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CASE REPORT

Ameloblastic carcinoma containing melanocyte and melanin pigment in the mandible: a case report and review of the literature

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A case of ameloblastic carcinoma containing melanocyte and melanin in a 66-year-old male with swelling and an ulcerating firm mass in the left submandibular region is presented. The diagnosis was confirmed by biopsy. The current histopathological diagnosis and management are discussed.
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Keywords: ameloblastic carcinoma; ameloblastoma; melanin pigment

A 66-year-old Caucasian male had become aware of a slow-growing lesion in his left mandibular region almost 1 year before but it had increased in size more rapidly over the subsequent 6 months. He complained of swelling and mild pain in the right mandibular region. The physical examination showed an ulcerating mass, 7×2 cm in diameter, with poorly demarcated swelling and some areas of nodular growth. The covering oral mucosa was normal in colour. He denied any history of trauma and his past medical history included surgical excision of the forehead skin with the diagnosis of basal cell carcinoma at another centre at 57 years of age and it revealed him to be a smoker. Clinical and radiological examinations of the neck region showed no lymphadenopathy.

Magnetic resonance imaging (MRI) showed a mass lesion in the left submandibular region invading the left submandibular gland with ill-defined margins (Fig. 1). The patient underwent surgical resection of the lesion. Macroscopically, it consisted of a $7.5 \times 7 \times 6$ cm mass with numerous nodules.

The patient remained under close follow up, and recurrent lesions were noted 8 and 12 months after the initial surgery in the anterior mandible and at the level of the left clavicle, respectively. The histological examination revealed a pattern the same as that of the former lesion. The patient experienced no recurrence in the 18 months following the last operation. The radiological examination revealed no metastatic lesions in the lungs.

The histopathological examination of the resected specimen revealed that the lesion had an infiltrative pattern of islands of neoplastic odontogenic epithelium within a mature fibrous stroma. There was no evidence of continuity between the tumour and the epidermis and derivatives of the skin (Fig. 2). The epithelium was rimmed by a single layer of cuboidal-to-columnar cells with palisaded nuclei, which were polarized away from the basement membrane in some areas. The cells within the epithelial islands occasionally exhibited a stellate, reticulum-like arrangement (Fig. 3). Individual epithelial cells often showed changes including increased nuclear hyperchromatism, nuclear/cytoplasmic ratio, and mitoses with typical and atypical forms (Fig. 4). In addition to clear cells, the presence of squamous metaplasia, cells with eosinophilic cytoplasm and rarely granular cell change was observed as well as widespread coagulation necrosis. It was revealed that the lesion had invaded the submandibular salivary glands, the mandibular bone and perineural tissue, and lacked a fibrous capsule microscopically.

An unusual finding of interest was the presence of dendritic-shaped cells in tumour islands and brownish-black pigment in both the tumour parenchyma and stroma (Fig. 5). The immunohistochemical investigation revealed that HMB-45 was positive in both (Fig. 6) and this pigment was also distinctly positive with Masson-Fontana's silver impregnation and negative for iron. The tumour cells were weakly positive with S-100 and vimentin immunohistochemically.

Comments

Both malignant ameloblastoma and ameloblastic carcinoma are quite rare; only 44 cases of malignant

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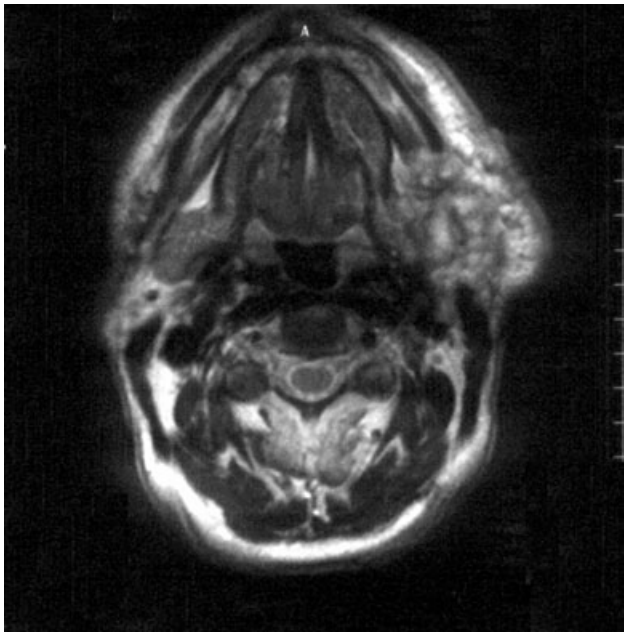

Figure 1 Magnetic resonance imaging at the time of diagnosis, a soft tissue density mass lesion showing infiltration to the skin in the left part of the mandible.

