CASE REPORT

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주의 깊은 검사를 요하는 백악질-골 이형성증 : 증례 보고

조주연

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ABSTRACT

Cemento-osseous dysplasia in need of careful examination : a case report

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Cemento-osseous dysplasia (COD) is a fibro-osseous lesion of the maxilla and mandible, originating from a periodontal ligament. It resembles a periapical lesion such as a periapical abscess or a cyst in its early stage. Usually, the affected tooth is vital and without symptoms.

A healthy 35-year-old female was referred to the endodontic department for evaluation and endodontic treatment of an amalgam restored mandibular first molar with an apical radiolucency. Since the tooth was sensitive to a cold stimulus, chronic apical periodontitis was suspicious at first sight. But after clinical and radiographic evaluations, multiple periapical COD involving vital teeth of the mandible was diagnosed. No specific treatment of COD was scheduled except for renewing the pre-existing restoration.

The present case is a confusing case since COD was associated with an asymptomatic tooth. Knowledge of the clinical features related to pulp vitality tests and cone-beam computed tomography (CBCT) helps determine the differential diagnosis between COD and a periapical lesion.

Key words : cemento-osseous dysplasia, periapical lesion, differential diagnosis, cone-beam computed tomograph

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I. Introduction

Cemento-osseous dysplasia (COD) is a fibro-osseous lesion of the maxilla and mandible, originating from a periodontal ligament, and resembles a periapical lesion such as a periapical abscess or a cyst, at its early stage. It usually affects women in their 40s or 50s and has been described as a common lesion among the fibro-osseous lesions of the jaws with a prevalence between 0.24% and 5.9%[2,3].

The latest classification of bone-related tumors and related lesions published by World Health Organization in 2017 includes ossifying fibroma, familial gigantiform cementoma, fibrous dysplasia, COD, and osteochondroma as its fibro-and chondro-osseous lesions[4]. In this classification, COD is still a controversial and debated term, on the basis that cementum and bone are indistinguishable. However, in order to regard COD as an odontogenic lesion rising in the periodontal ligament, the terminology COD was reverted back instead of osseous dysplasia, and three subtypes are described according to its clinical and radiographic findings: periapical, focal, and florid COD[5~7]. But the classification remains unclear, and still controversial[8].

COD has a radiolucent or mixed radiolucent and radiopaque lesion with calcified materials, usually associated with a vital tooth without symptoms. Therefore, COD are diagnosed accidentally by routine radiographic examinations, and often misdiagnosed as a periapical lesion, which leads to an unnecessary dental procedure such as endodontic treatment or even surgery[1,2,8].

Normally, an asymptomatic tooth with periapical radiolucency or mixed radiolucency with radiopacity is easily diagnosed as cemento-osseous dysplasia (COD). However, in this manuscript, lesions of a relatively young aged female with a symptomatic mandibular molar presenting a mixed periapical radiolucency turned out to be one of CODs after careful examinations with an aid of cone-beam computer tomography.

This article presents a rare case of COD that involved several root apices of mandibular teeth, which was confused with periapical lesions since the patient reported discomfort at the molar. Knowledge of such radiographic features is extremely important for the differential diagnosis of COD from other similar apical lesions.

II. Case report

A healthy 35-year-old female without any specific medical history was referred from the orthodontic department for endodontic treatment of an amalgam-restored mandibular first molar with an apical radiolucency. The patient had a history of orthodontic treatment 12 years ago. The patient visited for a check-up and reported occasional cold sensitivity at the left mandibular first molar. The x-ray of the first molar revealed approximately 10mm in diameter, mixed radiodensity, and oval-shaped apical radiolucency involving both roots. Therefore, based on this information, the orthodontist regarded it as a periapical lesion that needed an endodontic treat-

ment or surgical enucleation(Fig. 1).

