



## Case Report

## Peripheral ameloblastoma of the pterygomandibular space: A case report

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## ABSTRACT

Peripheral ameloblastoma is a rare benign tumor of the oral and maxillofacial region with similar histologic features to central ameloblastoma. It arises from rests of odontogenic cells and epitheliums and usually found in the tooth-bearing gingival area of the jaw. We report a case of peripheral ameloblastoma in the pterygomandibular space of a 60-year-old male patient. It is important to note that the lesion occurred in the extragingival area is rarely reported. We suggest that the peripheral ameloblastoma might occur outside of the jaw and report its unique clinical features.

### 1. Introduction

Ameloblastoma is a relatively common tumor that occupies 30% of whole benign odontogenic tumors [1]. It is originated from dental lamina remnants, reduced enamel epithelium, cell rests of Malassez, basal cell layer of surface epithelium and is generally developed in the mandible and grows gradually [2]. Although it has histologically positive features, it seldom shows aggressive invasiveness. Clinically, it is categorized as regular solid, multilocular, unicystic, desmoplastic, and peripheral ameloblastoma. Although most ameloblastoma usually occurs inside of the jaw bone, peripheral ameloblastoma is developed in the periphery of the jaw bone and this occupies 1–5% of the whole ameloblastoma [3].

Clinically, Peripheral ameloblastoma is categorized by the site of development and usually occurred in the gingiva overlying the tooth bearing area around the jaw. However, it is very rare to find a peripheral ameloblastoma that is occur in the remote area of the jaw [4]. If it is developed separately from the jaw, it is difficult to diagnose peripheral ameloblastoma through clinical and radiological examinations.

We report a case of ameloblastoma occurred in the pterygomandibular space, which is separate from the tooth-bearing gingival area. A review of literature also performed for studying features of ameloblastoma.

### 2. Case report

This study was conducted in full accordance with ethical principles of the World Medical Association Declaration of Helsinki. A 60-year old

male patient who visited the department of otorhinolaryngology at other hospital with the elevated lesion in his right inner part of the throat has been diagnosed as ameloblastoma through the punch biopsy at two months ago and he visited to our oral and maxillofacial department for surgical treatment. At the first visit of patient, we observed a lesion with firm mass at the right lateral wall of oropharynx. We have identified a relatively pure and light yellowish liquid mixed with blood by the needle aspiration examination. MRI examination found a solid or cystic lesion sized 1.5 x 1.8 cm with the well-shaped boundary from the right part of the pterygomandibular space (Fig. 1). Abnormality of neck lymph node, penetration within the bone and pitting or corrosion of the cortical bone were not identified. We have performed excisional biopsy of the lesion after the 5 cm incision and dissection of lateral wall of oropharynx through the intraoral approach to the patient under general anesthesia. A solid lesion with a well-defined capsule in diameter that attached to lingual nerve bundle was identified, dissected, and excised (Fig. 2). Although a transient neurosensory disturbance of lingual nerve was observed after the surgery, the patient was recovered well within 2 months and had no other complication after the surgery.

### 3. Discussion

Peripheral ameloblastoma is a rare odontogenic tumor that is histologically similar to central ameloblastoma because the dorsal epithelium is present in the connective tissue and the tumor cells are arranged in a fence shape [5,6]. However, it is clinically less aggressive than central ameloblastoma. Central ameloblastoma are invasive and recurrence rate are 50% within 5 years after surgery, while Buchner

