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Case report

Angiolipoma of the cheek: A case report with a literature review

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ABSTRACT

The case of an angiolipoma of the cheek in a 76-year-old man is reported. Angiolipoma is a benign tumor which is very rare in the soft tissue of the oral region, with only 21 reported cases, including our case. In our case, microscopically, the mass consisted of mature adipose cells intermixed with scattered vascular components. The tumor had a low proliferative capacity and the capsule was present, indicating a noninfiltrating type. After surgical treatment, the patient has done well with no sign of recurrence for 3 months.

1. Introduction

Angiolipoma (AL), a subtype of lipoma, is a benign tumor in which adipose tissue and vascular components are intermingled. The tumor occurs commonly in the trunk and extremities, especially in the forearm [1,2]. However, AL is very rarely found in the soft tissue of the oral region, with only 21 reported cases, including our case. Here, we describe a case of AL in the buccal mucosa and present a literature review.

2. Case report

The patient was a 76-year-old Japanese male with a chief complaint of a mass in the left buccal mucosa that he had noticed before our initial examination. He had a medical history of hypertension and lumbago and an unremarkable family history. Physical examination showed a good physique and a good nutritional status, with no abnormalities in the trunk and extremities. A head and neck examination showed a symmetrical facial configuration, no abnormal sensation or motor palsy of the cheek, and no significant cervical lymph node enlargement. A non-tender, well-defined, soft mass measuring 20 mm × 20 mm was noted in the left cheek below the buccal mucosa. The mass was slightly moveable and there were no abnormalities of the overlying mucosa such as changes of thickness or color (Fig. 1).

Based on the clinical diagnosis of benign tumor of the buccal mucosa, surgical excision was performed under local anesthesia. Since the mass was surrounded by a thin capsule and was not adherent to the surrounding tissue, detachment was easy and the mass was removable as a lump. The specimen was 15 mm × 15 mm in size and the surface was smooth and red-yellow in color. The specimen was solid and a cross section was yellowish in color (Fig. 2).

Microscopically, the mass consisted of mature adipose cells intermixed with scattered vascular components based on H-E staining (Fig. 3). In immunohistochemical staining, the vascular element was positive for CD34 and smooth muscle actin, consistent with components of vascular endothelial cells and blood vessel smooth muscle, respectively. Regarding the proliferative activity, the labeling indexes for proliferating cell nuclear antigen (PCNA) and Ki-67 were 5.3% and less than 1%, respectively. Based on these results, the final diagnosis was AL. After surgical treatment, the patient has done well with no sign of recurrence for 3 months.

3. Discussion

AL is a subtype of lipoma that was first reported by Bowen in 1912 [1]. Most ALs occur just below the surface of the skin in the extremities, abdomen, chest and back, and especially in the forearms, and tend to be multiple [2,3]. ALs can occur at any age, but are relatively common in adolescence and the twenties [2,3]. Histologically, ALs consist of mature adipocytes and proliferating vascular endothelial cells and are classified into a noninfiltrating type with a capsule and an infiltrating type [2,3].

AL in the oral cavity was first reported by Davis et al. [4] as a tumor occurring in the hard palate. To our knowledge, there are only 21 cases of AL in oral soft tissues [4–22] including our case;

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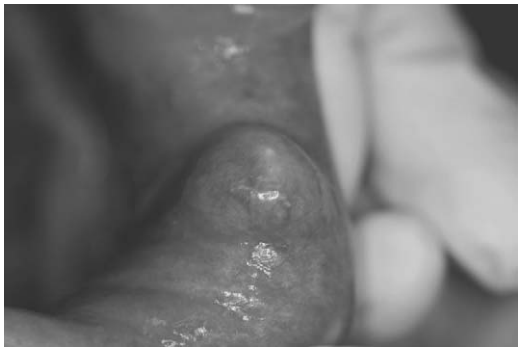

Fig. 1. The oral cavity at the first examination. A mass measuring 20 mm × 20 mm with normal overlying mucosa was present in the left buccal mucosa.