After the patient's referral, a dental panoramic radiograph and additional dental x-ray were taken for an initial examination. Several periapical lesions were seen in the mandible; right lateral incisor, left canine, left premolar, and the left first molar (Fig. 2,3). Surprisingly, all involved teeth except the first molar were asymptomatic, and all the teeth had intact lamina dura. Clinical examinations including electric pulp test (EPT), cold test, percussion, mobility, and probing were done. Only the left first mandibular molar was slightly more sensitive to a cold stimulus only, where the old amalgam restoration and several cracks existed(Fig. 4). All the teeth exhibited a normal response to percussion, mobility, and probing and all the involved teeth and the adjacent teeth showed EPT positive, within the score of 10 to 15. There were no significant soft tissue or bony abnormalities during an oral examination. In the radiograph, apical lesions of the left premolar and the first molar presented radiopaque mass surrounded or mixed with radiolucency which distinguished them from a simple periapical abscess, while the others looked like just radiolucent apical lesions. The patient's past dental panoramic views were able to be collected, which were taken just before starting the orthodontic treatment and just after finishing it with an interval of 2 years(Fig. 5). No apical radiolucency was detected on both x-rays. Therefore, it could be assumed that the lesions might have developed during the period between the present and after the orthodontic procedure.

Fig. 1. X-ray of the left first mandibular molar showing an apical radiolucency with radiopaque mass. A similar apical lesion is seen at the apex of the adjacent premolar.

Fig. 2. Panoramic radiograph showing several radiolucent and mixed radiopaque/radiolucent lesions at the root apices of mandibular teeth. (arrows)

Fig. 3. a), b) X-rays of left mandibular teeth: radiolucent periapical lesion of the canine, mixed radiopaque lesions of the premolar and molar.

Fig. 4. a), b) Intraoral photo of left mandibular 1st molar. An old amalgam restoration with several crack lines exists.

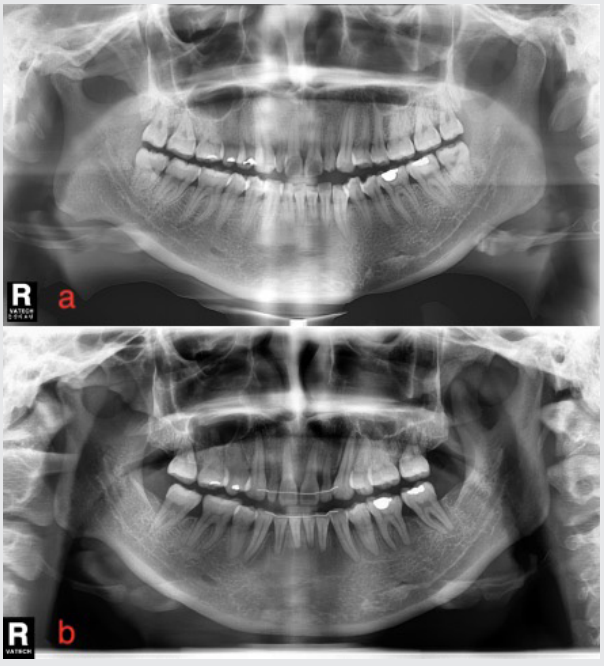

Fig. 5. a) Panoramic radiograph of the patient just before orthodontic treatment. b) Panoramic radiograph of the patient just after finishing orthodontic treatment. No definite apical lesions in both x-rays.

Then, a cone-beam computed tomography (CBCT) was collected for further diagnosis. In CBCT scans, multiple foci of bony dysplasia in various stages were found in the periapical regions of the mandibular anterior and posterior teeth. Radiopaque masses surrounded by radiolucent halos were revealed at the apex of the left premolar and the first molar with thinning of buccal cortical bone, and apical radiolucency was noticed at the apex of the right lateral mandibular incisor and the left mandibular canine. No definite bony expansion was presented(Fig. 6-9). The mixed hyper-and hypodense lesion was compatible with complex odontoma, and the hypo-dense apical lesion was mimicking a periapical endodontic lesion. The lesions were diagnosed as CODs after careful clinical and radiographic examinations studying the existing documentation and evidence. The slight sensitivity to ice pellet was mainly presented at the gap of the existing amalgam restoration, which supported the symptom did not originate from COD. The patient received information on COD and was instructed to check up every year or upon any sign or symptom, and there was no significant change at the time of follow-up in 7 months(Fig. 10).

