

Case report

Pigmented adenomatoid odontogenic tumour

Report of an undescribed case and review of the literature of pigmented intraosseous odontogenic lesions

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Summary. A pigmented adenomatoid odontogenic tumour occurring in the mandible of a 12-year-old Japanese girl is reported. In addition to the characteristic histopathologic features, fine granules and coarse aggregates of melanin pigment were widely distributed in the spindle-shaped epithelial cells between duct-like structures, and dendritic cells containing melanin pigment were also distributed in tumour epithelium. The possible histogenesis of melanin pigment in the epithelial cells of the odontogenic lesions is discussed, although no firm conclusions could be drawn.

Key words: Melanin pigment – Melanocyte – Adenomatoid odontogenic tumour – Odontogenic lesion

Introduction

Melanin pigment is widely distributed in the skin, the nervous system, certain types of mucosa, and the uveal tract, but is not normally present within bone. Pathologically, there are very few descriptions of intra-osseous melanin-pigmented lesions other than metastases of malignant melanoma, and all reported examples have occurred within the jaw bones.

Melanin-pigmented jaw lesions include melanotic neuroectodermal tumour of infancy (Pindborg et al. 1981), the pigmented variant of calcifying odontogenic cyst (Lurie 1961; Gorlin et al. 1964; Duckworth and Seward 1967; Abrams and Howell 1968; Chandi and Simon 1970; Soames 1982) and odontogenic keratocysts (Brown 1971; Brannon 1977). Racial pigmentation is thought to be one important factor in the occurrence of such

melanin-pigmented jaw lesions, with the exception of melanotic neuro-ectodermal tumour of infancy.

The author has studied the occurrence of melanin-pigmented lesions in jaw bones from Japanese patients; melanocytes in the epithelial lining of odontogenic keratocysts (1985a), the pigmented variant of calcifying odontogenic cyst (1985b, 1989a), melanin-pigmented odontogenic epithelia in complex odontomas (1987, 1989b), melanocytes in ameloblastic fibro-odontoma (1988) and odontameloblastoma (1989c). The present paper reports a case of adenomatoid odontogenic tumour containing numerous melanin-pigmented epithelial cells and melanocytes in the belief that this represents a previously undescribed component within the spectrum of odontogenic tumours.

Case report

A 12-year-old Japanese girl was referred to an oral surgeon by her dentist, because of swelling of the left buccal region and a cyst-like radiolucent lesion of the mandibular left premolar area. Clinical examination revealed a rather well circumscribed swelling of the buccal region of the mandibular left premolar area, and the lower left first premolar was unerupted. The lesion was 30 mm in diameter and of bone-like hardness due to expansion of the underlying bone. The overlying mucosa was normal in color. Radiographically, there was a well circumscribed radiolucent lesion containing multiple small radiopacities between the roots of the upper left canine and second premolar. The lower left first premolar was embedded near the lesion and its crown was embedded in the lesion. A clinical diagnosis of suspected dentigerous cyst or adenomatoid odontogenic tumour was made, and surgical resection of the lesion including an embedded tooth was performed. The postoperative course was uneventful.

The surgical specimen was a 15 × 20 × 18 mm fluid-filled sac within which was contained the crown of the premolar tooth. The sac was attached to the entire circumference of the tooth at its cervix, and the root projected from the sac. When the tooth and sac were separated the sac was sectioned, a 6 × 8 × 8 mm mural outgrowth with an irregular polypoid surface was observed.