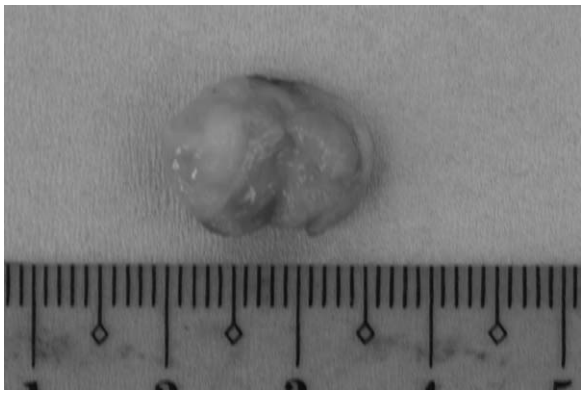

Fig. 2. The resected specimen. The specimen was 15 mm × 15 mm in size and surrounded by a thin capsule. The inside of the specimen was solid.

therefore, such cases are extremely rare. The 21 patients comprised 11 males and 10 females; i.e., the incidence in males is slightly higher. The age at the first consultation ranged from 1 to 81 years old and the mean age was 32.1 years old. The mean disease period was about 3 years and 2 months, and consequently the mean onset age was assumed to be about 29 years old (Table 1). This onset age does not differ significantly from that of 21 to 24 years old found in cases of systemic AL [2,3]. Compared with other common lipomas in the oral cavity, which have a mean onset age of 51.9–60.2 years old [23–25]. ALs in oral soft tissues appear to occur in younger patients. This may be because vascular components grow more rapidly than lipoma tissues.

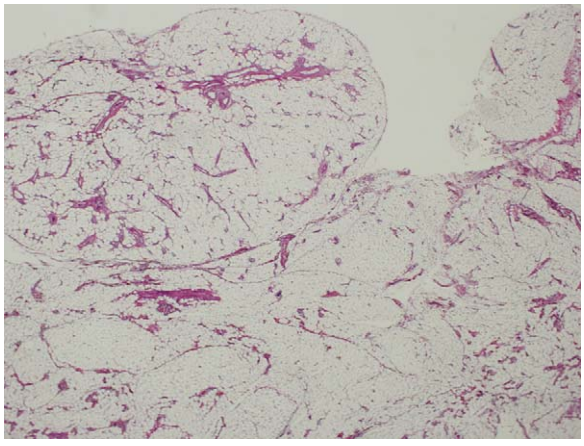

Fig. 3. Histopathologic findings. The mature adipose cells intermixed with scattered proliferation of vascular components were noted (haematoxylin and eosin, 40×).

Table 1
Cases of angiolipoma in the soft tissue of oral region (21 cases; 1976–2010).

Mean sideration age	32.1 (1–81)
Sex predilection	11:10 (male:female)
Chief complaint	Mass or swelling (tenderness, changes of overlying mucosa or skin)
Family history	(–)
Mean disease period	About 3.2 years (few days to about 20 years)
Region	Cheek: 13 cases (61.9%) Tongue: 3 cases (14.3%) Palate: 2 cases (9.5%) Lip: 2 cases (9.5%) Chin: 1 case (4.8%)
Mean major diameter	3.0 cm (0.5–8.0 cm)
Multiple tumors	(–)
Treatment	Excision: 11 cases (52.4%) Excision with surrounding tissue: 10 cases (47.6%)
Infiltrating type	Noninfitrating: 12 cases (57.1%) Infitrating: 7 cases (33.3%) Unknown: 2 cases (9.5%)
Recurrence	(–)

The chief complaints were a mass and swelling in all 21 patients and associated tenderness in 6 cases. Changes in the overlying skin or mucosa such as a yellow and pink coloring were observed in 7 patients and pain occurred in 4 of these cases (Table 1). In contrast, pain and discomfort are found in more than a half of patients with systemic AL, whereas abnormal findings in the overlying skin are rare [2,3]. AL in the oral cavity was not associated with family history or multiple onset (Table 1), whereas systemic AL occurred as multiple tumors in 197 of 248 patients and 11 of these patients had a family history of a subcutaneous mass [2,3]. These differences in pathosis suggest that AL in the oral region differs from systemic AL. Further evidence in more cases is required to evaluate this possibility.

