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CASE REPORTS

Orthokeratinized Odontogenic Cyst of the Mandible with Heterotopic Cartilage

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Abstract Cartilaginous metaplasia is a rare but well-documented phenomenon occurring in the wall of odontogenic keratocyst. The mural cartilage not associated with odontogenic keratocyst has been reported only once in a maxillary teratoid cyst of congenital origin to our knowledge. A case presented is a 38-year-old man with intraosseous keratinizing epidermoid cyst in the mandible, the wall of which contained a nodule of mature hyaline cartilage. The present lesion likely represents a previously undescribed, histologic hybrid consisting of orthokeratinized odontogenic cyst and cartilaginous heterotopia.

Keywords Cartilage · Heterotopia · Mandible · Orthokeratinized odontogenic cyst

Introduction

It has long been recognized that nasopalatine duct (incisive canal) cysts occasionally contain cartilaginous rests within their wall. They are vestigial remnants that persist in the region of palatal papilla without involution [1]. On the other hand, the heterotopic mural cartilage has been reported to occur in about 0.1–0.6% of odontogenic keratocysts [2]. This metaplastic change can be seen in association with a primary or recurrent lesion [2–7]. With a single exception of cystic teratoma in the maxilla [8], the cartilaginous wall has not been observed in any other type of jawbone cyst. This report describes a mandibular case of orthokeratinized odontogenic cyst-associated mural cartilage, which appears to be the first in English literature [9–12].

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Case Report

A 38-year-old man presented with an 8-month history of discomfort in the retromolar area of the left mandible. On admission, a fluctuant, non-tender swelling of the gingiva was seen distal to the second molar. Radiographic survey revealed a unilocular radiolucent lesion with well-corticated margins (Fig. 1). The keratinaceous nature of aspirated contents was sufficient to establish the clinical diagnosis of odontogenic keratocyst. During the surgical procedure, the lesion was found to involve the distal root of the second molar; so, the cyst was enucleated with extraction of the tooth. Grossly, the specimen consisted of a 14 × 7 × 6-mm cyst filled with keratin. Microscopic examinations showed a thick cyst wall, lined by the orthokeratinized squamous epithelium (Fig. 2a–c). The flattened basal cell layer lacked the palisading and the prominent granular cell layer was apparent (Fig. 2c). On one side of the fibrous wall was an elliptical nodule of well-developed hyaline cartilage (Fig. 2a, b). There was no foreign body reaction, necrosis, inflammation or hyaline degeneration. Also noted were areas of chondroid metaplasia in the periodontal ligament on the distal side of the second molar (Fig. 2d). Even in multiple sampling, characteristic features of odontogenic keratocyst could not be found in the epithelial lining. The lesion was finally diagnosed as orthokeratinized odontogenic cyst with cartilaginous nodule. Follow-up 3 years later revealed no sign of recurrence.