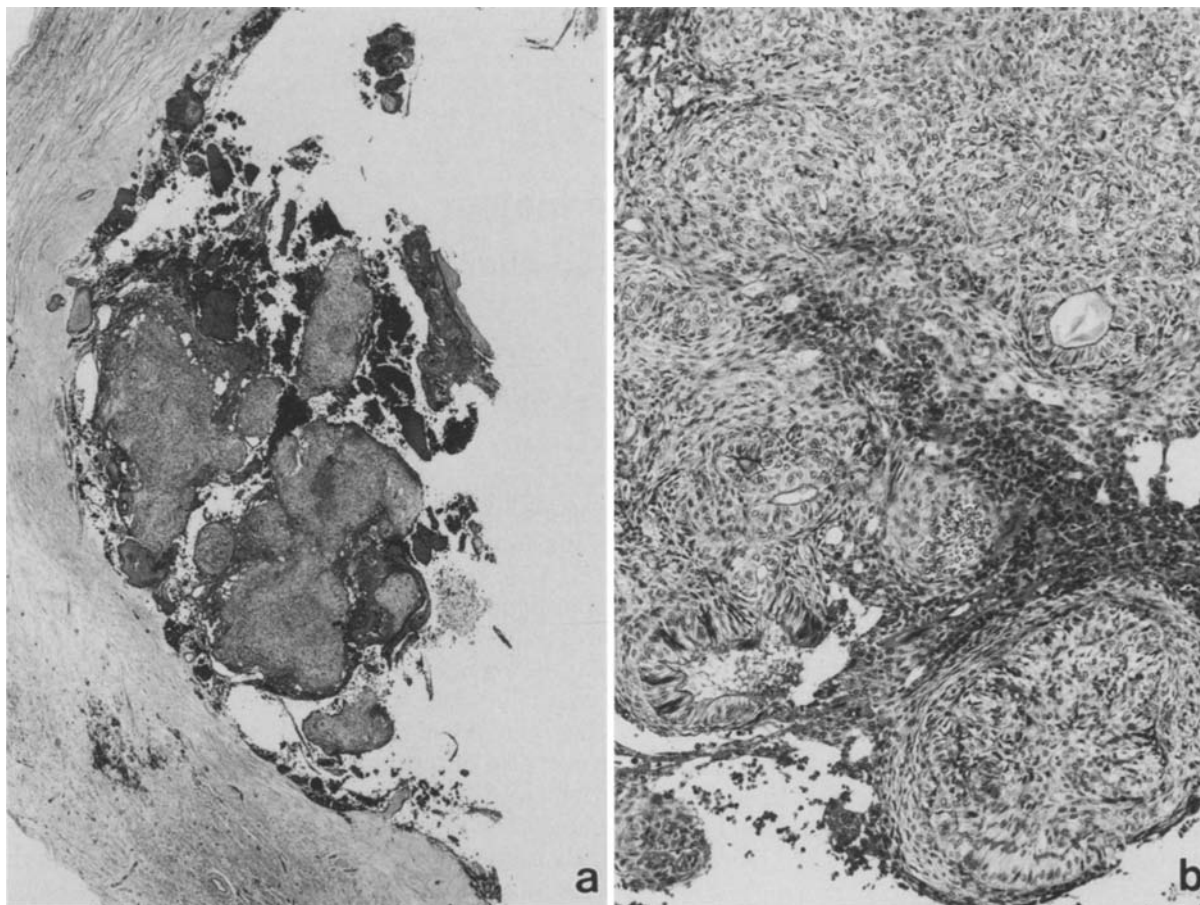


Fig. 1. Thick-walled cyst-like structure containing intraluminal epithelial proliferation. Various sized calcified bodies are scattered in and around the lesion (**a** $\times 25$). The lesion is composed of nodular epithelial masses and internodular epithelial component (**b** $\times 100$). Duct-like and whorled structures are evident in the epithelial masses

Results

Histopathological examination revealed a cyst lined by a thin layer of benign non-keratinizing stratified squamous epithelium. Arising from, and associated with the wall of, the cyst was a benign proliferation of odontogenic epithelium (Fig. 1a). The proliferating epithelium was in the form of strands, sheets and whorled masses, and various sized calcified masses were scattered in and around it. The lesion was composed of two distinct types of epithelium (Fig. 1b). The first consisted of solid nodules or islands of columnar to cuboidal epithelial cells. Duct-like spaces were located in many of the epithelial nodules and these structures were lined by similar columnar cells. Eosinophilic, amorphous material was present between the cells in some of the epithelial nodules, within the duct-like structures, and in some of these structures it separated the columnar cells from central invaginations of epithelial cells. Between the duct-like struc-

tures was the second type of epithelium, consisting of smaller spindle cells. This epithelium was also surrounded the epithelial nodules. A diagnosis of adenomatoid odontogenic tumour was made.

An unusual histological feature was that fine granules and coarse aggregates of dark brown pigment were widely distributed in the cytoplasm of spindle epithelial cells between the duct-like structures. Histochemistry indicated that the pigment was melanin, with strong positivity with Masson-Fontana's staining for melanin (Figs. 2 and 3a) and bleaching with hydrogen peroxide. Periodic acid-Schiff and stains for iron were negative. Dendritic cells containing melanin pigment in their cytoplasm were also identified (Fig. 3b).

Discussion

Melanocytes have been observed in several cases of calcifying odontogenic cyst (Lurie 1961; Gorlin et al. 1964; Duckworth and Seward 1965; Abrams

