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CASE REPORT

A case of extragingival peripheral ameloblasotoma in the buccal mucosa

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Peripheral ameloblastomas (PAs) of the extragingival areas are extremely rare. To the best of our knowledge, only five cases of extragingival PA have been reported. We present here a sixth case of extragingival PA of the buccal mucosa in an 80-year-old male. The tumor was surgically removed by blunt dissection and there is no evidence of recurrence for 7 months. We also discuss here the clinical characteristics, the origin, and the management of the tumor by reference to the relevant literature.

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Case

An 80-year-old Japanese man was referred to Renaiss Kyoto Hospital for evaluation of a swelling on the left buccal mucosa. The lesion had been present for about 3 months and had gradually increased in size. The patient was positive for hypertension and under hormonal therapy for prostate cancer that was in good control. In the left buccal mucosa a firm, non-mobile mass could be felt. The mass was well defined and circular, about 2 cm in diameter, lying anterior and medial to the mandibular ramus. The surface mucosa was normal and non-ulcerated. The mass was intact to the left parotid duct and there was unimpeded salivary flow from the left parotid duct. Cervical lymph nodes could not be palpated. The remainder of the physical examination was unremarkable. Computed tomography (CT) and magnetic resonance imaging (MRI) revealed a well-demarcated circular mass existed anterior to the mandibular ramus (Fig. 1). The tumor was slightly enhanced in contrast CT scan and showed low signal in both T1 and T2 weighted MRI images. An incisional biopsy revealed that the micrographic findings were consistent with the PA. The tumor was removed through an intraoral approach under general anesthesia. There was no attachment to the mandible or maxilla and no communication with the surface epithelium. The tumor was well encapsulated and dissected free by blunt dissection with relative ease and was totally removed. The tumor was not adherent to either the parotid duct or gland. The tumor was a single, firm, lobular, whitish mass, measuring $2.0 \times 2.0 \times 2.5$ cm. Histologic examination revealed that the tumor was encompassed with fibrous capsule (Fig. 2a) and displayed two histologic subtypes; the plexiform (Fig. 2a) and follicular type (Fig. 2b). Periphery of the tumor was mostly indicative of the plexiform pattern that consisted of a proliferation of epithelium that forms cords and strands. The central area was mainly follicular with peripheral palisading cells with their nuclei polarized away from the basement membrane. The central cell population was composed of stellate reticulum-type cells. No mitotic figures and no tumor invasion in the surrounding tissue were seen. Subsequent to the surgical treatment, the patient has had no evidence of recurrence for 7 months.

Comments

Peripheral ameloblastoma (PA) is a relatively uncommon disease (1, 2) and PAs of the extragingival areas such as the buccal mucosa and floor of the mouth are extremely rare. To the best of our knowledge, only five cases of extragingival PA have been reported (3–7). Because of its rarity, we report here an additional case of extragingival PA in the buccal mucosa. Strictly, PAs in extragingival locations are excluded from the diagnosis of PA (2). We use the term of extragingival PAs for PAs in the extragingival areas.

A summary of the clinical features of the five previously reported cases of extragingival PAs and our case is presented in Table 1. The extragingival PA preferably occurred in elder males. The age range of

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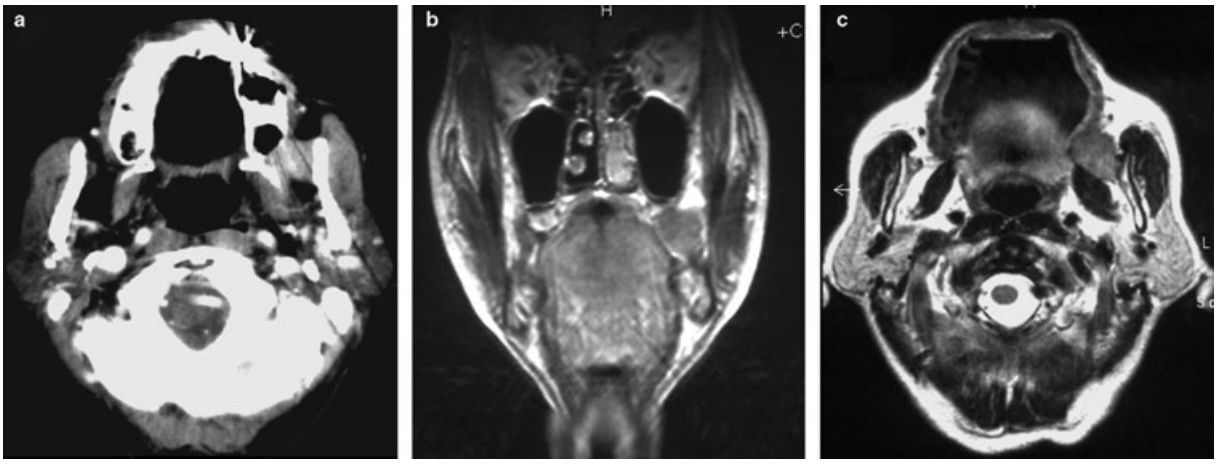

Figure 1 CT and MRI images. (a) An axial contrast CT scan shows a slightly enhanced mass in the anterior and medial portion of the left mandibular ramus. (b, c) Coronal T1 weighted and axial T2 weighted MRI images. Homogeneous soft tissue density areas are seen anterior to the left mandibular ramus.

