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CASE REPORT

Peripheral Ameloblastoma in the Maxillary Canine Region

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Abstract
This report is of a 71-year-old woman with a peripheral ameloblastoma of a granular cell type arising in the crevicular epithelium of the maxillary canine region. The osseous hard mass was pedunculated and spherical with normal surface mucosa. The excised specimen revealed ameloblastomatous tissue exhibiting areas of continuity with the crevicular epithelium. After surgery, no evidence of recurrence was observed.

Key words: Ameloblastoma, Granular cell tumour, Maxilla

Introduction
Peripheral ameloblastoma is a relatively rare odontogenic tumour that arises directly from the surface epithelium or from the residues of the dental lamina lying outside the bone.[1] Histologically, the tumour exhibits the characteristics of intraosseous ameloblastoma.[1,2] However, peripheral ameloblastoma is distinguished by a less aggressive behaviour, which causes mild saucerisation of the adjacent bone without infiltration into the bone marrow.[2] Moreover, whether or not peripheral ameloblastoma is a true neoplasm or hamartomatous lesion remains controversial.[3,4] This report is of a patient with peripheral ameloblastoma in the maxillary canine region.

Case Report
A 71-year-old woman was referred to the Division of Oral and Maxillofacial Surgery, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan, in March 1991 because of a swelling of the gingiva in the right maxillary canine region. The patient noticed that the swelling had been slowly increasing in size during the past year. The patient's medical and family histories were unremarkable.

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Clinical examination revealed an elastic hard mass measuring 19 x 17 x 9 mm in the buccal gingiva of the right maxillary canine region. The mass was pedunculated and spherical, and the surface mucosa was normal (Figure 1). Dental X-ray showed a well-circumscribed multilocular radiolucency in the alveolar ridge distal to the right maxillary canine (Figure 2). The clinical diagnosis was epulis or pyogenic granuloma.

The tumour was excised en bloc together with the maxillary canine and alveolar bone at the tumour base, under local anaesthesia. In addition, a layer of exposed bone surface was shaved with a round bur. Clinically, the excised tumour was 19 x 17 x 12 mm

Figure 1. Clinical appearance of the mass in the maxillary canine region.

Figure 2. X-ray showing a well-circumscribed multilocular radiolucency.

in size and had a bone-like hardness; the surface was smooth, with a colour similar to that of the oral mucosa, and the basal surface was covered by a connective tissue–like capsule. No recurrence was detected during the 15 years since surgery.

Histopathological Findings

The tumour consisted of relatively large circular granular cells, and the formation of large cysts was observed (Figure 3). The crevicular epithelia of the right maxillary canine were located in a cord-like configuration in the interstitium (Figure 4a), and a gradual replacement of flat epithelia with granular cell-type tumour cells was observed (Figure 4b). However, in other regions, no continuity of the oral mucosa with the tumour was observed. At the base of the tumour, loosely connected angular cells similar to the enamel organ's typical stellate reticular cells were present in the interstitium and an alignment of single-layered cylindrical cells encircling the tumour was observed (Figure 4c). Squamous metaplasia and acanthoma were partly observed. Furthermore, an

Figure 3. Bucco-lingual section through the right maxillary canine showing the formation of a large cyst (arrow). Tumour cells infiltrated the alveolar bone. (Haematoxylin and eosin stain; original magnification, x 7.)

infiltration of follicular-type tumour cells into the trabeculae of the spongy bone and the compact bone was observed. This region was also covered by a fibrous capsule (Figure 4d). On the basis of the above findings, the entity was diagnosed as peripheral ameloblastoma of a granular cell type.

