Acanthomatous Ameloblastoma with Melanin Pigmentation of the Maxilla: Report of a case

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Abstract- A case of acanthomatous ameloblastoma with melanin pigmentation occurring in the maxilla of a 27-year-old Japanese woman is reported. The microscopic examination of the biopsy and excision specimens of the tumor revealed proliferating ameloblastic tumor cells with a network of anastomosing strands. Within some epithelial strands, discrete foci of squamous metaplasia progressing to keratin pearl formation and dystrophic calcification were observed. Dark brown melanin granules were scattered in proliferating tumor nests and stromal connective tissue.

Key words: ameloblastoma, melanocytes, keratinization, calcification, ultrastructure

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Introduction

Ameloblastoma is defined by WHO as a benign, locally infiltrative neoplasm composed of proliferating odontogenic epithelium within fibrous stroma (1). Several variants have been recognized, namely, acanthomatous (2,3), basal cell, granular cell (4), unicystic (5), desmoplastic (6) and clear cell (7) types. Recently, Sia et al. (8) reported another variant of ameloblastoma presenting calcifying and keratinizing pearls. Several other investigators have discussed melanin pigmentation in odontogenic neoplasms and cysts (9-11). Takeda et al. (12) investigated 47 cases of odontogenic keratocysts and reported that melanocytes had been observed in five cases. This article presents a rare case of ameloblastoma showing both keratinizing pearls with calcification as well as melanin pigmentation.

Case Report

A 27-year-old Japanese woman was admitted to the Department of Oral Surgery, Dental Hospital of Tohoku University in December of 1990, with complaints of a painless swelling of the left cheek that had increased over the previous month. There was no relevant past or family history. Oral examination revealed marked bucco-palatal swelling in the left premolar region of the maxilla (Fig. 1). Slight pain upon pressure and parchment crepititation were present. The overlying mucosa was normal in color. A panoramic radiograph showed focal radiolucency in the apical region of the two left premolars and the first molar extending to the left

Fig. 1: Clinical photograph. A marked swelling of the left maxilla.

Fig. 2: Panoramic radiograph. Focal radiolucency (arrow) of the left maxilla.
maxillary antrum (Fig. 2). Resorption of the roots of the first and second premolars had occurred. An exploratory puncture was performed and a yellowish serous liquid was extracted. This lesion was clinically diagnosed as an odontogenic cyst. Histopathologic examination of the biopsy specimens indicated a diagnosis of a variant of ameloblastoma. In July of 1991, the lesion was excised under local anaesthesia. The excised specimen was an ill-defined granulomatous-like tissue with some dark brown tissue components. After the first operation, the lesion recurred twice, once after 7 months and again after 2 years. Re-excisions were performed and no sign of local recurrence was found after the last surgery.

**Histopathology**

**Light microscopic observations**

Microscopic examination of the biopsy and excision specimens revealed proliferating ameloblastic tumor cells in the connective tissue stroma. The tumor cells were arranged in a network of anastomosing strands (Fig. 3). The peripheral cells of the strands were columnar, cuboidal or polygonal. The central cells of the strands in some parts were stellate reticulum-like, but mostly either spindle, cuboidal or polygonal shaped.

The nuclei of the tumor cells were large and oval shaped. No cell atypia or mitosis was noted. Within the tumoral nests, discrete foci of squamous metaplasia progressing to keratin pearl formation and dystrophic calcification were observed (Fig. 4). The calcifying spots revealed prominent peaks of calcification which were identified as calcium and phosphorus by X-ray microanalysis (Fig. 4-Inset). Within the proliferating tumor nests and stromal connective tissue, dark brown granules were scattered. In addition, several dendritic cells with immunohistochemical positivity for S-100 protein and Fontana-Masson staining positivity were present (Fig. 5-a, 5-b).

![Fig. 3: Photomicrograph of the biopsy specimen. The tumor cells arranged in a network of anastomosing strands with discrete foci of keratin pearl formation. (H-E, ×40)](image)

![Fig. 4: Photomicrograph of the biopsy specimen. Dystrophic calcification in discrete foci of squamous metaplasia. (H-E, ×200). Inset: X-ray microanalysis. The calcifying spots showing the prominent peaks of calcium and phosphorus.)](image)

![Fig. 5: Melanocytes in the tumor nests. a: S-100 protein staining. b: Fontana-Masson staining.](image)

The re-excised specimens were fundamentally similar to the primitive lesion, but the squamous metaplasia and calcified keratin formation were more serious than the primitive specimen.