The onset regions were the cheek in 13 patients, the tongue in 3, the palate in 2, the lip in 2 and the chin in one. The mean major diameter of the tumor was 3.0 cm (range: 0.5–8 cm). Most other lipomas in the oral cavity are reported to occur in the buccal mucosa [23–25], with an incidence of 36.4–45.7% and a mean major tumor diameter of 0.8–2.2 cm. Similarly, most of the 21 reported ALs in the oral region occurred in the cheek, but the tumor size was larger.

The histological type was noninfiltrating in 12 cases, infiltrating in 7, and unknown in 2 (Table 1). In a comparison of tumor types, most ALs in the oral region were noninfiltrating. Seven infiltrating cases have been described, with the cheek [17,19,22] in 3, the lip [16,21] in 2 and one each with the tongue [10] and chin [9] (Table 1). Seven of the 23 patients with systemic infiltrating AL had pain [3], and there was a similar tendency in patients with infiltrating AL in the oral region, with tenderness in 3 of the 7 cases. Three of the 12 patients with noninfiltrating AL also had tenderness, which is lower than for systemic AL, in which more than half of cases. However, tenderness may be characteristic of AL in the oral region because other common lipomas are painless.

Howard and Helwig [2] proposed a pathogenic mechanism in which lipoma caused by adolescent hormones is subsequently stimulated by trauma, resulting in angioproliferation in the lipoma. However, AL occurs several years after birth in some cases and the true cause remains unknown. History of trauma was found only in one patient with AL in the oral region, but it cannot be ruled out that continuous slight stimulation is involved in the onset of AL in the oral region, particularly since stimulation by food in the mouth can occur easily.

Most ALs are originally diagnosed as lipoma, but case reports since 1990 have used MRI, CT, sonography and aspiration biopsy to distinguish between hemangioma, lipoma and AL [11–22]. However, the accuracy of these methods remains low for these tumors

and a definite diagnosis in most literature cases has been made by histopathology. Differences in the percentages of adipose and vascular components among individuals make definite diagnosis of ALs difficult. In our patient, the clinical diagnosis was lipoma, but AL was diagnosed histopathologically.

PCNA and Ki-67 immunohistochemical staining can be performed to evaluate the proliferative capacity of a tumor. Fregnani et al. [24] found mean labeling indexes of 13.2% and 2.8% for PCNA and Ki-67, respectively, for all lipomas, with the highest indexes of 17.3% and 4.8%, respectively, found for fibrolipomas in the oral cavity. In our case, PCNA and Ki-67 immunohistochemical staining gave labeling indexes of 5.3% and less than 1%, which suggests that the proliferative capacity of the tumor was relatively low, although a simple comparison of the results is difficult.

The appropriate treatment for noninfiltrating ALs is surgical excision, with excision including the surrounding tissues recommended for an infiltrating AL in which the capsule is absent or partially associated with the tumor. There has been no report of malignant transformation and recurrence of AL in the oral region [average follow-up duration: 17.5 months (range: 3–36 months)] (Table 1), but the recurrence rate of infiltrating AL is high, ranging from 35% to 50% [3]. The AL in our patient had a low proliferative capacity and the capsule was present, indicating a noninfiltrating type, but the patient requires continuous follow up. To prevent recurrence of AL, it is important to remove the tumor and surrounding tissues in cases when the capsule is difficult to separate from these tissues during surgery, since diagnosis of the infiltrating type of the tumor is difficult preoperatively.

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