

Case Report

Ameloblastoma of the soft tissue: an unusual presentation and literature review

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Abstract: Ameloblastoma or adamantinoma is the rarest of the three forms of odontogenic-type tumor. These tumors are benign, locally aggressive neoplasms arising from ameloblasts. They typically occur at the angle of the mandible, and the occurrence of this tumor in soft tissue, i.e., peripheral ameloblastoma (PAM), is extremely rare. PAM shows a male preponderance, and the most common sites are the mandibular canine and premolar lingual gingivae. In this paper, we present a case of ameloblastoma of cheek mucosa, which is considered a rare site of occurrence. The patient presented in the hospital with swelling of the left-side cheek. The mass was visible and involved the buccal vestibule, and upper and lower alveolus regions. Ameloblastoma was confirmed after the biopsy, then resection of the tumor was performed.

Keywords: Ameloblastoma, peripheral ameloblastoma, odontogenic tumor, cheek mucosa

Introduction

Ameloblastoma is a rare primary bone tumor that commonly arises in the jaw; it has been described in the appendicular skeleton, e.g., the tibia. It is a benign, slow growing, and locally invasive odontogenic tumor [1, 2]. It accounts for 11% of all odontogenic tumors. The tumor frequently develops in the mandible (80%) and maxilla (16%). However, peripheral ameloblastoma (PAM) located in the soft tissue accounts for the remaining 1%-9.3% [3-5]. Thus, occurrence of the tumor in soft tissue is extremely rare. It occurs in all age groups, but the lesion is most commonly diagnosed in the third and fourth decades of a person's life [6]. The tumor is usually asymptomatic, painless, and present with bony deformity. Wide surgical excision with adequate safe margins is the treatment choice for this tumor. Here, we present a rare case, an ameloblastoma of oral soft tissue that was originally misdiagnosed as cancer. We also report clinical and pathological data with a review of the available literature on PAM.

Case presentation

An 82-year-old woman presented in the department of stomatology, Tianjin Medical University

General Hospital with swelling of the left-side cheek, which is associated with pain during chewing. The symptoms have been present for 2 months. Intra-oral inspection showed a mass in the left cheek measuring approximately 5.0 cm × 5.0 cm × 3.5 cm with an unclear border. The mass was visible and involved the buccal vestibule, and upper and lower alveolus regions. Mouth opening was limited. The mass bulged out from the buccal mucosa, kermesinus, slightly hard in quality, and obviously indurate (**Figure 1A** and **1B**). The MRI results revealed a soft tissue mass that has large multiloculated cystic expansile lesions arising from the buccal space (**Figure 1C**). The mandible and maxilla were not damaged. The masseter muscle and medial pterygoid muscle were involved in the MRI attenuation and dimensions. No obvious cervical lymphadenopathy was observed. The patient had no history of tumor. When the patient was admitted, benign tumor was not initially considered, and buccal cancer was favored. Laboratory findings were as follows: WBC: 7.59 × 10⁹/L, HB: 121 g/L, CREA: 65 umol/L, AST: 13 U/L, ALT: 8 U/L, TP: 59 g/L and ALB: 40 g/L. Biopsy was performed, and the pathological section of the tumor had a distinctive morphological characteristic of ameloblas-

