SIALOLIPOMA OF THE FLOOR OF THE MOUTH: A CASE REPORT

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Intra-oral lipoma is a well-known entity, but lipomatous tumors including salivary gland tissue containing clustered or peripherally located ducts and acinar cells are uncommon. They are a newly recognized entity of salivary gland lipoma, designated sialolipoma. We describe a case of sialolipoma arising in the floor of the mouth presenting with apparently normal salivary gland tissue, as demonstrated by both histologic and immunohistochemical findings, in a 67-year-old female. Complete surgical removal of the tumor with preservation of the sublingual gland was implemented after a careful examination confirming that the lesion did not originate from the sublingual gland.

Key Words: lipoma, sialolipoma, mouth floor, minor salivary gland
(*Kaohsiung J Med Sci* 2004;20:410–4)

Intra-oral lipoma is a well-known entity, but lipomatous tumors characterized by salivary gland tissue containing duct and acinar cells are infrequent. They are a newly recognized entity of salivary gland lipoma, termed sialolipoma [1]. To our knowledge, only four cases of intra-oral sialolipoma of minor salivary gland origin have previously been reported in the English medical literature [1,2]. Here, we report an additional case of intra-oral sialolipoma arising in the floor of the mouth.

CASE PRESENTATION

A 67-year-old female noticed a painless, non-tender, slowly growing mass over the left floor of her mouth for about 1 year. She complained that this mass caused interference with speech, mastication, and movement of her tongue. There was no history of trauma or bleeding tendency. Clinical evaluation revealed a 3 × 2 cm soft-tissue lesion. The oral cavity was otherwise healthy. The patient's medical history was unremarkable. Head and neck, as well as whole-body examinations revealed no abnormalities. The differential diagnoses were ranula, salivary gland neoplasm, dermoid and epidermoid cysts, and lipoma. The patient underwent surgical treatment, during which a yellowish, soft, well-circumscribed mass was found (Figure 1). The mass shelled out easily, with no adhesion to the sublingual gland or submandibular gland duct. The patient is well and has no evidence of disease after 2 years.

On gross examination, the tumor was well circumscribed and measured about 3 cm in maximum diameter. It was soft in consistency and yellowish in color. The cut surface was yellowish-white with a smooth greasy texture (Figure 2). Histologically, the tumor was encapsulated by a thin fibrous tissue and was predominantly lipomatous in nature (Figure 3). Islands of salivary gland tissue composed of mucous acini and duct dilatation with fibrosis were found around the periphery of the tumor (Figures 3 and 4). Multiple enlarged congested vascular channels were present (Figures 3 and 4). The acini in the tumor consisted of periodic acid Schiff- and mucicarmine-positive mucous glands. Immu-

Received: March 1, 2004 Accepted: April 2, 2004
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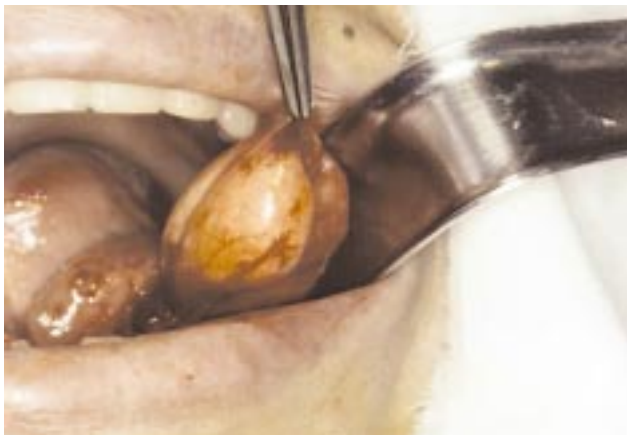