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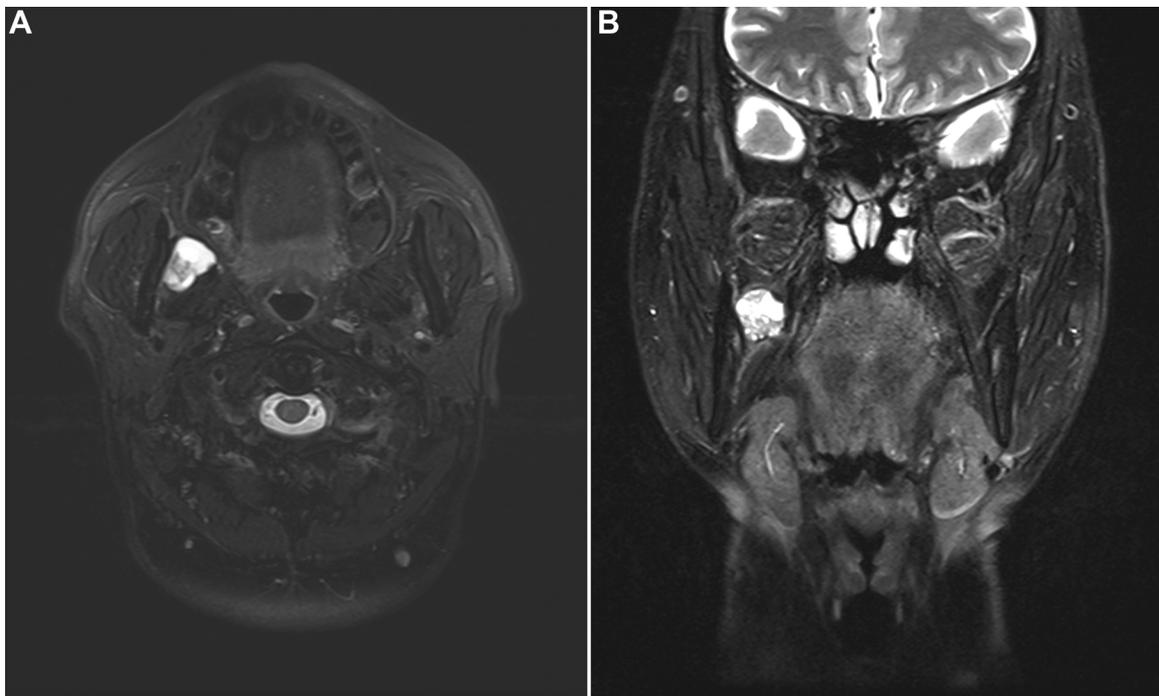
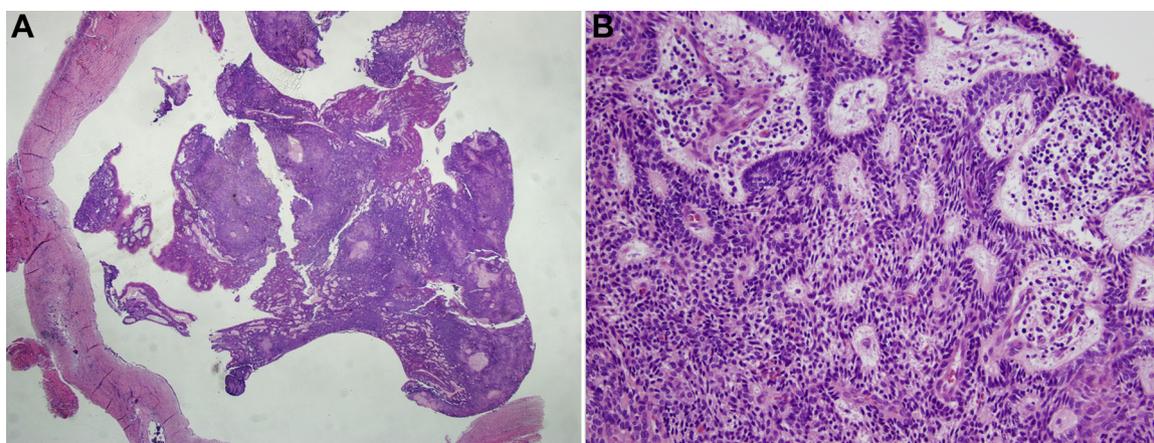


Fig. 1. T2 weighted axial (A) and coronal (B) MR image shows solid and cystic lesion in the right pterygomandibular space.



Fig. 2. A well capsulated tumor of 2 cm in diameter from the pterygomandibular space (A) is enucleated (B).