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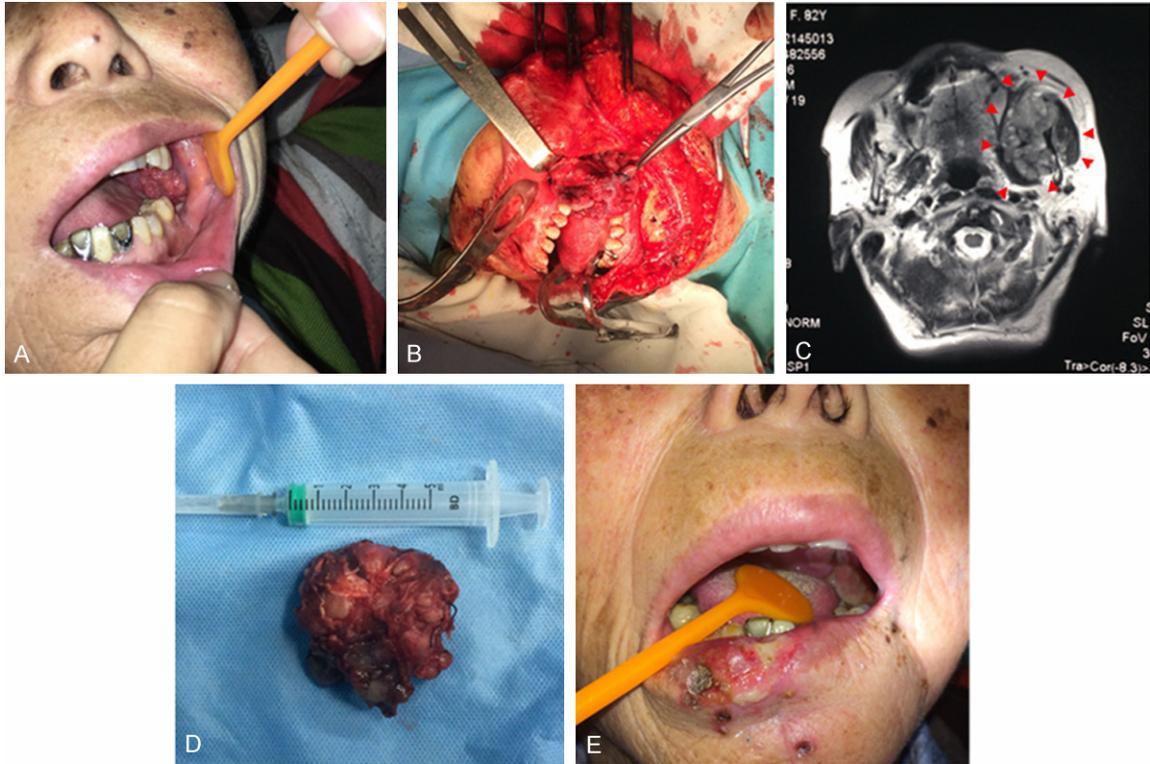


Figure 1. PAM in the cheek. A, B. Photograph shows the patient's cheek PAM. C. MRI shows the mass in soft tissue (arrows indication). D. The tumor. E. Surgical wound healing was good (one month after the operation).

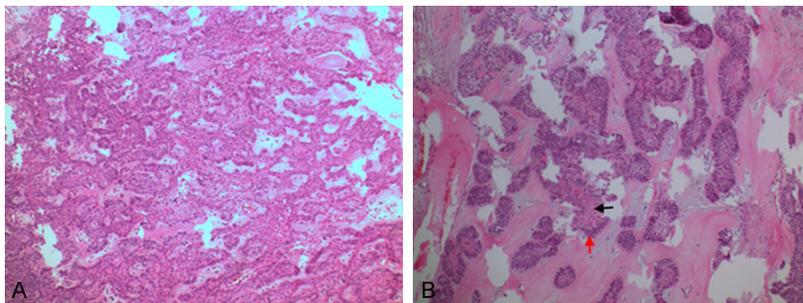


Figure 2. Hematoxylin and eosin-stained tumor section (A \times 100, B \times 200). The typical epithelial islands or cords constituted by two kinds of cells, one is cuboidal or columnar cell (red arrow), another is similar to the stellate reticulum (black arrow).

toma, so it was easily to diagnose as ameloblastoma (Figures 2 and 3). Under the microscope, epithelial islands were obviously mixed with fibrocollagenous tissue. The outermost layer of epithelial island was composed of tall columnar cells with polarization of the nuclei away from the basement membrane. The central portion of the island was composed of loose network of cells (Figure 4). Based on the above mentioned findings, resection of tumor

was planned. Under general anesthesia, the specimen was taken out, and the defect was covered by a cellular allograft dermal matrix membrane. The tumor did not recur after 20 months of treatment.