Figure 1. *Yellowish, soft, well-circumscribed mass found during surgery.*

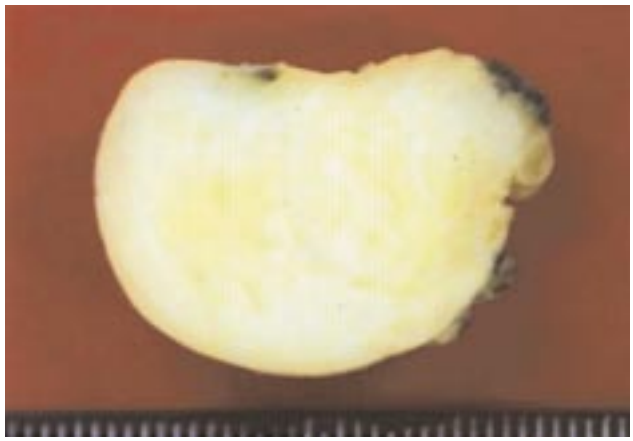

Figure 2. *Yellowish-white cut surface with smooth greasy texture.*

nohistochemical findings are summarized in the Table. Briefly, both the duct and acinar cells in the tumor were positive for keratin (AE1/AE3) (Figure 5) and epithelial membrane antigen. There were muscle-specific actin-positive cells and S-100-positive cells, suggesting myoepithelial cells, surrounding the acini (Figure 6). No Ki67 (MIB1)-labeled cells were identified in the tumor. Consequently, a diagnosis of sialolipoma of the floor of the mouth was made.

Discussion

The incidence of lipoma is higher in females than males, but intra-oral lipoma shows a predilection for males over the age of 40 years [3]. Most cases of intra-oral lipoma are slow growing and may reach a static size, usually less than 2 cm in diameter, with the only symptom a painless, palpable tumor [3]. However, lipoma occurring in the floor of the mouth may occasionally develop to an abnormal size [4], and may interfere with speech, mastication, or swallowing [5], as observed in the present case. Surgical excision as soon as possible is the treatment of choice. As far as we know, recurrence, associated with incomplete removal, has only occurred once [3].

Although trauma is considered an etiologic factor in lipoma occurring in other parts of the body, the etiology of intra-oral lipoma is not fully understood, and predisposing factors are not well defined [6]. No trauma history was identified in this case. It has been suggested that trauma might contribute to a patient's awareness of a pre-existing, asymptomatic lipoma rather than being the cause [7].

Microscopically, overt salivary gland tissues with both duct and acinar cells were observed in the present case. To

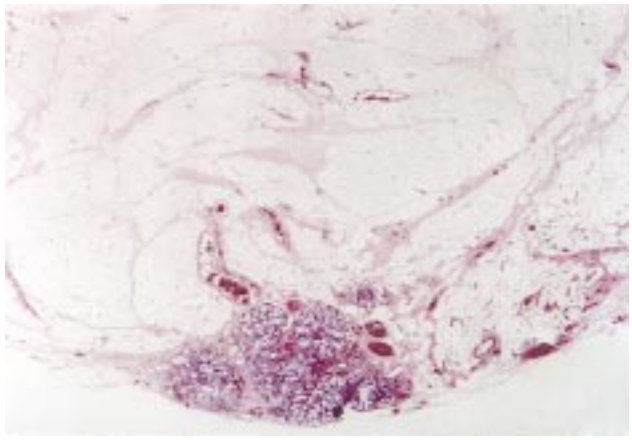

Figure 3. *Photomicrograph of the tumor illustrating its lipomatous nature and highlighting the presence of the encapsulating thin fibrous tissue; note the islands of salivary gland tissue localized around the periphery of the tumor (hematoxylin & eosin, original magnification ×40).*