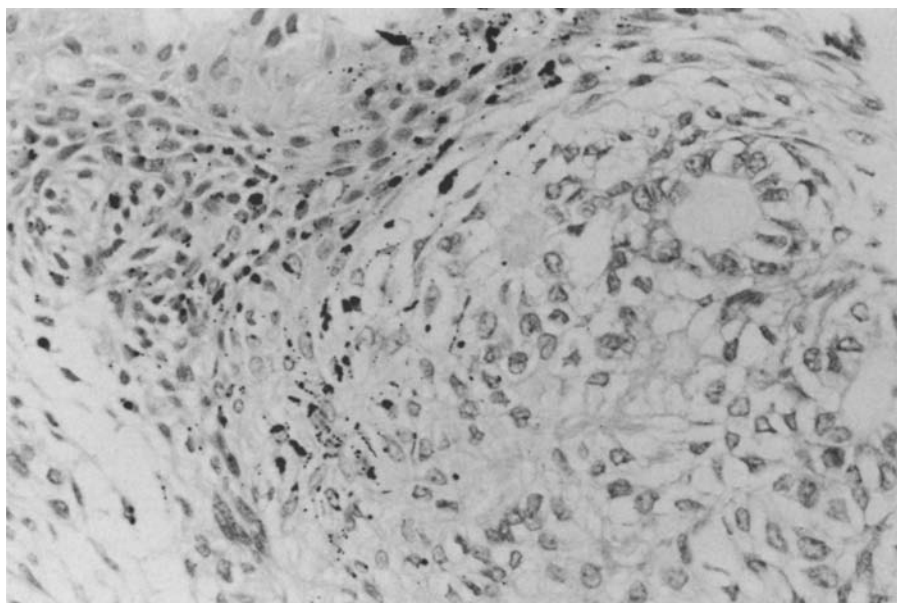


Fig. 2. Widely distributed fine granules and coarse aggregates of pigment in spindle-shaped epithelial cells between duct-like structures (Masson-Fontana's staining for melanin, $\times 200$)

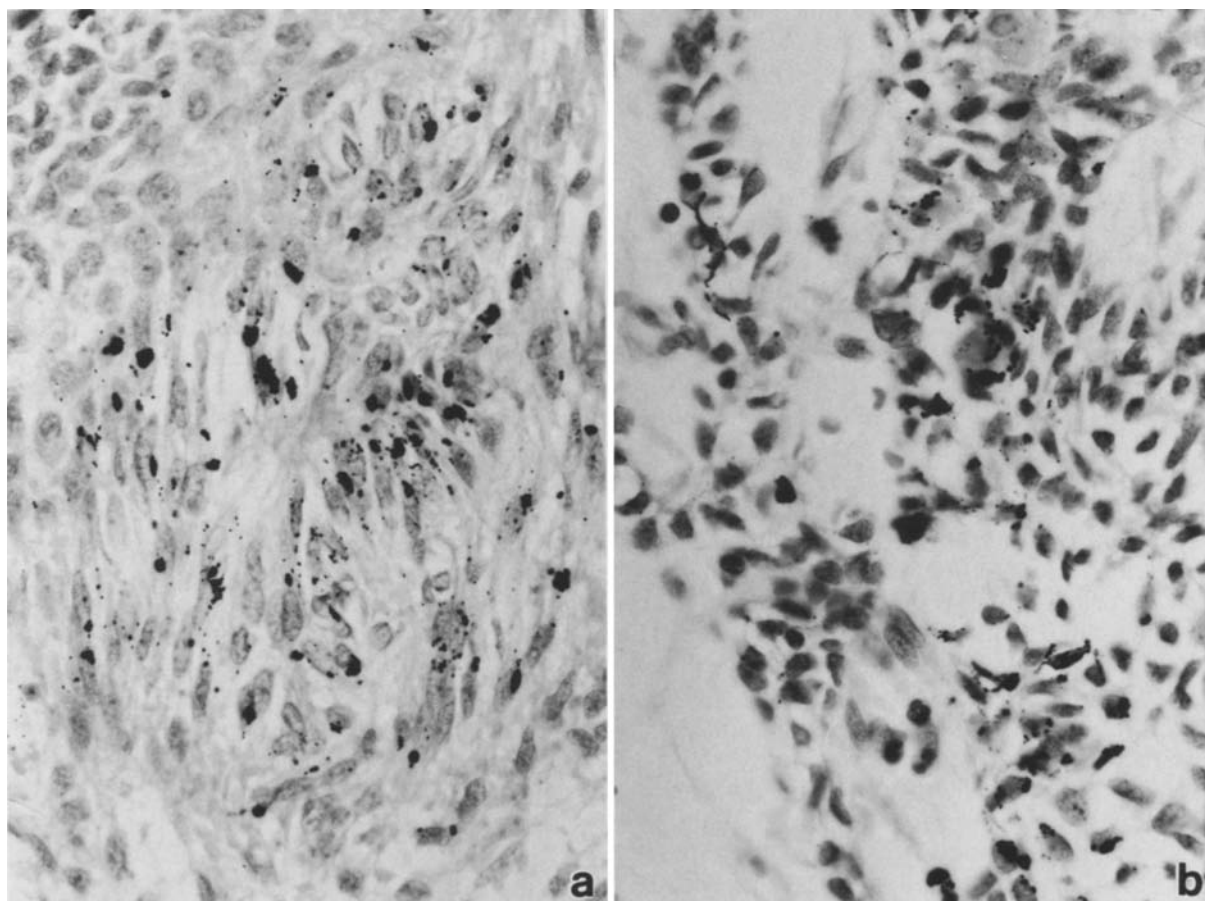


Fig. 3. High magnification of fine granules and coarse aggregates of pigment in spindle-shaped epithelial cells between duct-like structure (**a**, Masson-Fontana's staining for melanin, $\times 400$). Dendritic cells containing pigment in their cytoplasm are also identified in the tumour epithelium (**b**, Masson-Fontana's staining for melanin, $\times 400$)

