J Oral Pathol Med (2006) 35: 376–8

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CASE REPORT

Sialolipoma of the hard palate

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Sialolipoma is a new variant of salivary gland lipoma, which was first proposed by Nagao et al. (*Histopathology* 2001; 38: 30) in 2001. We report this rare case of sialolipoma in the hard palate. A 60-year-old Japanese woman was referred to our department complaining of a painless swelling on the right side of the hard palate. Intra-oral examination revealed a soft, elastic, dome-shaped mass with 1 cm in diameter located in the posterior part of the hard palate. Magnetic resonance imaging examination revealed high intensity on T_1-weighted image and isointensity on T_2-weighted image. Incisional biopsy revealed that the tumor was encapsulated by fibrous tissue, consisted of adipose tissue, and also contained normal salivary gland tissue peripherally. First diagnosed as an ordinary lipoma of the hard palate, the tumor was excised. According to the recent criteria of histologic findings of sialolipoma, we rediagnosed the tumor as sialolipoma of the hard palate.
J Oral Pathol Med (2006) **35**: 376–8

Keywords: hard palate; lipoma; minor salivary gland; sialolipoma

Case report

A 60-year-old Japanese woman was referred to the First Department of Oral and Maxillofacial Surgery, Osaka University Dental Hospital, complaining of a painless swelling on the right side of the hard palate in April 1999. The patient had noticed this swelling more than 10 years ago. Intra-oral examination revealed the presence of a well-defined, dome-shaped mass that was $1.2 \times 1.8 \times 1.0$ cm and was located in the hard palate just to the right of the midline. The posterior margin of the lesion was approximately on the posterior border of the hard palate, and the lesion was compressible. The overlying mucosa was intact and of normal color. Magnetic resonance imaging (MRI) examination revealed hyperintensity on T_1-weighted image and isointensity on T_2-weighted image, which are similar to that of subcutaneous fat (Figs 1 and 2). Under clinical diagnosis as the minor salivary gland tumor, an incisional biopsy was carried out, and the soft yellowish tissue encapsulated by fibrous tissue was observed (Fig. 3). Microscopic examination revealed that the tumor encapsulated by fibrous tissue consisted of adipose tissue, and salivary gland tissue was located peripherally within this tumor. These findings allowed us to diagnose as this lipoma of the hard palate. The tumor excision was performed with a 1-cm margin under local anesthesia. Histopathologically, the lesion consisted of mature adipocytes containing minor salivary glands, and they were encapsulated by thin fibrous tissue (Fig. 4). The palatal minor salivary gland adjacent to the tumor was compressed by the tumor. We initially diagnosed the tumor ordinary lipoma, which contains ectopic salivary gland tissue, but we changed the diagnosis to sialolipoma based on the first report of Nagao et al. (1) in 2001. The post-operative course was uneventful, and no sign of recurrence was observed.

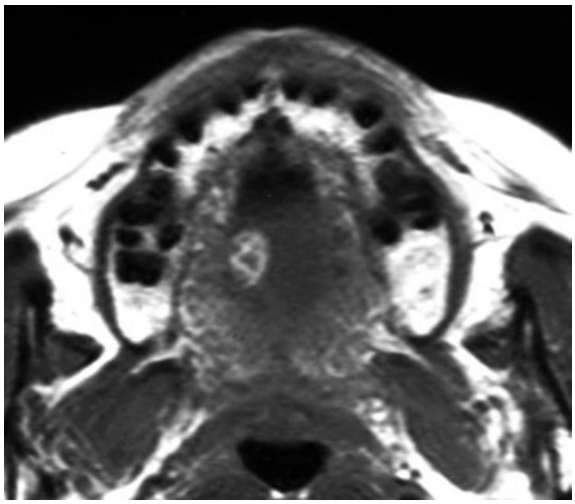

Figure 1 T_1-weighted axial magnetic resonance (MR) image shows an ovoid mass, its high signal intensity similar to that of subcutaneous fat, at the right posterior aspect of the hard palate.