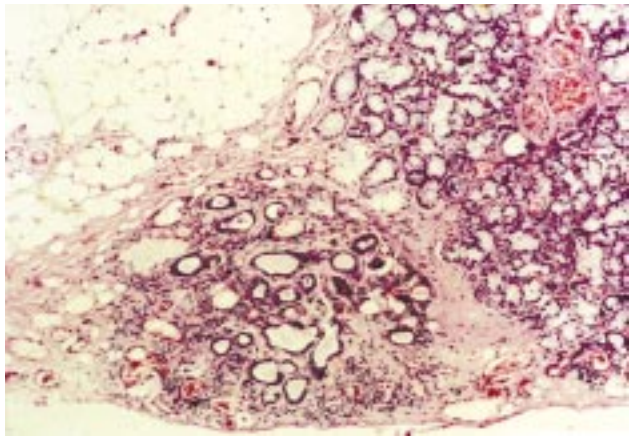

Figure 4. *Islands of mucous acini and duct dilatation with fibrosis within the encapsulating thin fibrous tissue (hematoxylin & eosin, original magnification ×100).*

Table. Summary of immunohistochemical findings

	Duct cells	Acinar cells	Adipose tissue	Antibody source (dilution)
Keratin (AE1/AE3)	+	+	–	DAKO* (1:50)
Epithelial membrane antigen	+	+	–	DAKO* (1:100)
Muscle-specific actin (HHF35)	–	+	–	Enzo Diagnostics† (1:50)
S-100 protein	–	+	+	Nichirei Corp‡ (1:10)
Ki67 (MIB1)	–	–	–	Immunotech SA§ (1:200)

*DAKO, Copenhagen, Denmark; †Enzo Diagnostics Inc, New York, NY, USA; ‡Nichirei Corp, Tokyo, Japan; §Immunotech SA, Marseilles, France.

our knowledge, benign tumors containing both adipose and salivary gland tissue were previously reported chiefly in the major salivary glands (parotid and submandibular glands) [1,8–12] and only four times in the minor salivary gland of the oral cavity (two on the palate [1], one each on the tongue and in the buccal cavity [2]). Therefore, the present case is the fifth documented case of intra-oral lipoma containing salivary gland tissue from the minor salivary glands.

Almost any site within the mouth may be involved, with most intra-oral lipoma occurring in the cheek, but also occasionally in the floor of the mouth [3]. To our knowledge, about 15 cases of intra-oral lipoma located in the floor of the mouth have been reported in the English medical literature [2,4–7,13–18], none of which showed involvement of salivary gland tissue. Therefore, the current case may be the first of intra-oral sialolipoma occurring in the floor of the mouth.

In 2001, Nagao et al proposed that a well-circumscribed mass composed of mature adipose elements and salivary gland tissues, which may be clustered or peripherally located within the tumor, is a distinct variant of salivary gland lipoma and designated it sialolipoma [1]. It has also been claimed that sialolipoma contains both duct and acinar cells, while another related lesion, salivary gland lipoadenoma, is composed of adipose tissue and duct components without an acinar component [12]. Our case has close histologic resemblance to, and consistent immunohistochemical findings with, cases described by Nagao et al [1].

Since the glandular tissue was located at the periphery of the tumor, it is postulated that the glandular components in this case originated from secondary entrapment from the adjacent minor salivary gland in the floor of the mouth during lipomatous proliferation rather than as an overt neoplastic element. This view is supported by the absence of Ki67 immunostaining, indicating low cell-proliferative activity in the glandular component of the tumor, and is also consistent with the findings of both Nagao et al [1] and Fregnani et al [2]. In addition, as shown by the immunohistochemical and histologic findings in the present case, glandular components closely resembled the cellular and structural compositions of normal salivary gland tissue. However, whether the salivary gland tissues were "ad-

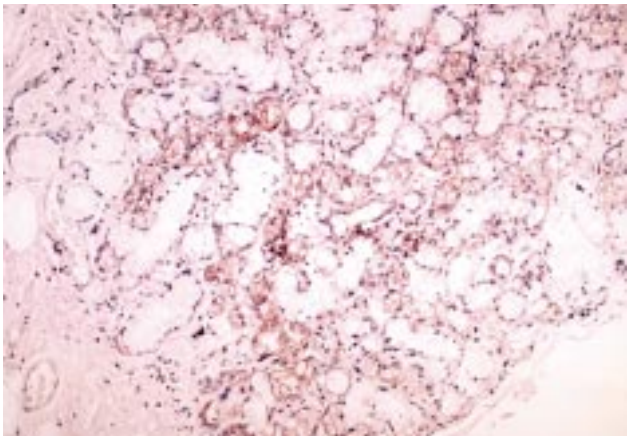

Figure 5. *Both the duct and acinar cells in the tumor are positive for keratin (AE1/AE3) (avidin-biotin-peroxidase-complex stain, original magnification × 40).*