III. Discussion

COD is a benign fibro-osseous lesion that sporadically appears in Black and Asian middle-aged females predominantly[1,2]. Familial occurrence is

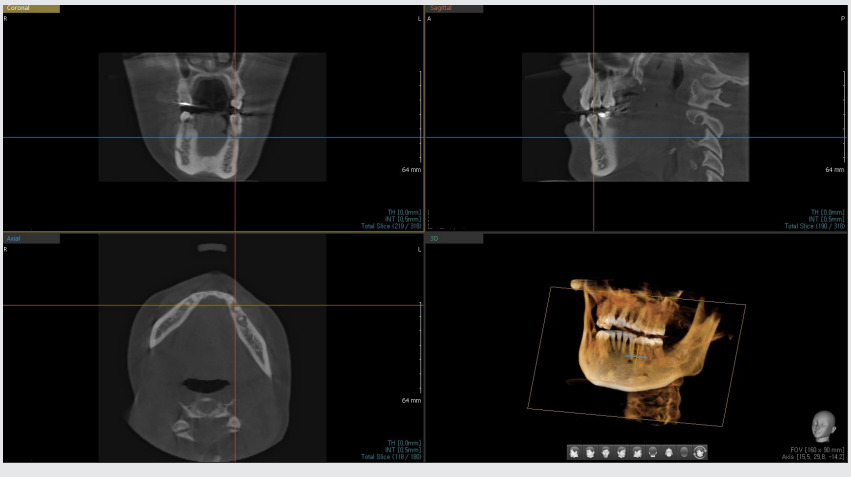

Fig. 6. Dental cone-beam computed tomography of the patient presenting cemento-osseous dysplasia lesions at the left mandibular premolar and molar.

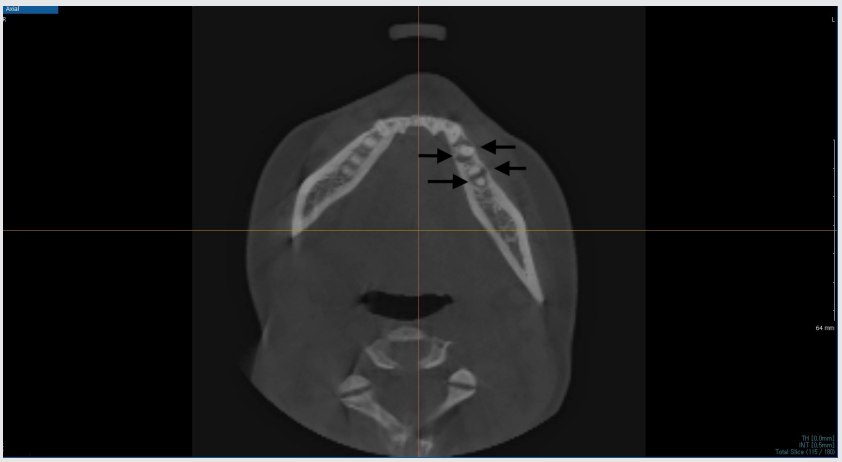

Fig. 7. Axial view of mandible. Thinning of buccal cortical bone and related radiolucent lesions are seen.