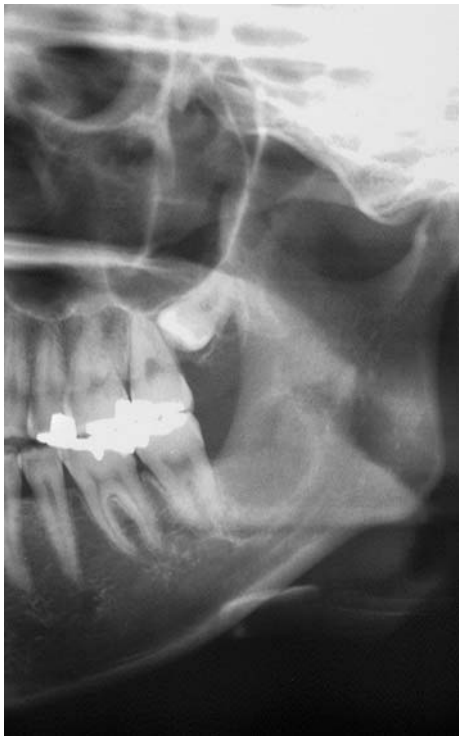

Fig. 1 Unilocular radiolucency in the mandible with corticated inferior border

Discussion

At least three histogenetic explanations for the occurrence of cartilage in the present cystic lesion can be offered: (1) the odontogenic keratocyst exhibiting mural cartilaginous metaplasia [2–7]; (2) the coincidence of orthokeratinized odontogenic cyst [9–12] or epidermoid implantation cyst [13] and ectopic cartilage in a single lesion; and (3) the intraosseous dermoid/teratoid cyst containing cartilage as an embryonic mesodermal component [8, 14–16]. We prefer to suggest our case unique "collisions" of two distinct simultaneously developing lesions, orthokeratinized odontogenic cyst and cartilaginous heterotopia for the following reasons: (1) the epithelial lining did not demonstrate characteristics of odontogenic keratocyst; (2) the cartilaginous wall of odontogenic keratocyst was interpreted as chondroid rather than true cartilage and showed multifocal distribution [2–7]; (3) the thick band of connective tissue separated a nodule of mature hyaline cartilage from the intact lining epithelium; (4) the patient had a negative past history of facial trauma or jawbone surgery with the osteocartilaginous grafting [13]; and (5) the cyst wall contained neither skin appendages nor respiratory/gastrointestinal epithelium [8, 14–16].

Finally, the cartilaginous wall of jawbone cysts is of academic interest only. This change has customarily been explained by mural cartilaginous metaplasia in the odontogenic keratocyst [2–7]. In 3.6–6% of odontogenic

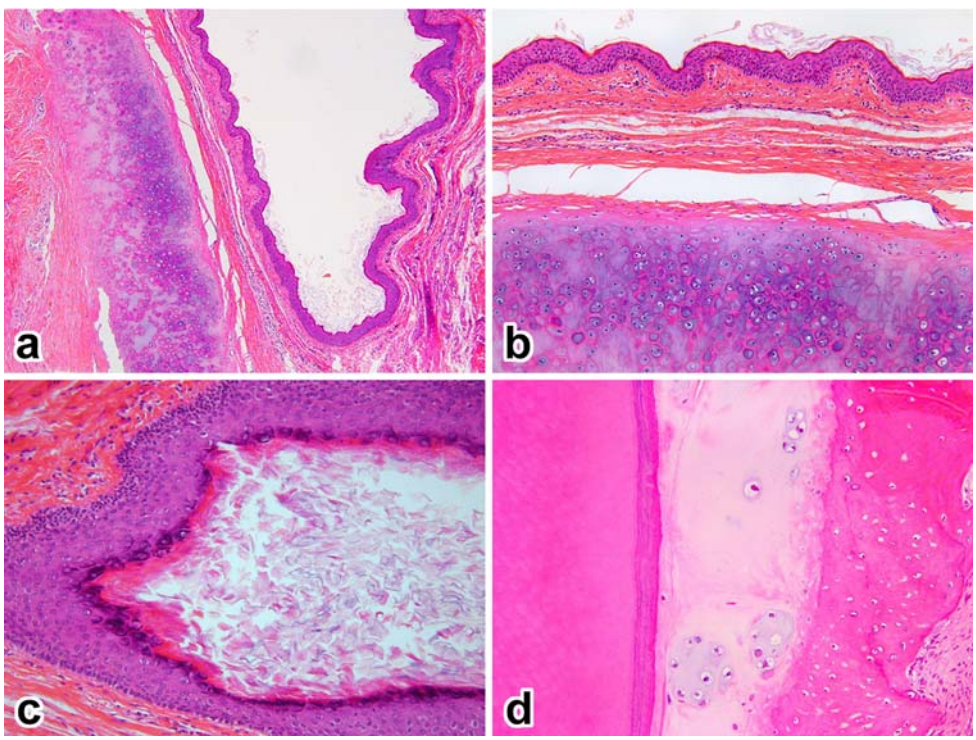

Fig. 2 **a** Orthokeratinized odontogenic cyst and cartilaginous nodule, **b** mature hyaline cartilage, **c** well-developed granular cell layer, **d** cartilagious metaplasia in periodontal ligament (Hematoxylin-Eosin. **a**, 40×; **b**, 100×; **c** and **d**, 200×)

keratocysts, there were areas of orthokeratinization with the granular cell layer [17–19]; however, our patient's lesion lacked the corrugated surface of parakeratin. It is thus our belief that the cartilage might be formed ectopically in the wall of orthokeratinized odontogenic cyst as a consequence of unknown stimuli [20]. The contingency of this occurring may be supported in part by the incidental microscopic finding of chondroid metaplasia in the periodontal ligament space of the second molar [21]. The lack of tumorous growth of the present cartilaginous nodule could be considered evidence in favor of heterotopia rather than its synonym, choristoma.

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