CASE REPORT



Simple bone cyst with severe root resorption: a case report



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Abstract

Background A simple bone cyst (SBC) is a non-neoplastic intraosseous cavity without an epithelial lining. It is usually asymptomatic and has little effect on the surrounding structures. SBC associated with root resorption of the involved tooth has seldom been reported.

Case presentation A 63-year-old Korean woman was referred to our hospital for a cystic lesion of the left mandibular second molar. Radiographic examination revealed a well-demarcated radiolucent lesion with severe root resorption in the involved tooth. The tooth was extracted, and surgical exploration revealed an empty cavity with only a small amount of fibrous connective tissue. Histological examination revealed external root resorption in the left mandibular second molar and a cystic structure consisting of fibrous collagenous tissue devoid of an epithelial lining.

Conclusions This case of SBC associated with root resorption offers valuable insight into the diagnostic process for radiolucent lesions accompanied by root resorption.

Keywords Simple bone cyst, Traumatic bone cyst, Root resorption, Tooth resorption

Background

According to the World Health Organization classification, a simple bone cyst (SBC) of the jaws is categorized as an intraosseous pseudocyst owing to the lack of epithelial lining [1]. It is typically filled with serous or sanguinous fluid, or may be empty [1]. SBC of the jaws was first described in 1929 [2]. The diagnostic criteria, which were established in 1946 [3], include a single lesion

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⁴Department of Oral and Maxillofacial Radiology, Daejeon Dental Hospital, Wonkwang University College of Dentistry, Daejeon, Korea devoid of epithelial lining, absence of infection, and a cystic cavity that is either empty or filled with only fluid or connective tissue [3]. It has a predilection for the mandibular body and mandibular angle during the second and third decades of life, accounting for approximately 1% of cysts in the jaws [4–6]. Various terms, such as traumatic bone cyst, extravasation cyst, or hemorrhagic bone cyst have been used to describe SBC. This diversity in nomenclature is attributed to its uncertain pathogenesis [5, 6]. Although the exact etiology remains unclear, SBCs are thought to be localized abnormalities that occur during normal bone remodeling or metabolic processes [7, 8].

SBC presents as a slow-growing, non-expansile lesion that is usually asymptomatic and typically identified during routine radiographic examinations. On radiography, SBC appears as a well-defined radiolucent lesion, with or without a sclerotic rim, and may have scalloped borders when extending between tooth roots. While these lesions



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typically do not affect the surrounding teeth, there have been rare reports of tooth displacement, resorption, and the loss of the lamina dura [9-11].

Herein, we report an unusual case of an SBC associated with severe root resorption of the involved tooth in the left mandible. Additionally, we discuss potential explanations for this uncommon presentation, in conjunction with a review of the relevant literature.

Case presentation

A 63-year-old woman was referred to our hospital with a cystic lesion in the left posterior mandible. The patient had been experiencing pain on chewing in the area around the lesion for the past 2 months. She had a medical history of hypertension but was otherwise healthy. Panoramic and periapical radiography showed a well-demarcated and unilocular radiolucent lesion in the left mandible with severe root resorption of the left mandibular second molar (Fig. 1-a, b). Cone-beam computed tomography revealed a well-defined, low attenuated lesion without bony expansion (Fig. 1-c, d). The lesion was separate from the inferior alveolar nerve, and displacement of the nerve canal was not observed. Intraoral examination of the left mandibular second molar revealed no evidence of dental caries or other abnormalities. The tooth responded positively to electric pulp testing, with no signs of mobility or sensitivity to percussion. These findings indicated that the lesion was not associated with periapical infection. Based on the clinical and radiographic findings, severe idiopathic root resorption



Fig. 1 (a) Panoramic radiography shows a unilocular radiolucent lesion with root resorption of the left mandibular second molar. (b) Periapical radiography reveals progression of root resorption close to pulp horn of the second molar. (c) Axial view of cone-beam computed tomography (CBCT) shows a unilocular radiolucent lesion without cortical expansion or thinning. (d) Sagittal view of CBCT shows severe root resorption with the left mandibular second molar. Severe periodontal bone loss with the left maxillary second molar is also evident

with cystic degeneration was suspected. Benign odontogenic tumor was also suspected, but the radiologic features were unusual. Surgical exploration of the cystic lesion, with extraction of the second molar and histopathologic examination were performed.

During surgical exploration, an empty space containing a small amount of fibrous tissue was discovered. The second molar was extracted, the space was curetted, and bone graft material was subsequently placed within the defect.

