Case report of a pigmented dentigerous cyst and a review of the literature on pigmented odontogenic cysts

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(Received 30 November 1999 and accepted 2 March 2000)

Abstract: This paper reports the first case of a dentigerous cyst containing melanin-pigment and melanocytes in the lining epithelium, and the first case of a odontogenic cyst with macroscopically visible pigmentation in the cyst wall. The patient was a 29-year-old Japanese male with a cystic lesion in the left retromolar area of the mandible. Pathologic examination revealed the lesion to be a dentigerous cyst with or without mild surface keratinization, and numerous granules of melanin-pigment were distributed in the basal cells of the epithelial lining. Furthermore, dendritic melanocytes were scattered in the basal layer. Review of the literature revealed that pigmented odontogenic cysts are uncommon, and only 11 cases have been documented; eight were odontogenic keratocyst, one was a gingival cyst, one was a botryoid odontogenic cyst, and one was a lateral periodontal cyst. The possible origin of melanocytes in odontogenic lesions is discussed. (J. Oral Sci. 42, 43-46, 2000)

Key words: melanin-pigment; melanocyte; dentigerous cyst; odontogenic cyst.

Introduction

Melanocytes and melanin-pigment are widely distributed in the skin, the nervous system, certain types of mucosa, and other areas, but are not normally present within bone in mammals. 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 bone.

Melanin-pigmented jaw lesions include melanotic neuroectodermal tumor of infancy (1), calcifying odontogenic cyst (2), adenomatoid odontogenic tumor (3,4), odontogenic keratocyst (5,6), lateral periodontal cyst (7), and a few other types of odontogenic lesions. We have studied the occurrence and distribution of melanocytes and melanin-pigment in intra-osseous odontogenic lesions, and discuss their possible histogenesis and pathological significance in the jaw lesions (8-16). The present paper reports a case of dentigerous cyst with melanin-pigmented epithelial lining in the belief that this represents a not previously described component within the spectrum of odontogenic cysts. In addition, the literature of melanin-pigmented odontogenic cysts is reviewed.

Case Report

A 29-year-old Japanese male was referred to an oral surgeon by his dentist because of swelling of the left buccal region and a cyst-like radiolucent lesion of the mandibular retromolar area. Clinical examination revealed a rather well circumscribed swelling of the buccal region of the mandibular left retromolar area. The lesion was 25mm 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 cystic lesion surrounding the crown of an unerupted third molar. A clinical diagnosis of suspected dentigerous cyst was made. At surgical resection of the lesion including an unerupted tooth, the cyst wall-like tissue was torn in several pieces. The
postoperative course was uneventful.

The surgical specimens were cyst wall-like tissues within which was contained the crown of the molar. The cyst wall-like tissue was attached to the entire circumference of the molar at its cervix. The luminal surface was smooth and had a pale white color, but brown pigmentation was found in part (Fig. 1).

Histological examination revealed a cystic structure consisting of a thick layer of loose fibrous tissue and a thin layer of epithelial lining which was composed of cuboidal and flattened cells. Mild surface keratinization was seen in portions of the epithelial lining. The brown-pigmented area, noticed by macroscopic view, consisted of slightly thickened epithelium with mild surface keratinization (Fig. 2a). Further, fine granules and coarse aggregates of dark brown-pigment were distributed in the cytoplasm of the basal cells. Histochemical examination indicated that the pigment was melanin, given the strongly positive Masson-Fontana's staining for melanin (Fig. 2b), and bleaching with hydrogen peroxide and potassium permanganate solution. Dendritic cells containing melanin pigment in their cytoplasm were also identified by Masson-Fontana's staining. These dendritic cells were immunohistochemically positive for S-100 protein (1:2000 dilution, polyclonal; Dako, Santa Barbara, CA, USA) after bleaching (Fig. 2b, inset).

**Discussion**

The existence of melanin-pigment and/or melanocytes in odontogenic cysts has been noted to be exceedingly rare in odontogenic cysts, with only 11 such cases documented (Table 1). In 1964, the first case was reported by Grand and Marwah (17) whose case was a 'pigmented gingival cyst' of a 62-year-old American Black male. In a series of 104 cases of odontogenic keratocyst, Browne (5) found numerous melanocytes in the epithelial lining of multiple cysts removed from a West Indian. In another series of 278 odontogenic keratocysts, only one patient, who was an American Black, exhibited melanin-pigmentation. In a study of 47 Japanese patients with odontogenic keratocysts, we found melanocytes and/or melanin-pigment in five
patients(8). Only one case of pigmented odontogenic keratocyst was reported in a White patient (18). Altini and Shear (7,19) described a case of pigmented botryoid odontogenic cyst in a 42-year-old African Black, and a case of pigmented lateral periodontal cyst in a 38-year-old Black was described by Buchner et al. (7). To our knowledge, the present paper is the first report on the existence of melanin-pigment and melanocytes in the lining epithelium of a dentigerous cyst. We have examined histologically surgical-resected specimens of over 100 dentigerous cysts, and have identified only one case of melanin-pigmented lesion, as presented here. Warter et al. (3) reported a case of 'dentigerous cyst associated with adenomatoid odontogenic tumor' containing melanocytes and melanin-laden epithelial cells, but were not able to exclude the possibility that their case may have been an adenomatoid odontogenic tumor with a prominent cystic structure. Furthermore, the present case is the first report of a odontogenic cyst with macroscopically visible pigmentation in the cyst wall.

Our previous study of pigmented odontogenic lesions (14) suggested that melanocytes play a role in hard tissue induction in tumorous odontogenesis under certain condition, since all pigmented odontogenic tumors were composed of odontogenic epithelium and mesenchyme with induction of dental hard tissue or calcification. However, even when considering the present case, no pathologic significance of melanocytes in odontogenic cysts can be concluded.

The origin of the melanocytes in odontogenic lesions is speculative. Melanocytes form part of the oral epithelium. The occasional presence of melanocytes in odontogenic lesions can be expected, since the dental lamina originates from the primitive oral lining. In fact, Lawson and his co-workers (20) studied the facial skeletons obtained from human fetuses, and found melanocytes in all six Black fetuses and in three of 11 Caucasian ones within the dental lamina and outer enamel epithelium.

Another possible origin of melanocytes in odontogenic lesions is that the melanocytes migrate through the mesenchyme, not within the ectoderm. Our previous study (16) showed that melanocytes exist in mesenchymal tissue around the dental anlage in dog fetuses, and that melanocytes appear neither in the oral epithelium nor in the epithelial element of the dental anlage at that fetal stage. The racial factor may play an important one in pigmented odontogenic lesions, since most patients with pigmented odontogenic lesions are Asian or Black (7,8-15). However, the limited number of cases does not give any statistical value to the data.

Although no conclusion can be drawn as to the specific origin and pathologic significance of melanocytes in the odontogenic lesions, it may be of interest for future investigation.

<table>
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<tr>
<th>Authors (Year)</th>
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<th>Age</th>
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*ND*, not described.
References