Discussion

Peripheral ameloblastoma is defined as having the histological characteristics of ameloblastoma occurring in the jawbone, in that it occurs in the gingiva, separate from the jawbone, and multiplies like a tumour.[2] Peripheral ameloblastoma is distinguished from tumours occurring on the oral mucosal epithelium other than the gingiva. However, since the oral mucosal epithelium has odontogenic potential, even after the development of teeth, a suggestion has been made that tumours that occur in the buccal or labial mucosa should also be classified as peripheral ameloblastomas.[5]

Peripheral ameloblastoma is a relatively rare type of tumour and accounts for 2% to 10% of all ameloblastomas.[6] Philipsen et al, in a study of 160 patients with peripheral ameloblastoma, showed that the mean age of onset was 52.1 years and the tumour

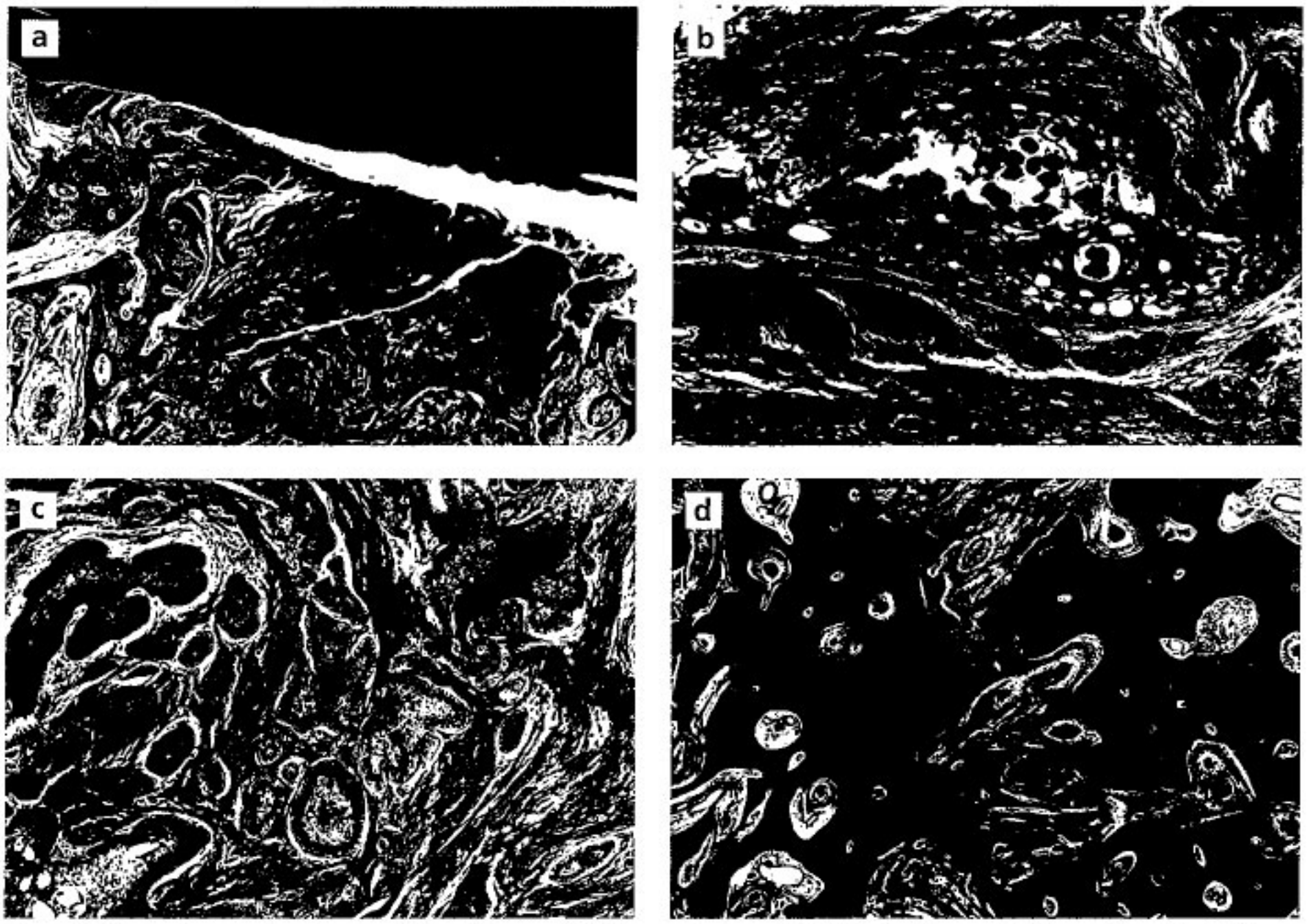

Figure 4. Photomicrographs showing different tissues. (a) The crevicular epithelia located in a cord-like configuration in the interstitium (haematoxylin and eosin stain; original magnification, x 40); (b) gradual replacement of flat epithelia with granular cell–type tumour cells (haematoxylin and eosin stain; original magnification, x 100); (c) loosely connected angular cells similar to the enamel organ's typical stellate reticular cells were present in the interstitium at the base of the tumour (haematoxylin and eosin stain; original magnification, x 100); and (d) infiltration of the follicular-type tumour cells into the trabeculae of the spongy bone and the compact bone (haematoxylin and eosin stain; original magnification, x 100).

most commonly occurred in people in the sixth decade of life.[6] The ratio of men to women was 1.9:1. These researchers also reported that peripheral ameloblastoma tended to occur among older people, in comparison with ameloblastoma.[6] Peripheral ameloblastoma occurred more frequently in the mandible (70.9%) than in the maxilla (29.1%).[6] Most of the tumours that occurred in the upper jaw were found in the posterior part of the hard palate or in the soft palate.[6] Tumours in the labial anterior part of the maxilla, such as in this patient, are rare.

According to the World Health Organization classification, the histopathological variants of peripheral ameloblastoma are mostly of the acanthoma type or the plexiform type.[2,6] There have been no reports of granular cell–type variants, as seen in this patient. The origin of peripheral ameloblastoma is likely to be the basal cells of the oral mucosal epithelium or the remnant odontogenic cells in the soft tissue. Suzuki et al traced the origin of peripheral ameloblastoma to the pluripotency of the oral mucosal epithelium, on the basis of the continuity between the tumour and the oral mucosal epithelia.[7] Philipsen et al asserted that, since tooth germ is embryologically formed on the lingual side, the remnant epithelia are more likely to persist on the lingual side, and the lingual gingiva is more likely to be the origin of peripheral ameloblastoma.