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Accepted for publication December 6, 2005

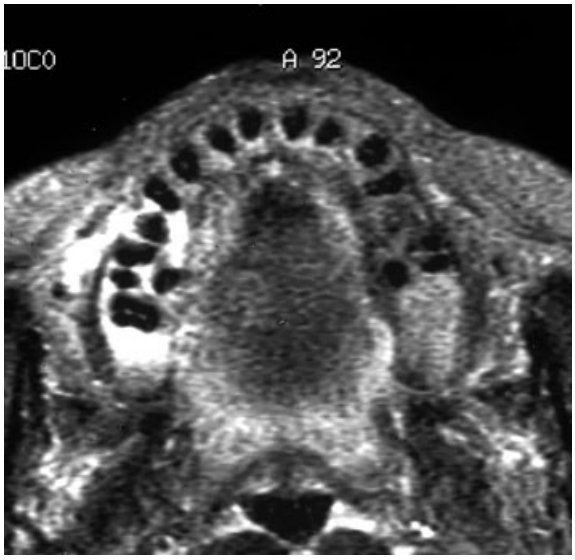

Figure 2 T_2-weighted axial magnetic resonance (MR) image shows an isointensity ovoid mass at the right posterior aspect of the hard palate.

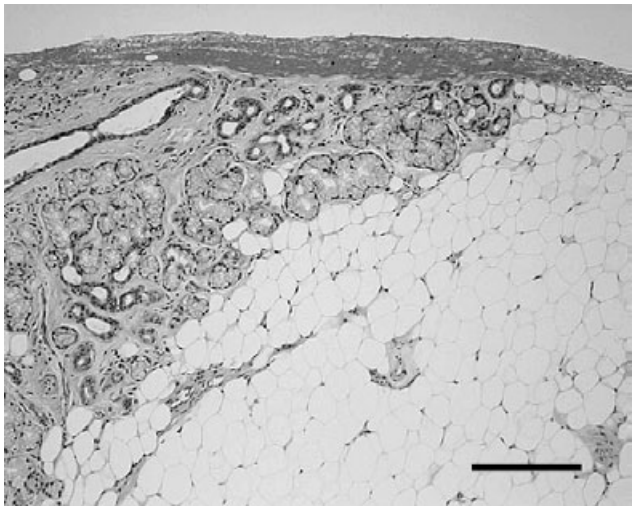

Figure 3 Photomicrograph of the biopsy specimen shows the tumor consisted of mature adipose tissue and mucous salivary gland with clearly defined margin [haematoxylon and eosin (H & E), bar: 100 μm].

Comments

Lipomas in the oral cavity are relatively rare. The most common locations of lipomas in the oral cavity have been reported to be in the buccal mucosa, a region abundant in fatty tissue (2), followed by the tongue. The hard palate has very little fatty tissue, and the incidence of lesions here is quite low (2, 3). In fact, to the best of our knowledge, only seven cases of lipoma in hard palate have been reported: three cases of angiolipoma (4–6), two cases of fibrolipoma (7), one case of spindle cell lipoma (8), and one case of ordinary lipoma (9). In addition to these, three cases of lipoma in the hard palate were discussed in previous reviews: two cases by Furlong et al. (2) and one case by Nagao et al. (1).

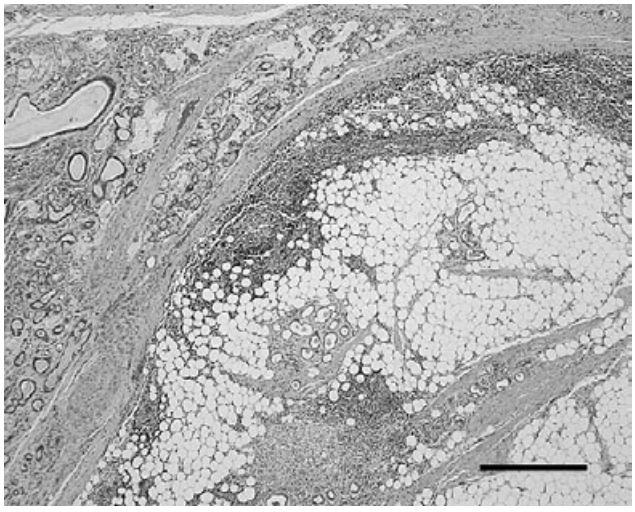

Figure 4 Photomicrograph of the excised specimen shows that the original palatal minor salivary gland is presented beside the tumor and was clearly separated from the tumor by fibrous tissue [haematoxylon and eosin (H & E), bar: 200 μm].