Table 1. Melanin-pigmented epithelial cells and melanocytes in odontogenic lesions reported by author

Odontogenic lesions	Number of cases	Melanin pigment	Melanocytes	Hard tissue formation
Odontogenic keratocyst (1985a)	5	±	+	
Calcifying odontogenic cyst (1985a, 1989a)	3	±	+	Calcified bodies and dental hard tissues
Complex odontoma (1987, 1989b)	2	+	±	Dental hard tissues and calcified bodies
Ameloblastic fibroodontoma (1988)	1	±	+	Dental hard tissues and calcified bodies
Odontoameloblastoma (1989c)	1	±	+	Dental hard tissues
Adenomatoid odontogenic tumour (present case)	1	+	+	Calcified bodies

and Howell 1968; Chandi and Simon 1970; Pindborg et al. 1981; Soames 1982; Takeda et al. 1985b, 1989a) and odontogenic keratocyst (Brown 1971; Brannon 1977; Takeda et al. 1985a), but it seems that the presence of melanocytes in other odontogenic lesions is exceedingly rare (Takeda et al. 1987, 1988, 1989b, c). Though the histogenesis of melanocytes in calcifying odontogenic cyst and odontogenic keratocyst has been discussed previously, no conclusion can be drawn as to its specific origin in intraosseous odontogenic lesions.

Melanocytes could be embedded within the bone by melanoblasts which have failed to complete their migration from the neural crest to the periphery (Zimmermann and Becker 1959). However, when intraosseous melanocytes are associated with invading odontogenic epithelium, it is reasonable to examine the possibility that the melanocytes form part of that epithelium. Certainly, since the presence of melanocytes in the oral mucosa is not uncommon, and since the dental lamina originates from the primitive oral lining, the occasional presence of melanocytes in odontogenic lesions must be expected. Furthermore, some investigators have suggested that the role played by racial pigmentation must be considered in pigmented odontogenic lesions, since most of the patients reported previously have been coloured (Soames 1982; Takeda et al. 1985a, b). Lawson et al. (1976) have studied the distribution of melanocytes in the dental primordium of human fetuses, and found melanocytes within the dental lamina or tooth bud in 3 of 11 Caucasian and all 6 of Negro fetuses; they considered that the presence of melanocytes in the dental primordium points to a contribution of the neural crest in the formation of teeth in mammals. Such evidence of melanocytes in dental anlage may also explain the histogenesis of melanocytes in odontogenic lesions. However, one point remains unclear about the melanocytes in odontogenic lesions; most of cases of odontogenic lesions with melanocytes have been calcifying odontogenic cyst or odontogenic keratocyst, but melanocytes have not been observed in ameloblastoma, which is a

common epithelial odontogenic tumour in all races.

The adenomatoid odontogenic tumour is an uncommon histological type of odontogenic tumour which is characterized by the formation of duct-like structures by the epithelial component of the lesion. It occurs typically in young persons in the second or third decade, and females are more commonly affected than males. The tumour is generally situated in the lateral incisor, canine or premolar region, and is frequently associated with an unerupted tooth. The present lesion associated with an unerupted tooth was found in the mandibular premolar area of an 12-year-old girl, and the proliferating epithelial lining of the cystic cavity showed typical histopathological features of adenomatoid odontogenic tumour. An additional unusual feature of the present case, which has not been documented previously, is wide deposition of melanin pigment in the cytoplasm of the spindle-shaped tumour cells between duct-like structures. Furthermore, dendritic cells containing melanin pigment were also scattered in the tumour epithelium. Although no conclusion can be drawn as to the specific origin and the pathological significance of melanin pigment and melanocytes in the intraosseous odontogenic lesion, the author's cases (1985a, b, 1987, 1988, 1989a, b, c) including the present one suggest that melanin pigment and melanocytes are present not only in some cases of calcifying odontogenic cyst and odontogenic keratocyst reported previously, but also in some cases of other intraosseous odontogenic lesions (Table 1). It is interesting that all pigmented odontogenic lesions, excepting for odontogenic keratocyst, are associated with formation of dental hard tissues or prominent calcification. The pathological significance of such phenomena must await further investigations.

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