**Fig. 3.** On gross examination, the tumor was composed of mural nodules with fibrous capsule (A, H&E staining, x1.25). Microscopically, the mural nodules showed epithelial strands and cords that form anastomoses. The peripheral cells of the strands were columnar epithelium with centrally polarized nuclei. The most of inner cells were having small and ovoid nuclei with scant cytoplasm; i.e. basal-like cells. Other inner cells showed stellate reticulum-like or squamous epithelial cells (B, H&E staining, x20).

et al. reported 19% of recurrence after surgical treatment of peripheral ameloblastoma which is not common in recurrence relatively [6]. Therefore, the surgical method for peripheral ameloblastoma is usually chosen conservatively though the treatment is different by the histological features [7,8]. The lower recurrence rate of peripheral ameloblastoma suggests that thick cortical bone play a role as a barrier, and it is argued that recurrent lesion is the result of insufficient resection of the lesion, not of aggression of the lesion. However, the long-term follow-up is necessary because malignant differentiation is rarely reported [3].

Peripheral ameloblastoma is common in males (1.9:1) and developed after middle age (mean 52.1 years old), which supports the view that they are genuine neoplasms rather than embryological hamartoma [3]. The lesion occurs mostly in the mandible (70.9%) than in the maxilla (29.1%), while mostly in the maxillary tuberosity (11.1%) if it occurs in the maxilla. There is controversy surrounding the location of the lesion in relation to the diagnosis of peripheral ameloblastoma. However, case distant from the tooth-bearing area is rare [4]. The origin of the peripheral ameloblastoma is largely divided into two theories, one originating from the extra-osseous dental lamina residue and the other originating from pluripotent cells in the basal cell layer of the oral mucosa. On the basis of this argument, the lesion completely separated from the epithelium and the lesion connected to the epithelium are observed at the same time [5,9]. Philipsen et al. excluded all of the peripheral ameloblastoma originating of the extralingival area from their report of summary about 160 cases of peripheral ameloblastoma because they most likely represent basal cell adenoma with a histopathological resemblance to an ameloblastoma or the rare ameloblastoid variant of the squamous cell carcinoma [3]. Zhu et al. claimed that they should be included in peripheral ameloblastoma in not only the tooth-bearing area but also other oral mucosa, including the mucosa, lips, and palate [10]. They experimented with rats that were transplanted of the dental papilla and oral epithelial cells, including the dental plaque into the renal space, they confirmed the formation of teeth and dental sac keratin cysts three weeks after transplantation, which is the basis for the differentiation of oral epithelial cells into ameloblast [9,10]. Shiba et al. also confirmed that the peripheral ameloblastomas in the oral floor were histologically identical to central ameloblastoma, so that the extralingival lesion was a peripheral ameloblastoma [11]. This case was observed as an elevated soft tissue lesion with no epithelium changing. Radiologically, adjacent cortical bone erosion was not observed, so that inflammatory lesion or acute proliferative lesion could be excluded clinically. However, it might be considered that the lesion was a benign tumor of the salivary gland if

there was no histological examination at the previous hospital because it was a mass that developed in the pterygomandibular space above the retromolar trigone area, which is not the area of general ameloblastoma.

The classification of WHO tumors does not include the histological definition of peripheral ameloblastoma. Gardener et al. reported that both peripheral ameloblastoma and basal cell carcinoma have the same growth pattern and histologic features, so that both diseases are generally expressed in the same tumor [7]. Nauta et al. reported that 53 cases of peripheral ameloblastoma did not have distinctive features to distinguish between basal cell carcinoma and peripheral ameloblastoma [12]. On the other hand, Greer and Hammond et al. reported that peripheral ameloblastoma polarize the nucleus of the peripheral cell toward the central region, whereas genuine basal cell carcinoma is characterized by a clear distinction between peripheral and central reticular cells [13]. In this case, the tumor was composed of mural nodules with fibrous capsule (Fig. 3a). Microscopically, the mural nodules showed epithelial strands and cords that form anastomoses. The peripheral cells of the strands were columnar epithelium with centrally polarized nuclei. The most of inner cells were having small and ovoid nuclei with scant cytoplasm; i.e. basal-like cells. Other inner cells showed stellate reticulum-like or squamous epithelial cells. The connective tissue showed cystic changes (Fig. 3b). No necrosis or atypical mitosis were noted. These findings were consistent with ameloblastoma, mainly basaloid type with some foci of plexiform type. The fibrous capsule was also lined by ameloblastomatous epithelium. Though tumor did not locate near the gingiva in this case, but it was diagnosed as peripheral ameloblastoma because of the histologic findings.

#### Conflict of interest

None declared.

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