A pigmented odontogenic keratocyst- Case report

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The odontogenic keratocyst is histopathologically characterized by a thin cystic corrugated parakeratinized epithelium with a well-polarized basal layer and a thin capsule of fibrous connective tissue. The presence of melanin pigment in the cystic lining is rare. This article reports a case in a Chinese female.

Key words : Key words: odontogenic keratocyst, melanin

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The odontogenic keratocyst (OKC) is a clinicopathologically distinct form of odontogenic cyst. Histopathologically, the OKC is characterized by a thin corrugated parakeratinized epithelium composed of six to ten cell layers, with a well-polarized basal layer and a thin capsule of fibrous connective tissue, mostly free from inflammatory cell infiltrates [1]. The presence of melanin pigment in the cystic lining is a rare occurrence, with only eight cases previously reported [1-4]. This report describes an additional Chinese-female case.

Report of Case

A 60-year-old Chinese female was referred to the Oral Pathology Department at Kaohsiung Medical University because of a swelling over the symphysis region. On clinical examination, a hard bone-like swelling due to expansion of the underlying man-

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dible, extending between the bilateral lower first premolars. The overlying mucosa of the swelling was normal in color. Radiographic examination revealed a cyst-like radiolucent lesion, and a provisional diagnosis of suspected cystic lesion was made. The patient was admitted and the cyst lining enucleated under naso-endotracheal anesthesia. The recovery course was uneventful.

A cystic structure lined by a stratified parakeratinized squamous epithelium was revealed from histological examination, consistent with OKC. A fine-grained brownish pigment was widely dispersed throughout the cytoplasm of the epithelial lining (Fig 1). The Masson-Fontana technique was used to determine that the pigment was melanin, however, the dendritic melanocytes could not be identified (Fig 2).

Discussion

For cases of OKC, the existence of melanin pigment and/or melanocytes is exceedingly rare, with only nine documented to date (including this study)[1-4]. Investigating 104 instances of OKC, Browne noted melanin pigments in the cystic epithelium in samples from multiple cysts which had been enucleated from a West Indian patient[1]. Brannon reviewed 278 cases of OKC, with only one African American subject revealing melanin pigmentation[2]. Further, in a study of 47 Japanese subjects with OKCs, melanin pigment was determined for

Fig 1. Cyst lining showing slight thickened epithelium with brown pigmentation almost over the whole layer of the lining (hematoxylin and eosin stain, × 100)

Fig 2. Black-stained fine granules in the epithelial cells of the representative area of the cyst lining (Masson-Fontana's stain, × 600)

five cases[3]. Additionally, there has been only one instance of a pigmented OKC noted for a white patient[4]. In a review of more than 200 cases of OKC in our department, melanin pigmentation was revealed for a single patient. To our knowledge, the present case is the first instance of pigmented OKC reported for a Chinese patient.

Apart from OKC, the presence of melanin has also been reported for other odontogenic cysts[5], as well as for some odontogenic tumors[6]. The origin of the melanin pigment in these odontogenic lesions remains incompletely understood. As the dental lamina develops from the primitive oral lining in which melanocytes comprise part of the oral epithelium, the occasional occurrence of melanocytes in the odontogenic lesions can be expected. Lawson et al observed melanocytes in the dental primordium, which appeared more frequently for black fetuses compared to white[7]. By contrast, Takeda discovered that melanocytes occur in the mesenchymal tissue but not the oral epithelium of dental tissues sampled from dog fetuses[8]. Thus, melanocyte migration through the mesenchyme, not the ectoderm, is another possible origin for melanin in odontogenic lesions. As most patients exhibiting pigmented odontogenic lesions are of Asian or African origin[3-5], it seems reasonable to propose a strong relationship for racial factors and the occurrence of pigmented odontogenic lesions, however, a larger number of cases are needed to confirm this association.

Histologically, two patterns of melanin/melanocyte distribution have been noted for pigmented OKC samples[3]. Firstly, numerous melanocytes distributed in the basal layer, but relatively little melanin determined throughout the epithelial lining. Secondly, inconspicuous melanocytes within the epithelial lining but abundant melanin pigment within the basal epithelial cells. The histological findings for our case are opposite to the first of these, with numerous melanin pigments revealed within the epithelial lining, and inconspicuous melanocytes in the basal layer.

Since all pigmented odontogenic tumors consist of an odontogenic epithelium and mesenchyme, with the induction of hard dental tissue, it seems reasonable to suggest that melanocytes may play a particular role in hard-tissue formation during tumorous odontogenesis[9]. As yet, however, the pathological characteristics for melanocytes in pigmented odontogenic cysts have not been determined

for any OKC case, including the case reported herein.

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含黑色素之齒源性角質囊腫病例報告

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齒源性角質囊腫之組織病理特徵是它有一層很薄的角質化表皮，其基底層排列整齊，並且有一薄的纖維結締組織的被囊。唯於角質化表皮中發現黑色素是罕見的現象，本文乃報告一例發生於一名中國女性的含黑色素之齒源性角質囊腫。

關鍵字：齒源性角質囊腫，黑色素
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