of which were capped by serous demilunes and serous acinar differentiation. It was further characterized by the proliferation of intercalated, striated or oncocytic ducts. This is in contrast to the cases reported by Nagao *et al.* (1) which lacked ductal proliferations and is diametrically opposite to that of lipoadenoma or oncocytic lipoadenoma (2, 3). By definition, lipoadenoma is an encapsulated lesion characterized by neoplastic proliferation of both adipose and ductal components, but lacks acinar differentiation and myoepithelial component (2, 3). The possibility of pleomorphic adenoma with lipometaplasia was considered. As there are no areas that morphologically represent pleomorphic adenoma, it cannot be deemed as such (4). As ductal proliferation and acinar differentiation are not consistent with a diagnosis of either sialolipoma or lipoadenoma, it would be more appropriate to call this lesion as

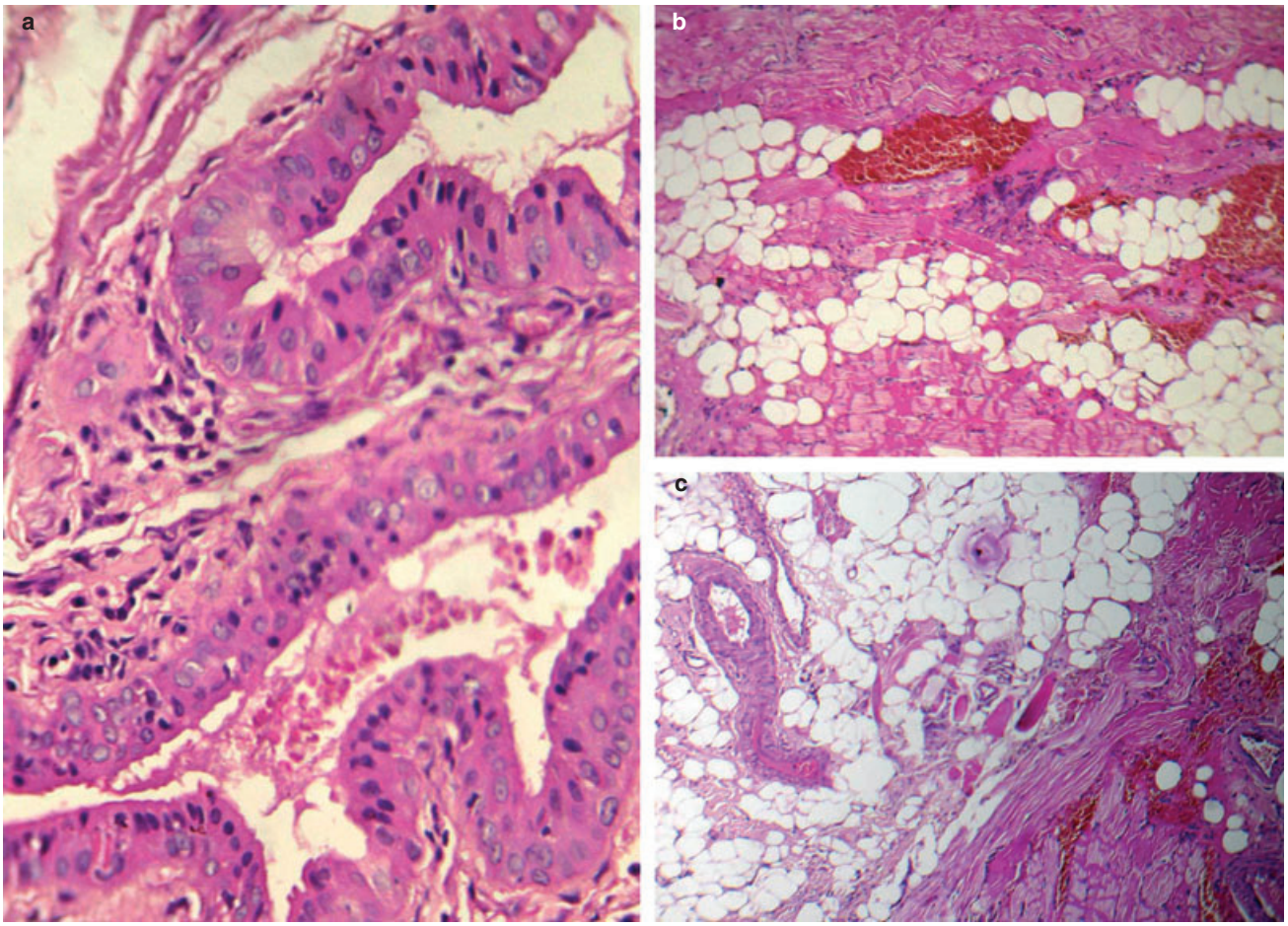


**Figure 4** Case 2: shows bilayered oncocytic cell linings with papillary projection into the lumen of the duct (a). Mature adipocytes and blood vessels infiltrating the muscle in (b) and the bulk of the adipocytes are found external to the muscle tissue in (c). Note: relatively large caliber vessel (Original magnification ×240, ×60, ×60, H&E).

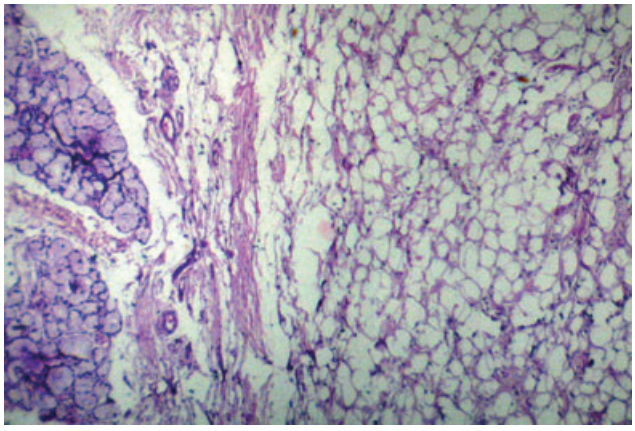

**Figure 5** Case 3: shows mature adipocytes and mucous salivary gland separated by fibrous tissue (Original magnification ×40, H&E).

'sialolipoadenoma' as it implies proliferation of both ductular and adipose components along with the presence of acinar units. However, as Nagao *et al.* (1) have stated that 'although the phenomenon of intermingling of salivary glandular tissue appears to be residual, it was still difficult to exclude a benign neoplastic process'. We therefore, exercise caution in interpreting this lesion as a distinct pathogenetic entity.