Histopathological examination of the extracted tooth revealed marked external root resorption with the formation of osteodentin and presence of osteoclasts on the resorbed dentin surface (Fig. 2-a, b). The pulpal tissue showed signs of mild inflammation; reparative dentin deposition was evident. The tissue specimen obtained from the cavity revealed a band of fibro-collagenous tissue without an epithelial lining (Fig. 2-c). Additionally, amorphous eosinophilic calcified material and reactive new bone formation were observed (Fig. 2-d).

The lesion healed without complications after surgery. Six months post-operatively, the missing teeth, including the left maxillary second molar, which had been extracted due to periodontitis, were restored with implants. A panoramic radiograph acquired 1 year after surgery showed normal bone regeneration within the lesion, without evidence of recurrence (Fig. 3).



Fig. 2 (a) A tooth showing severe root resorption and reparative dentin formation with mild inflammation in the pulp chamber. (b) The resorbed surface is lined with osteoclasts (arrow) and replaced with osteodentin (arrowhead). (c) The cystic wall consists of cancellous bone within fibrous connective tissue with hemorrhage. (d) Cystic components present with amorphous eosinophilic materials and cementum-like calcification



Fig. 3 Panoramic radiography showing uneventful healing in the left posterior mandible after 1-year follow-up

Conclusions

To the best of our knowledge, only seven cases of SBC of the jaw associated with root resorption have been reported [9–11]. One case involved a 52-year-old woman with an SBC associated with an impacted third molar in the right mandible, which showed significant loss of tooth structure [9]. This case was similar to ours in that it presented with severe root resorption in a tooth associated with an SBC in a middle-aged woman. However, it was unclear whether the root resorption resulted from internal or external resorption. In contrast, our case demonstrated external root resorption confirmed by histological findings, suggesting an association with the cystic lesion beneath the tooth. Suei et al. [11] reported root resorption in 5 of 31 SBC cases. Among these five cases with root resorption, four recurred after treatment. However, the extent of root resorption and detailed radiographic findings were not mentioned in that study.

The clinical manifestation of SBC in older individuals often differs from that in younger individuals [6, 12]. Older patients tend to present with atypical features, such as the loss of lamina dura of the affected tooth or more frequent multiple cysts [6, 11, 12]. These atypical SBCs are often associated with a higher recurrence rate [11, 13]. While the bony defect in this case healed uneventfully, atypical SBCs may require longer follow-up.

Studies on root resorption in jaw lesions are limited. Among the odontogenic cysts or tumors, ameloblastoma is usually the lesion associated with root resorption, occurring in approximately 81% of cases [14]. Root resorption is reported in 55% of dentigerous cysts, 36% of nasopalatine duct cysts, and 18% of radicular cysts [14]. Despite their aggressive nature, the incidence of root resorption in odontogenic keratocysts is low [14]. The mechanism of root resorption in jaw lesions is not fully understood and does not appear to be solely related to the lesion's aggressiveness. One theory is that lesions originating from the dental follicle can lead to the resorb dental hard tissues [14], while another suggests that intracystic pressure-induced ischemia causes root resorption [15]. However, SBC does not originate from the dental follicle, nor does it have significant intra-cystic pressure. Root resorption in SBC is likely to arise from a different mechanism. We also considered the possibility that idiopathic tooth resorption may progress aggressively and form a cystic space mimicking SBC.

SBCs in middle-aged patients often occur in conjunction with benign fibro-osseous lesions such as cementoosseous dysplasia (COD) [16–19]. SBCs may arise from the progression of COD [17] or from large cysts formed by coalescing microcysts in fibrous dysplasia [20, 21]. These findings indicate that fibro-osseous lesions might contribute to the occurrence of SBC. The amorphous eosinophilic calcified material in this case may represent an early sign of COD. However, typical histological features of COD were not observed in our case. Additionally, hypercementosis is more commonly seen than root resorption in COD, which is inconsistent with this case's findings. This study presents a rare case of SBC associated with severe root resorption. The atypical presentation of SBC in older women, potential for recurrence, and need for longer follow-up periods highlight the importance of proper diagnosis and management of this condition. Although the mechanism by which root resorption occurs in SBC remains unclear, this case underscores the importance of considering SBC in the differential diagnosis of jaw lesions associated with root resorption.

Abbreviations

COD Cemento-osseous dysplasia SBC Simple bone cyst

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Not applicable.

Author contributions

This study was conceived by J.H.Y. and D.E.K who also analyzed and interpreted the histopathologic data and wrote the manuscript. H.G.Y. interpreted the radiologic data and wrote the manuscript. B.C.K. recorded and analyzed the clinical information and wrote the manuscript. All authors read and approved the manuscript.

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Data availability

All data analyzed during this study are included in this published article.

Declarations

Ethics approval and consent to participate

The study was approved by the Institutional Review Board of Daejeon Dental Hospital, Wonkwang University (W2304/013 – 001).

Consent for publication

Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

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