**Electron microscopic observations**

The tumor cells revealed a round or oval nucleus containing one or two nucleoli. The cytoplasm was comparatively sparse with numerous mitochondria and rough endoplasmic reticulum and also glycogen particles (Fig. 6). The tumor cells were densely arranged and had small intercellular spaces and numerous desmosomes. The inner cells of the strand were similar to the peripheral cells and contained some organelles, but they were loosely arranged and had somewhat wide intercellular spaces. In some areas, the tumor cells of the strand appeared flat and resembled squamous epithelium with an irregular nucleus and sparse cytoplasm. Many bundles of tonofilaments were scattered throughout the tumor cells.

The melanocyte, characteristically, was a large cell with a small round or ovoid nucleus and numerous dendritic processes. Melanin granules were observed at different stages of maturation from premelanosomes to melanosomes in the cytoplasm (Fig. 7). In some areas, fine granules and coarse aggregates of melanin pigment were also distributed in the epithelial cells.
Discussion

Among the large number of odontogenic tumors found in the oral cavity, the ameloblastoma is one of the most common (13,14). The acanthomatous type usually occurs in a follicular type but rarely does this occur in the plexiform type (15). There is extensive squamous metaplasia, sometimes with keratin formation, within the acanthomatous islands of the tumor cells (1). The present case showed squamous metaplasia and keratin pearl formation in the plexiform type with calcification and melanin pigmentation.

Melanin pigment is widely distributed in soft tissues such as the skin, nervous system, certain types of mucosa, and also the uveal tract, but is not normally present within bone (16). Melanin pigmented jaw lesions include melanotic neuroectodermal tumor of infancy (17-20). This tumor usually occurs in an infant under the age of one year, and histologic features do not resemble any type of ameloblastoma because of a lack of columnar basal cells and cells similar to stellate reticulum cells. It, therefore, is not thought to be odontogenic in origin, but rather of neural crest origin (21-23). Except for melanotic neuroectodermal tumor of infancy, few cases of melanotic ameloblastic tumor have been reported in adults (10,16,24). Melanin was found within epithelial cells in five out of the thirty ameloblastomas that Lurie (24) had examined. Subsequently, similar cases have been reported in the literature as either melanotic ameloblastic odontoma (16) or melanotic ameloblastic fibro-odontoma (10). Melanocytes have been observed in several cases of calcifying odontogenic cysts (25), odontogenic keratocysts (12) and other odontogenic lesions (11). Eda et al. (10) reported a melanotic ameloblastic fibro-odontoma in a 22-year-old woman and suggested that odontogenic epithelium was able to produce melanin pigment. Melanocytes could be
embedded within the bone by melanoblasts which have failed to complete their migration from the neural crest to the periphery (26). Certainly, since the presence of melanocytes in the oral mucosa is not uncommon (27) and since the dental lamina originates from the primitive oral lining, the occasional presence of melanocytes in odontogenic lesions can be expected. Furthermore, some investigators have suggested that the role played by racial pigmentation must be considered in pigmented odontogenic lesions (25). Lawson et al. (28) found melanocytes within the dental lamina in all six Negro and three of eleven Caucasian fetuses and in two instances within the tooth bud itself in the outer epithelial layer. They concluded that the regular occurrence of melanocytes within the developing tooth might also serve to explain the formation of certain pigmented odontogenic lesions.

Electron microscopic observations showed that the tumor cells of ameloblastoma were either central star-shaped and peripheral cuboidal or columnar in the follicular type (15). However, these two cell types were not always found in all cases of the plexiform type in which cells resembled squamous epithelium (15, 29). In the present case there was no significant difference between the peripheral and inner cells. In some areas, the tumor cells resembled squamous epithelium but the differentiation to squamous epithelium was less evident from standpoints of cellular attachment and/or keratinization than that seen in normal surface epithelium.

The distinct tendency of ameloblastoma towards local invasiveness and recurrence following excision, as in this case, has generally been considered common (30, 31). In general, histologic characteristics of the recurrent ameloblastoma were similar to the primitive lesion, but differentiation of tumor cells was usually less than that in the primitive tumor cell. In the present case, the histologic features of the re-excised lesion were similar to the first biopsy, but squamous metaplasia and calcified keratin formation were more serious than in the first specimen. As ameloblastomas are known to recur after many years of apparent cure, long-term follow-up is indispensable.

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References
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