Review and discussion

Ameloblastoma is a benign odontogenic tumor of epithelial origin. It was described in 1827 by Cusack and designated as an adamantinoma in 1885 by the French physician Louis-Charles Malassez [7]. It was renamed as ameloblastoma in 1930 by Ivey and Churchill [7, 8]. Its etiology has not been determined. It constitutes about 1% of all head and neck tumors and about 11% of teeth-originating tumors. Ameloblastoma can be present in a wide range of ages, but these are usually diagnosed between the third and fourth decades of a person's life,

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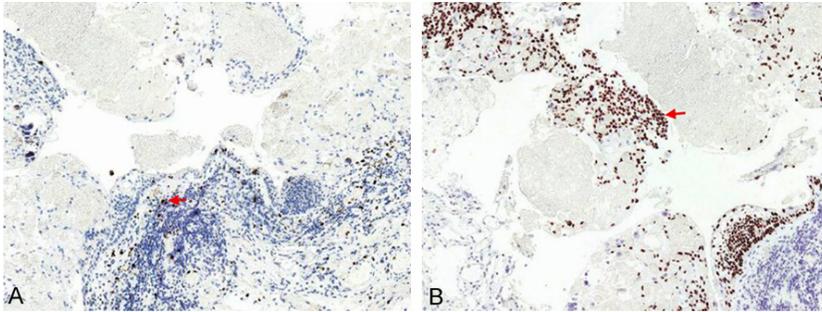


Figure 3. A. Immunohistochemistry for ki67 showing positive staining in the tumor cells $\times 100$. B. Immunohistochemistry for cytokeratin showing focal positivity in the tumor cells $\times 100$.

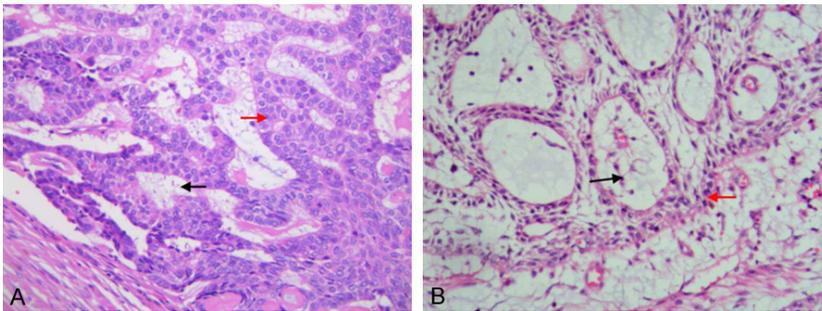


Figure 4. Hematoxylin and eosin-stained tumor section ($\times 400$, A and B are two different parts of the slice). The outermost layer of epithelial island was composed of tall columnar cells with polarization of the nuclei away from the basement membrane (red arrow). The central portion of the island was composed of loose network of cells (black arrow).

except in the case of the unicystic variety (20-30 years old). No gender predominance is noted [9].

Usually, it occurs in the mandible near premolar and molar teeth, but more rarely in its anterior part. The very rare location is the gingiva or the cheek mucosa. Different treatment modalities are surgical excision, enucleation, curettage, cryotherapy, radiotherapy, and chemotherapy. Wide surgical excision was advocated for its management to prevent its recurrence [10-13]. Surgical excision involves complete removal of tumor with negative margin of 15-20 mm [14]. Ameloblastomas are histologically classified into follicular, granular, plexiform, desmoplastic, basal cell, and acanthomatous varieties [15]. Diagnosis is usually made by fine needle aspiration or biopsy, which reveals a palisaded basal cell layer with stellate reticulum like epithelium. Differential diagnosis of ameloblastoma includes calcifying epithelial odontogenic tumor (CEOT), odontogenic myxoma, central giant cell granuloma, or ameloblastic fibroma.