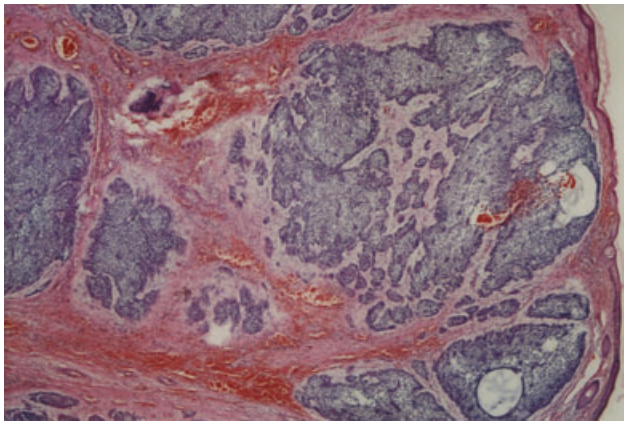

Figure 2 Ameloblastic carcinoma. The tumour consists of islands and cluster of cells and there is no continuity between the tumour and the epidermis (haematoxylin–eosin, original magnification ×40).

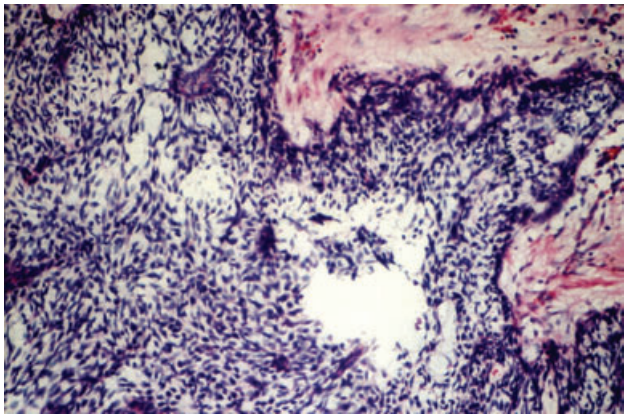

Figure 3 Focally, oedematous less-cellular spindle cell regions mimic stellate reticulum (haematoxylin–eosin, original magnification ×200).

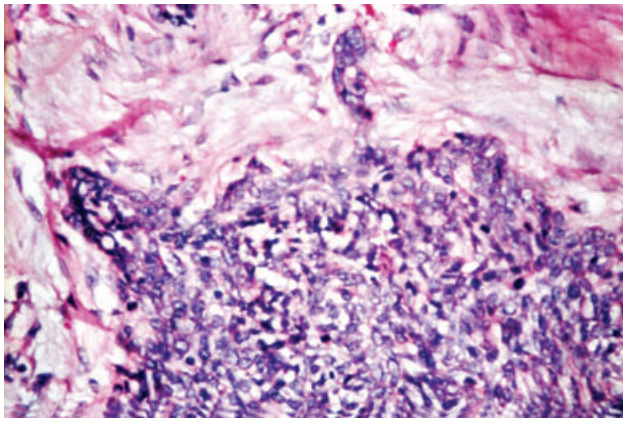

Figure 4 Ameloblastic carcinoma exhibiting high cellularity and frequent mitosis (haematoxylin–eosin, original magnification ×400).

