Letter to the Editor

To the Editor:
Ameloblastoma is the most common benign odontogenic tumor, consisting of odontogenic epithelium, which resembles enamel organ that has not undergone differentiation to the point of hard tissue formation, lying in the fibrous stroma. Ameloblastoma generally occurs in the intraosseous region of the premolar and molar regions of the mandible, but it rarely appears in the peripheral region. Peripheral ameloblastoma is also known as extraosseous ameloblastoma, soft tissue ameloblastoma, ameloblastoma of mucosal origin, or ameloblastoma of the gingiva. In peripheral ameloblastoma, almost all reported cases have originated in the soft tissues overlying the tooth-bearing area. However, peripheral ameloblastoma of the buccal mucosa origin is extremely rare. To date, there have been three reports of peripheral ameloblastoma arising in the buccal mucosa. The present report describes a case of peripheral ameloblastoma of the right buccal submucosa.

A 75-year-old woman presented to Division of Oral and Maxillofacial Surgery at Nagasaki University Graduate School of Biomedical Sciences with a slowly enlarging mass, of which she had been unaware, in the right buccal mucosa region. At the time of the first visit, her medical history was uneventful except for chronic gastritis. Physical examination revealed a 40 × 25 × 15 mm, relatively well-defined, elastic hard and painless mass in the right buccal submucosal region (Fig. 1). The patient did not know when the swelling had begun. The mucosa over the growth was intact but slightly immovable. Its upper extent was immediately to the anterior and superior of the orifice of Stensen’s duct, from which there was unimpeded clear salivary flow. The lesion did not extend to the maxillary or mandibular alveolar ridge. She had no facial paralysis, paresthesia, or anesthesia and no palpable regional lymphadenopathy.

Panoramic radiography did not demonstrate a direct relationship to the maxillary and mandibular bones. However, computed tomography revealed a 30 × 18 mm encapsulated tumor with a relatively clear margin and a soft-tissue-like low-density area in the right buccal space (Fig. 2). Clinically, a diagnosis of benign tumor was made. Enucleation was performed via an intraoral approach under general anesthesia.

![Figure 1](image1.jpg)  The mass was measured at 40 × 25 × 15 mm. It was a relatively well-defined, elastic hard and painless mass in the right buccal submucosal region (arrows).

![Figure 2](image2.jpg)  Axial computed tomography scan showing a 30 × 18 mm encapsulated tumor with a relatively clear margin and a soft-tissue-like low-density area in the right buccal space (arrows).
The extirpated specimen measured $35 \times 21 \times 15$ mm, and was a totally encapsulated, elastic hard, and whitish-yellow substantial mass (Fig. 3). The histological examination revealed that the tumor was well circumscribed by fibrous connective tissue, and no continuity to the mucosal epithelium was found. The tumor consisted of a cystic appearance including a substantial plexiform pattern extending from the cyst wall to the cystic lumen. The peripheral cells of the tumor were palisaded with their nuclei polarized away from the basal membrane, and these cells delineated a stellate reticulum-like area. These histological features were consistent with plexiform unicystic ameloblastoma. Some scattered follicular nests were observed in the stroma (Fig. 4). On the basis of clinical, radiographical and histopathological examinations, the diagnosis of peripheral ameloblastoma was established.

Peripheral ameloblastoma is a rare odontogenic tumor that has been reported to account for approximately 1–5% of all ameloblastomas. The first case of peripheral ameloblastoma of the soft tissue overlying the bone was reported by Stanley and Krough. Since then, many such cases have been reported, and referred to as mucosal, extramedullary, extraosseous and soft-tissue ameloblastomas. In the reported cases, an origin in the soft tissues overlying the tooth-bearing area was typical. Other types of peripheral ameloblastoma have arisen in the extragingival regions. Three cases of the lesions originated in the buccal mucosa and one in the floor of the mouth.

The biological behaviors of peripheral ameloblastoma, malignant transformation and proliferative potential in particular, are very important clinically. According to Baden et al., the frequency of malignant transformation of peripheral ameloblastoma is 6% (3/50 cases), relatively more frequent than that of intraosseous ameloblastoma (0.5–4%). However, the documentation of additional cases of peripheral ameloblastoma in the literature since their report, without a proportional increase in reported malignancies, has shown a decrease in frequency to 1.88% (3/160 cases). Although peripheral ameloblastoma is comparatively less aggressive than its intraosseous counterpart, cases of malignant transformation, recurrence as severe dysplasia and metastasis to the supraclavicular lymph nodes have been reported.

The origin of peripheral ameloblastoma has been considered to be the rests of the dental lamina, or the basal cells of the surface epithelium. However, it is difficult to explain the origin of peripheral ameloblastoma as the buccal submucosa on the basis of dental lamina rest proliferation, because dental lamina rests occur in such a location extremely rarely. Champion et al. noted that oral mucosa has the potential to differentiate into ameloblastic cells, and Klinar and McMain reported that buccal mucosa may have an abortive odontogenic potential. In an animal experiment, in which the oral epithelium and the dental papilla were removed from 17.5-day-old C3H mouse embryos and transplanted to the renal subcapsular space of 3-month-old syngeneic mice, the formation of teeth and odontogenic keratocysts were seen after 3 weeks. Zhu et al. explained that oral epithelium has the potential to differentiate into ameloblasts and to form teeth and odontogenic lesions, as the histogenesis of the development of peripheral ameloblastoma. However, we could not obtain the obvious evidence on the histogenesis in the present case.
REFERENCES