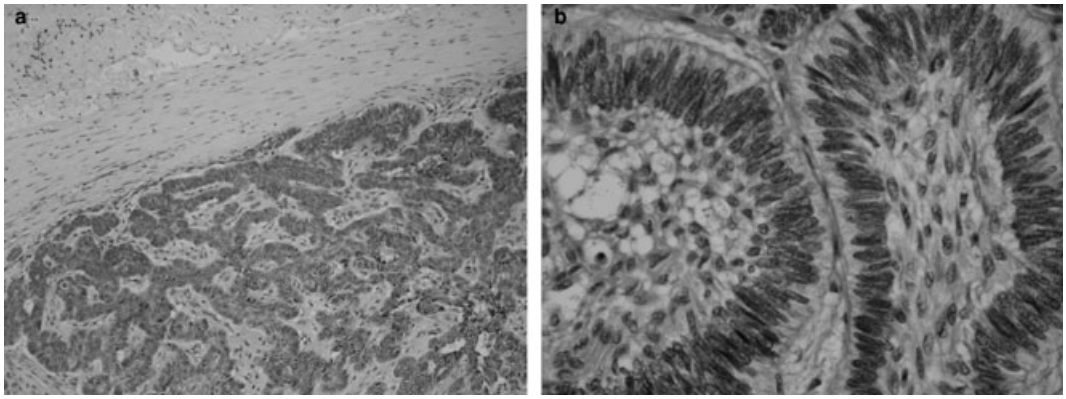

Figure 2 Histologic findings. (a) Periphery of the tumor shows fibrous capsule around the mass. Under the capsule, an anastomosing growth of the epithelium in strands and sheets is seen (HE, ×100). (b) The peripheral cells are palisade with their nuclei polarized away from the basement membrane. These cells delineate stellate reticulum-like areas with occasional spindled cells and acanthomatous areas (HE, ×400).

patients with extragingival PA was similar to that of patients with PA (2). With regard to gender of the patients, extragingival PAs occurred in five men and one women, while the male/female ratio in PAs and intraosseous ameloblastomas are reported to amount to 1.9:1 (2) and 1.2:1 (8), respectively. All patients with extragingival PA underwent surgery for the treatment. Among them only one patient received an excision with a wide margin, and in three out of the six patients the tumor was removed totally. No patient showed reappearance of the disease, although the follow-up periods were relatively short in every case.

Potential sources of extragingival PA include odontogenic remnants of vestibular lamina, pluripotent cells in the basal cell layer of the mucosal epithelium (6) and pluripotent cells of minor salivary glands (4). Shibata et al. thought that extragingival PA originates in the stratified squamosus epithelium because two out of the four patients with extragingival PA in the buccal mucosa showed continuity with the basal layer of the overlying epithelium (7). However, as Klinar and McManis described (4), the communication between the surface epithelium and the tumor can derive from expansion of the tumor stimulating concurrent growth of the normal epithelium. In our case, we could not find continuity between the tumor and the surface epithelium, suggesting that cells in the surface epithelium are not the origin of the tumor. We think the late occurrence and rapid growth are important characteristics of extragingival PA in understanding the pathogenesis of the tumor. The mean onset of the disease is 62.8 years of age in the six cases previously reported including our case. In three out of the six cases, the disease rapidly developed for 2–3 months before their first medical consultation. Braunstein and Mass (3) described their case as being unusual because of its rapid development. The development of the tumor in our case was so rapid that we had suspected the lesion of malignancy in our first examination. The rapid development of the tumor indicates the duration between the onset of the disease in practice and the time patients noticed it is short, suggesting the extragingival PA is a late-onset disease different from the intraosseus ameloblastoma. Therefore, we think that

Table 1 Cases of extragingival peripheral ameloblastoma

	Braunstein and Mass (3)	*Klinar and McManis (4)*	*Ramnarayan et al. (5)*	*Woo et al. (6)*	*Shibata et al. (7)*	*Yamanishi et al. (This report)*
Year	1949	1969	1985	1987	1990	2006
Age, sex	63, male	68, male	65, male	52, female	49, male	80, male
Location	Buccal mucosa	Buccal mucosa	Floor of the mouth	Female buccal mucosa	Buccal mucosa	Buccal mucosa
Ulcerated	–	+	–	+	+	–
Encapsulated	+	+	+	+	Unstated	+
Contact with the surrounding tissue	–	Surface epithelium	Surface epithelium	Surface epithelium, Stensesen's duct	Surface epithelium	–
Treatment	Blunt dissection (total removal)	Excision with wide margin	Excision	Total removal	Excision	Total removal
Size	$2 \times 2.5 \times 1.5$	$5.0 \times 4.0 \times 2.5$	$2 \times 1 \times 1$	$3.0 \times 2.5 \times 1.5$	$3.5 \times 2.5 \times 1$	$2.0 \times 2.0 \times 2.5$
Follow-up	4 months FOD	5 months FOD	6 months FOD	9 months FOD	12 months FOD	8 months FOD

FOD, free of disease.

factors that trigger generation of the extragingival PA would accumulate with aging or are directly related to genetic changes with aging.

For the treatment of the tumor, we chose total removal of the tumor by blunt dissection without wide surgical margin. The fibrous capsule was recognized in five out of the six cases with extragingival PA (unstated in the remaining one case). Multicystic and solid type of intraosseous ameloblastomas and PAs usually do not possess fibrous capsules (9). Capsules cannot be recognized in intraosscous ameloblastomas penetrating into the surrounding soft tissue (10). It is unknown why extragingival PA possess fibrous capsules, but this clinical characteristic would suggest the benign nature of the extragingival PA. We think that the first choice of treatment for extragingival PA is total removal of the lesion by blunt dissection. In one case, however, tumor cells were found in the capsule (4). Follow-up is very important after surgical treatment.

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