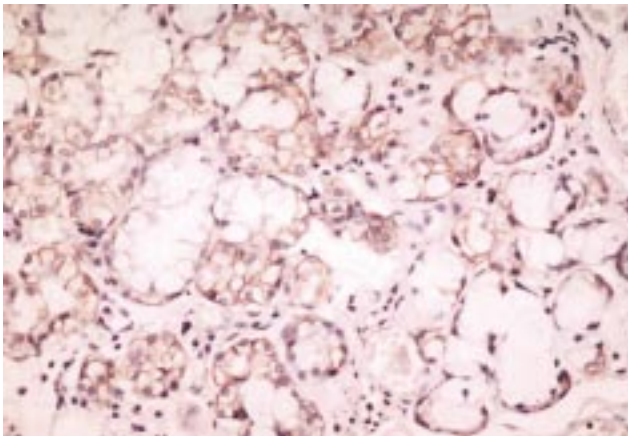

Figure 6. *S-100-positive cells, suggesting myoepithelial cells, surrounding the acini (avidin-biotin-peroxidase-complex stain, original magnification × 100).*

mixed" or "secondarily entrapped" within the lipoma or "merely included" within the capsule of the lipoma remains to be clarified. Ultrastructural study using electron microscopy could be helpful to solve this issue, but unfortunately, fresh tissue was not available. Therefore, further study in a greater number of cases is necessary before a conclusion can be reached.

Salivary gland lesions with extensive adipose tissue such as lipomatosis [19] and pleomorphic adenoma with extensive fatty component [20] should also be included in the microscopic differential diagnosis. The presence of a fibrous capsule can easily distinguish salivary gland lipoma from lipomatosis. On the other hand, the presence of apparently normal salivary gland tissue with only duct dilatation and fibrosis preclude the possibility of pleomorphic adenoma with extensive fatty element. Furthermore, malignant transformation of sialolipoma has not yet been reported.

Regarding the treatment of choice in the present case, complete surgical removal of the tumor is regarded as an appropriate treatment, similar to the ordinary lipoma, and there are no reported cases of recurrent sialolipoma. However, it is important to remember that oral floor lesions can develop that may not originate from the sublingual gland, as in our case. Therefore, prudent examination of the origins of lesions in the floor of the mouth is very important, and unconditional removal of the sublingual gland should not be the standard treatment.

In conclusion, the present case report is unique in two aspects compared to other previously reported cases of intra-oral lipomas. First, it documents an uncommon case of intra-oral lipoma characterized by salivary gland tissue. Second, it is a case of sialolipoma occurring in the floor of the mouth, which has not been previously described.

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發生在口底之含唾液腺脂肪瘤(sialolipoma)—病例報告

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脂肪瘤 (**lipoma**) 是一種已廣為人知的良性腫瘤，但在脂肪的腫瘤中含有唾液腺組織 (包括唾液腺管道與濾泡細胞) 並不常見。這種腫瘤是唾液腺的脂肪瘤中的新分類，稱之為含唾液腺脂肪瘤。本篇報告一個 **67** 歲女性發生在口底一個含有正常外觀唾液腺組織的含唾液腺脂肪瘤，並且從組織學與免疫化學染色得到證實。經過仔細地檢查，確定此一腫瘤不是從舌下腺而來，以手術方式將它完整從口底取出並保留完整的舌下腺。

關鍵詞： 脂肪瘤，含唾液腺脂肪瘤，口底，小唾液腺

(高雄醫誌 **2004;20:410—4**)

收文日期：**93** 年 **3** 月 **1** 日

接受刊載：**93** 年 **4** 月 **2** 日

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