The second case is interesting as it is a diffuse lipomatous lesion with entrapped residual acinar tissue and dilated ducts lined by a double layer of oncocytes that could be confused with papillary oncocytic cystadenoma or oncocytic lipoadenoma. The cystic spaces in papillary oncocytic cystadenoma are characteristically lined by a double layer of oncocytes and evenly distributed oncocytes admixed with lipocytes are features of oncocytic lipoadenoma but both are well-circumscribed lesions (3, 9). Moreover, in lipoadenoma, oncocytes are arranged in an acinar or microglandular pattern and not as cystically dilated ducts (3), and lipomatous stroma is not a feature of papillary oncocytic cystadenoma (9). The oncocytic changes, noted here, are more likely to be the result of metaplasia of the duct system that occur with increasing severity in the older subject (10). Other alternative diagnoses that need to be differentiated in this case include, infiltrating angiolipoma or intramuscular angiolipoma and lipomatosis. The fact that the bulk of the lipocytic proliferation was external to the muscle tissue and the presence of

relatively thick vessels together with the lack of microthrombi favor a diagnosis of infiltrating lipoma (11). On the other hand, exclusion of lipomatosis can be made on the basis of lack of clinical findings and pertinent histories attributable to Madelung's disease (5). Traumatic pseudolipoma, in view of the short duration of the present lesion, may also be considered. But it characteristically presents immediately or within hours of the initial injury in a young patient (median age 20 months) with specific site predilection (12).

Our third case resembles the cases reported by Lin *et al*. (7) and Sakai *et al*. (8), in that the main lesion lacked admixture of salivary glandular elements and lipocytes, although it was circumscribed. With exceptions, almost all parts of the oral cavity contain salivary glands although at variable amounts. So, any intra-oral lipoma that occurs in these sites may intermingle with adjacent salivary glandular elements. But to categorize a lesion as sialolipoma in the context of minor salivary glands, we believe in line with Nagao *et al*. (1) that the amount of adipose tissue and entrapped glandular elements should be in equal proportions limited peripherally by a fibrous capsule. Therefore, the cases reported by Lin *et al*.(7) and Sakai *et al*. (8), as well as our third case may simply be a lipoma rather than sialolipoma.

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