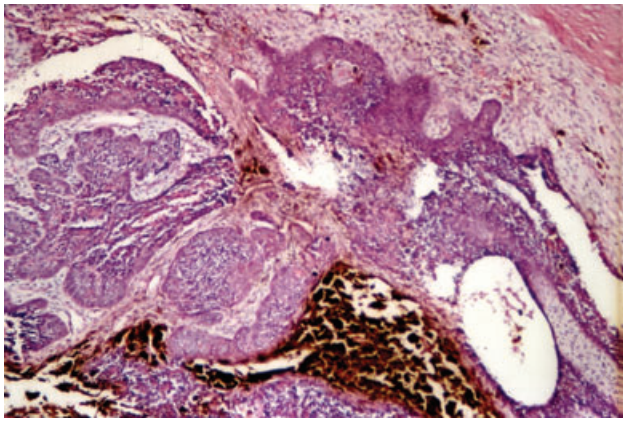

Figure 5 Many dendiritic-shaped cells, containing granular and brownish-black pigment in both the tumour parenchyma and stroma (haematoxylin–eosin, original magnification ×100).

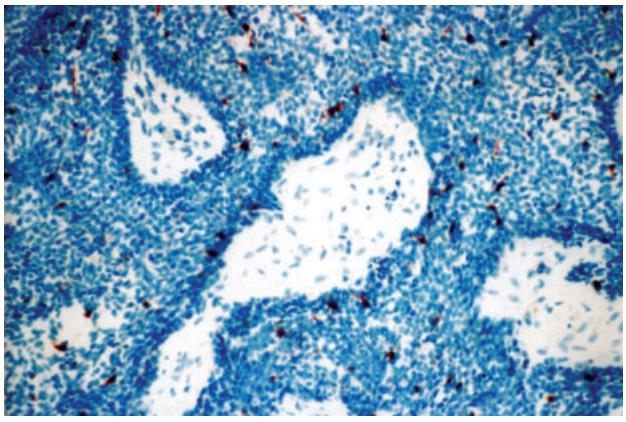

Figure 6 HMB-45 staining confirmed that these pigmented dendritic cells were indeed melanocytes (streptavidin peroxidase, AEC chromogen, original magnification ×100).

ameloblastoma and 29 cases of ameloblastic carcinoma have been reported (1). The term ameloblastic carcinoma is used to refer to any ameloblastoma in which there is histological evidence of malignancy in the

primary tumour or the recurrent tumour, regardless of whether it has metastasized. In this case, the capability of these lesions to metastasize should be anticipated (2). Ameloblastic carcinomas affect males slightly more than females, exhibiting a 5:1 mandibular predilection. The mean age at initial surgery is 33.5 years. Radiographically, the lesions are less defined; and perforation of the cortical bone with extension into surrounding soft tissue is not rare (1).

Melanocytes and melanin pigment are widely distributed in the skin, the nervous system, certain types of mucosa, the uveal tract, etc., but are not normally found within the bone in mammals. Pathologically, there are very few descriptions of intraosseous melanin-pigmented lesions other than metastases of malignant melanomas; interestingly, all reported examples have occurred within the jawbone (3). These include melanotic neuroectodermal tumour of infancy, calcifying odontogenic cyst, lateral periodontal cyst, ameloblastic fibrodentinoma (3), odontoma, odonto-ameloblastoma, odontogenic keratocyst, ameloblastic fibro-odontoma and adenomatoid odontogenic tumour (4). The origin of the melanocytes in the odontogenic lesions is speculative. Lawson et al. found melanocytes in all six Negro foetuses and in three of 11 Caucasian ones within the dental lamina of the maxilla and mandible and outer enamel epithelium. The regular appearance of melanocytes within the developing tooth may also serve to explain the formation of certain pigmented odontogenic lesions (5). Takeda and Uyeno showed that melanocytes exist in mesenchymal tissue around the dental anlage in dog foetuses and that melanocytes appear neither in the oral epithelium nor in the epithelial element of the dental anlage at that foetal stage (6). Some have thought that melanocytes in the odontogenic lesions become activated and melanin pigment is produced under certain conditions, and some have proposed that there is a relationship between melanocytes in odontogenic lesions and race (3). This report describes a very rare case of ameloblastic carcinoma exhibiting confirmed histopathological evidence of focal melanin pigment and melanocytic cell differentiation in both the nests and fibrous stroma of the tumour.

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