PAM was first described by Kuru in 1911 [16] and a case report in 1949 was considered to be the first well-established case of PAM [17]. In the literature, 1%-9.3% ameloblastomas are peripheral cases, PAM accounted for 37%-67% of all peripheral odontogenic tumors [3]. The age of patients with PAM ranged from 3 years old to 92 years old. Most of the patients were from 40 years old to 59 years old, with an average age of 52 years old. The mean age was significantly higher than that of the intraosseous ameloblastoma (37 years old). PAM shows a male preponderance, the male to female ratio is 1.8:1 [18]. The mandible is a more common site than the maxilla, with the ratio being 2.5:1.

The most common site for PAM is mandibular premolar region (32.6%) followed by anterior mandibular region (20.7%) and maxillary soft palatal tissue of the tuberosity area (11.1%) [19] (**Table 1**). However, cases of PAM in the extralingival area are extremely rare. Only six cases of extralingival PAM have been reported [17, 20-25] (**Table 2**).

The size of the lesions was 0.5-4.5 mm, and it can be asymptomatic. The lesion also be manifested as an external mass. The color is similar to the surrounding mucosa. The surface is smooth or verrucous and papillary. As a result of local damage, the mass can become an ulcer or undergo keratinization. Bone absorption can occur under the oppression, but the X-ray showed no lesion on the jaw. If PAM occurs in the alveolar ridge, it may affect the pronunciation of words uttered by the patient [26]. Some cases are multicentric lesions reported in the literature [27, 28]. The pathology of PAM and intraosseous ameloblastoma is similar, but acanthomatous lesions are much

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Table 1. Comparison of intraosseous ameloblastoma (AM) and PAM

| | First reported year | Incidence | Mean age | Sex | Region | Pathology | Treatment |
|-----|---------------------|---------------------------------|----------|-------------------------|---|--|------------------------|
| AM | 1827 | 11% of teeth-originating tumors | 37 | No gender preponderance | Mandible near premolar and molar teeth | Classified into follicular, granular, plexiform, desmoplastic, basal cell, and acanthomatous varieties | Wide surgical excision |
| PAM | 1911 | 1%-9.3% of AM | 52 | Male preponderance | Mandibular canine and premolar lingual gingivae | Similar to AM, but acanthomatous lesions are common | Conservative resection |

Table 2. Reported cases of PAM in the extragingival area

| Authors | Year | Nation | Sex | Region | Treatment | Follow-up |
|--|------|--------|-------|---------------------|--------------------------|---|
| Edward Braunstein, Brighton Mass | 1949 | USA | Man | Left buccal mucosa | Excised | No evidence of recurrence 4 months later |
| Karl L. Klinar, Lieutenant Colonel, James C. McManis | 1969 | USA | Man | Right buccal mucosa | Excised with wide margin | No evidence of recurrence 5 months later |
| K. Ramnarayan, R.G. Nayak, Antony G. Kavalam | 1985 | India | Man | Floor of mouth | Locally excised | No evidence of recurrence 6 months later |
| Sook-Bin Woo, Jennifer E. Smith-Williams, James J. Sciubba, et al. | 1987 | India | Woman | Right buccal mucosa | Locally excised | No evidence of recurrence 9 months later |
| Toshikatsu Shibata, Naoe Kaneko, Kyoko Hokazono, et al. | 1990 | Japan | Man | Left buccal mucosa | Excised | No evidence of recurrence 12 months later |
| Tadashi Yamanishi, Shoji Ando, Tomonao Aikawa, et al. | 2007 | Japan | Man | Left buccal mucosa | Locally excised | No evidence of recurrence 7 months later |

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more common in PAM; calcification also can be seen in some cases. Hiroyuki et al. reported ameloblastin, a gene coding for a protein found in tooth enamel, was overexpressed in PAM [29].

