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Case Report

A Case of Orthokeratinized Odontogenic Cyst Suspected to be a Radicular Cyst

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Abstract

This report describes a case of orthokeratinized odontogenic cyst arising in the mandibular molar region of a 39-year-old man. Under the initial clinical diagnosis of radicular cyst, root canal treatment was performed on the mandibular right second molar. The treatment that continued for six months did not achieve healing. Subsequently surgical intervention was selected since the tooth fracture was found, and the prognosis was judged to be poor. After atraumatic tooth extraction, the apical cystic lesion was enucleated, and the tooth was replanted. A definite diagnosis of orthokeratinized odontogenic cyst was made by histopathological examination of the biopsy specimen. The radiograph taken seven months after the operation showed an improvement in the radiolucent lesion. No clinical signs of tooth mobility, pain, and swelling were present. The tooth was then successfully retained with the final restoration. Careful follow-up is needed in order to detect any signs of recurrence.

Key words: Orthokeratinized odontogenic cyst—Radicular cyst—Intentional replantation

A part of this study was presented at the 127th annual meeting of the Japanese Society of Conservative Dentistry (Nov. 8, 2007).
Introduction

Primary diagnosis of apical periodontitis that requires root canal treatment is based on radiographic examination in addition to assessment of clinical signs and symptoms. Although the most prevalent diagnosis is radicular cyst or radicular granuloma, other odontogenic cystic and neoplastic diseases can present with the similar radiologic features.

Keratocystic odontogenic tumor (KCOT), formerly known as odontogenic keratocyst (OKC), is a benign unicystic or multicystic intraosseous neoplasm of odontogenic origin, with a characteristic lining of parakeratinised stratified squamous epithelium. Its potential for locally destructive behavior, recurrence, and tendency for multiplicity has received particular attention. However, it is important to note that cystic jaw lesions that are lined with orthokeratinizing epithelium do not form part of the spectrum of KCOT. They are designated as orthokeratinized odontogenic cysts (OOCs).

This paper reports a case of OOC occurred at the mandibular second molar, initially diagnosed as radicular cyst. The possibility of a treatment modality which consists of surgical removal of the apical lesion after tooth extraction followed by intentional replantation is discussed.

Case

A 39-year-old man visited Tokyo Dental College Suidobashi Hospital after developing swelling around distal root of lower right second molar. Panoramic and dental radiography revealed a radiolucent lesion at the distal root (Figs. 1, 2a), and no sign of root resorption was observed. The systemic history was unremarkable. Acute apical periodontitis was suspected clinically, and root canal treatment was performed. Although the treatment reduced pain, a fistula formation was observed. At the sixth month, an incomplete fracture of the distal root was detected. An increase in size was radiologically observed at the apical lesion (Fig. 2b).

Attempt was made to repair the fractured area with 4-META/MMA-TBB adhesive resin (Super-Bond C&B, Sun Medical, Moriyama, Japan). A ligature wire was also used to temporarily fix the tooth. However, the suppuration persisted in spite of the continued treatment. At this stage, extraction of the tooth was indicated, but the patient expressed a strong desire to keep the tooth. Therefore, intentional replantation was offered as an alternative choice. After atraumatic tooth extraction, the apical lesion was carefully enucleated, and the visible fracture line of the distal root was further repaired with the adhesive resin (Fig. 3a). Then retro-filling of the root was

Fig. 1 Panoramic view at the first examination
performed, and the tooth was replanted. Throughout the procedure, periodontal ligament tissue was carefully treated without mechanical damage and drying. The enucleated tissue (Fig. 3b) was fixed in 15% neutral buffered formalin solution and routine pathological examination was carried out. The lesion showed cystic structure, and the cystic space was filled with a lot of keratinized tissue. The lining epithelium had thick onion-like surface orthokeratinization, prominent granular cell layer and flattened or cuboidal basal cell layer. Epithelial-connective tissue interface was flat and devoid of rete ridges (Fig. 4). Daughter cysts and epithelial islands were not seen in thin fibrous connective tissue of the cyst wall. Finally the lesion was diagnosed as OOC.

By the seventh month post-operation, a gradual improvement in radiolucency of the lesion was observed (Fig. 5a-c). With the resolution of clinical symptoms, the tooth was

Fig. 2
a: At the first examination.
b: At month 6. A crack was found in the distal root. Note the increase in lesion size.

Fig. 3
a: Surgical intervention. The tooth was atraumatically extracted, and the apical lesion was enucleated. Then the fracture was repaired with 4-META/MMA-TBB adhesive resin, and tooth was replanted.
b: Macro finding of the enucleated apical lesion. Cyst-like structure with thin connective tissue was identified.

Fig. 4 Enucleated specimen showing orthokeratinizing epithelial lining (H-E staining)
The lining epithelium has thick onion-like surface orthokeratinization, prominent granular cell layer and flattened or cuboidal basal cell layer. Epithelial-connective tissue interface is flat and devoid of rete ridges.

Fig. 4 Enucleated specimen showing orthokeratinizing epithelial lining (H-E staining)
is now designated as KCOT with the recommendation by the World Health Organization (WHO) Working Group in 2005. KCOTs comprise approximately 11% of all cysts of the jaws. They occur most commonly in the mandible, especially in the posterior body and ramus regions. Although the prevalence of orthokeratinizing variant, OOC, is considered to be low, information regarding OOC is still limited.

KCOTs as well as OOCs are difficult to diagnose clinically due to a relative lack of specific clinical and radiographic characteristics. Other cystic and neoplastic diseases, such as lateral periodontal cyst, fissural cyst, radicular cyst and benign bone tumor can all present with the similar radiologic features. OOC is clinicopathologically different from other types of odontogenic cysts and may constitute a distinct entity.

Aggressive treatment for KCOT addresses the “neoplastic nature” and could include peripheral ostectomy, chemical curettage or en bloc resection. Therefore, it is extremely prudent to distinguish between KCOT and OOC. In case of OOC, more conservative treatment (cyst-oriented) is selected to preserve anatomical structures including teeth. In the present case, conventional root canal treatment was initially selected, since the lesion was clinically diagnosed as acute apical periodontitis. The treatment was continued but failed to heal the lesion. Therefore, presence of radicular cyst was suspected. The subsequent root fracture prompted us to extract the tooth, which made it possible to access the apical lesion for enucleation. Without histopathological examination, definite diagnosis of the case would not have been possible.

We cannot determine precisely when the crack had developed in the distal root. Resulting inflammatory reaction, together with predisposing OOC was thought to have contributed to the refractory nature of the lesion. Recent report from our group demonstrated clinical efficacy of intentional replantation for teeth with refractory apical periodontitis. As for the long-term prognosis...
of the intentional replantation of vertically fractured roots after reconstruction with dentin-bonded resin, Hayashi et al.\textsuperscript{6} reported that longevity was 88.5\% at 12 months after replantation, 69.2\% at 36 months, and 59.3\% at 60 months. In the present case, the tooth was preserved to fulfill patient’s need, although poor prognosis would be predicted.

Through the experience of treatment for the present case, we reconfirmed the importance of concise histopathological examination as well as careful clinical diagnosis of an individual case. Although a review of literature found no published data on recurrence rate of OOC, it is considered to be lower than that of KCOT. In the present case, multiple procedures were required to preserve the near hopeless tooth. Therefore, careful follow-up is especially needed in order to detect any signs of recurrence.

References