[6] In this patient, as the oncocytes and crevicular epithelia on the distal side of the dextral maxillary canine were observed to be continuous and the tumour developed on the side of the lip or cheek where tooth germ is less likely to be formed embryologically, it was thought that the tumour had originated in the oral mucosal epithelium, specifically in the crevicular epithelium.

Granular cells in ameloblastoma are thought to originate from the odontogenic epithelia, and biochemically and morphologically resemble lysosome granules.[8] However, opinions differ as to whether the appearance of the granules is due to degeneration or cell activation.[8-10] It has been suggested that the appearance of the granules is due to degeneration subsequent to ageing or inflammation, on the grounds that granular cells are frequently observed in patients with a relatively slow progression of disease.[11,12] However, it has also been suggested that the appearance of the granules is due to the high activity of the tumour, on the grounds that the focus of necrosis is not observed in the tumour, although active multiplication is observed, and that phenomena subsequent to degeneration such as pyknosis, cell degeneration, and atrophy are not observed.[9,13]

In this patient, as continuity between the epithelia and the granular cell–type tumour was observed, it was thought that the neoplastic granular cells originated in the oral mucosal epithelium. It is most likely that the tumour was caused by the degeneration of granular cells, given that this patient was an elderly woman and disease progression was slow; a large cyst was formed from the granular cells, and a large amount of necrotic material was observed in the cyst. Furthermore, the tumour in this patient infiltrated into the alveolar bone, and only follicular tumour cells were present in the tumour lesion in the trabecula. This was thought to have occurred because the multiplication activity was higher in the follicular tumour cells than in granular cell–type tumour cells.

In general, ameloblastoma tends to spread through the spongy bone, without infiltrating into the Haversian canal.[1] However, in this patient, the tumour infiltrated into the Haversian canal in the compact bone of the alveolar bone, suggesting the possibility that peripheral ameloblastoma may infiltrate into the compact bone or the Haversian canal. These histological findings support the hypothesis that peripheral ameloblastoma is a true neoplasm, and not a hamartomatous lesion.

As shown, this patient had rare histopathological findings. There have been no symptoms of recurrence 15 years after surgery. However, long-term follow-up is necessary, because progression to malignancy and recurrence in the form of severe epithelial dysplasia have been reported.[14]

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