PAM derived from Serres epithelial remnant. A lesion can be diagnosed as PAM if it is consistent with the morphological features of ameloblastoma histologically, as follows: with a connective tissue gap between the lesion and the surface mucosa; it is located in the upper of the lamina propria; and it does not extend to the periosteum. However, some PAM lesions connect with oral mucosal surface, such as in our case, which can be interpreted in two different ways, as follows. First, PAM may originate from oral mucosa because oral epithelial has the potential to differentiate into dental epithelium. Second, the formation of "collision tumor" could occur because of the dental epithelium fuse with oral mucosal in the process of teeth eruption. Studies have shown that the cytokeratin of PAM is similar to teeth embryo, but different from oral mucosa [26, 30, 31].

PAM needs to be differentiated from basal cell carcinoma, basal cell adenoma and adenocarcinoma [32, 33]. Whether basal cell carcinoma could occur in oral mucosa have different opinions, some scholars argue that basal cell carcinoma could not occur in oral mucosa, basal cell carcinoma occurring in the bearing teeth area actually is PAM. There is also another view that PAM and basal cell carcinoma are the same disease, both of them derive from the surface epithelium, but PAM lacks bone invasion potential. If the tumor lacks typical ameloblastoma histological features, distinguishing them is difficult. Studies have reported that the PAM occurred in a nonbearing teeth area, such as the maxillary tuberosity, buccal mucosa, or mouth floor, although the lesions are similar to ameloblastoma in histology, they are diagnosed as basal cell adenoma or basal cell adenocarcinoma [34, 35].

Ameloblastomas are well-known for their recurrence. Lau et al. reported that recurrence rates were 3.6% for wide resection, 30.5% for enucleation, and 16% for enucleation followed by Carnoy's solution [36]. Several factors influence the rate of recurrence, as follows: clinicopathological variant of tumor, anatomic site, safe margins during surgery, and histological

variant. The main factor of recurrence is incomplete resection. Treatment of ameloblastoma by curettage leaves a small tumor island in the bone, and this may later cause recurrence [37, 38]. The solid variety of ameloblastoma has the greatest propensity for local infiltration and recurrence [39-41].

The invasion and recurrence rate of PAM was significantly less than that of the ameloblastoma; the recurrence rate of the PAM was 19% [31]. PAM has a good prognosis, which is related to the following factors. (a) Its growth in the gingiva allows easy early detection and surgical resection. (b) The dense connective tissue of the gingiva has much more resistance capability than the thin connective tissue between the trabecular bone in the tumor infiltration. (c) The bone tissue below the gingivae can prevent tumor invasion [26]. PAM is more likely to expose to the local effects of carcinogens, but malignancy is rare. Only three malignant transformed cases have been reported. Some cases showed epithelial dysplasia. A few scholars believe that the concept of PAM might be dangerous, because clinicians may not understand the biological behavior of PAM. Therefore, unnecessary surgery is performed to extend resection [42]. At present, the treatment of PAM is conservative resection, which preserves the margin of safety without removing bone or teeth [42, 43] (**Table 1**). Follow-up after surgical treatment is considered to be important [44, 45].

After admission, we found the prominent mass originating from the buccal mucosa in a cauliflower-like manner, which involved a wide range. No relationship was found between the mass and jaws. MRI showed that the tumor was mainly located in the pterygomandibular space, unlike the PAM that was reported previously. Thus, the tumor was considered as a buccal malignant tumor initially. However, biopsy revealed that the tumor had pathological presentation of ameloblastoma. It was diagnosed as PAM, and a review of literature showed that PAM located in cheek is very rare. As we know, only six cases of extralingival PAM have been reported, five of them located in cheek, one of them located in mouth floor (**Table 2**). This is a report of a sixth case of a PAM arising in the buccal mucosa. We resected the mass conservatively, and the tumor did not recur after 20 months of treatment.

Conclusions

PAM should be considered among other differential diagnoses in the evaluation of cheek mucosa tumors. The invasion and recurrence rates of PAM are significantly less than those of ameloblastoma. Thus, conservative resection is recommended.

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Disclosure of conflict of interest

None.

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