Lipomas can be classified histopathologically into subtypes, and their frequency in the oral cavity is different among the reviews. Fregnani et al. (3), who analyzed 46 cases of intra-oral lipomas, showed that the most common histologic subtype is the ordinary lipoma followed by the fibrolipoma, but Furlong et al. (2) reported that spindle cell lipoma was rather common. It is well known that major salivary glands are the most common anatomical location of lipomas in the oral and maxillofacial region (2), and several histologic variants of lipoma, but not ordinary lipomas, are found in salivary glands, e.g. angiolipoma, pleomorphic lipoma, spindle cell lipoma, and lipoadenoma. On the other hand, some reports showed cases of well-circumscribed tumors composed of a lipomatous element and glandular tissue in varied anatomical sites except the oral and facial regions (1). This histologic feature can often be seen in cases of lipoma in the salivary gland, and Nagao et al. (1) retrospectively reviewed 2051 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 similar features; i.e. the tumors were well circumscribed and encapsulated by thin fibrous tissue and were composed of mature adipose tissue and salivary gland tissue that was clustered or peripherally located within the tumor. According to these results, Nagao et al. (1) advocated the concept of a new variant of lipomatous tumor, which they termed 'sialolipoma', occurring in the salivary glands and showing the histologic features, described above. Our present case, with a tumor showing normal minor salivary gland tissue within mature adipose tissue with fibrous encapsulation, allowed us to diagnose the tumor as sialolipoma.

Since Nagao et al. (1) proposed the new variant, two cases have been reported. Lin et al. (10) showed a case of sialolipma on the oral floor and Hornigold et al. (11) reported a case of congenital sialolipoma in the parotid

gland. On the other hand, Fregnani et al. (3) also presented two cases of intraglandular lipomas, one located in the tongue and one in the buccal sulcus, showing very similar histologic features to sialolipoma, but they did not use the term 'sialolipoma' for their diagnoses. As Nagao et al. (1) showed one case of sialolipoma in the hard palate, our present case is the second report of sialolipoma in the hard palate.

Some lipomatous lesions need detailed histopathologic examinations to distinguish from sialolipoma (1, 10). The lipoadenoma has histologic features similar to sialolipoma, but can be differentiated by the lack of acinar cells; lipoadenoma is composed of adipose tissue and duct components without acinar cells, while sialolipoma contains both acinar cells and duct components (1, 10). However, to distinguish them clearly, immunohistochemical analysis may often be required with the antibodies specific to acinar cells such as α-amylase and epithelial membrane antigen. Both lipomatosis in the salivary gland and pleomorphic adenoma with extensive fatty tissue should be distinguished as being different from sialolipoma (1, 10). The presence of fibrous capsulation can be easily distinguished from lipomatosis (1, 10) and the presence of normal salivary gland tissue with duct dilation and fibrosis can distinguish from pleomorphic adenoma (10).

The salivary gland components located in the tumor are considered to have originated from secondary entrapment from the adjacent minor salivary gland during lipomatous proliferation rather than representing true neoplastic elements (1). However, Lin et al. (10) indicated that it is uncertain whether the salivary gland tissue admixed, was secondarily entrapped within the lipoma, or merely included within the capsule of the lipoma, and they noted the necessity to study a large number of cases in the future.

In conclusion, sialolipoma can occur in any locations with salivary gland and adipose tissue, and future retrospective histologic analysis may reveal a higher incidence of this new variant, because some cases previously diagnosed as ordinary lipoma which was closely related salivary gland can possibly be rediagnosed as sialolipoma. Future analysis may clarify the pathogenesis of the salivary gland tissue within the tumor and also confirm this new histologic variant of lipomas.

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