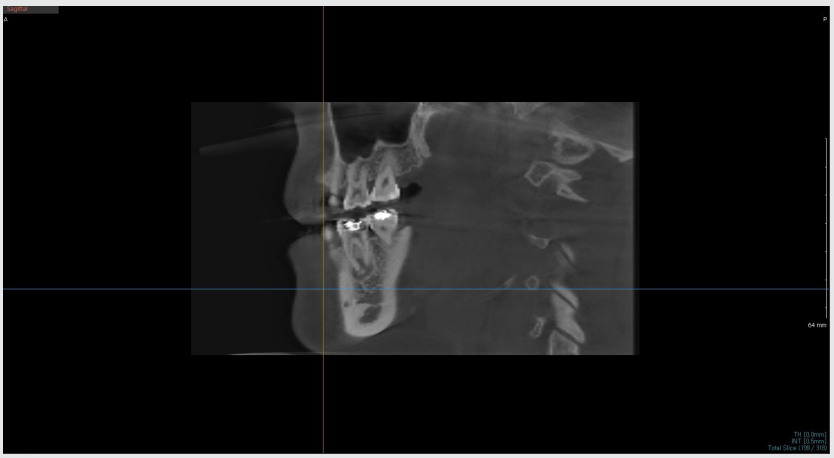

Fig. 8. Sagittal view of left mandibular 1st molar presenting radiolucent halo with radiopaque focus.

Fig. 9. 3-dimensional reconstruction of the patient's jaw assembled from dental cone beam computer tomography.

Fig. 10 Follow-up panoramic radiograph after 7 months, lesions were still unchanged.

very rare, and the hereditary cases also occur in younger ages, grouped in the florid type, and have no prevalence in female and/or black skin color. In the latest classification, three clinical forms of COD are distinguished: 1) periapical COD; located in the apex of premandibular teeth, 2) focal COD; which appears in the molar region, 3) florid COD; autosomal dominant, which occurs in any quadrant of both jaws, and usually affects 3 or 4 quadrants, has expansile potency[5]. It is reported FCOD shows a marked tendency for symmetry, with regard to the affected sextant, rather than mirror image symmetry[9]. There is a possibility that the single or initial lesion may develop into the florid form[1,10]. However, there is still controversy regarding the exact classification of COD. In the present case, the patient had COD located in both the anterior mandible and left mandibular molars, which is distinguished as a florid type of COD. The patient's familial history of COD was not investigated. And when comparing the patient's post-orthodontic x-ray with the recent x-rays, post-orthodontic remodeling and following root resorption were what was seen in the apices of #43,41,32,31 in the post-orthodontic x-ray. Because the radiolucent lesions of those teeth disappeared in the recent x-ray.

COD has three stages: 1) osteolytic stage: radiolucent lesions, 2) mixed stage: radiolucent and radiopaque lesions, and 3) osteogenic stage: radiopaque lesions[11]. This disease has clinical importance since it resembles a periapical endodontic lesion when it's in the radiolucent stage. Interestingly, the histologic and radiographic features of the COD after the third stage of periapical COD appeared to be back to normal in one report[3]. In this report of 12 years' follow-up, the radiographic appearance of periapical COD after the final stage revealed less radiopacity than in the third stage and it also showed a more normal bony trabecular pattern. However, as most COD lesions are asymptomatic, diagnosis of the disease mainly relies on a routine dental examination, and it is usually very straightforward. The margins of COD are sclerotic and well-defined. The lesion is mostly ovoid and focusing the root apex, and the maturity of the lesion is characterized by its internal radiodensity[12]. The discovery of multiple periapical radiolucencies at the vital teeth is sufficient evidence to evoke a diagnosis of COD. Biopsy of the lesions is not suggested because of the risk of postoperative infection because it is reported that these lesions lack vascular supply[1,13,14]. Since COD tends to have a scarce vascularization, it paves the way for the infection. Any uncontrolled, infected COD may lead to chronic osteomyelitis, therefore strict control is recommended for the patients with COD. For example, an ill-fitted prosthesis could lead to the resorption of alveolar bone above COD, and contributes to the exposure of the lesion to the oral cavity, which may cause infectious complications[14]. Also, the diagnosis of COD can be made based on clinical and radiographic analysis without histopathological analysis[12]. Although biopsy and following histologic analysis were not done in the present case, a main histologic feature of COD is composed of fibrous stroma and mineralized tissues which consist of woven or lamellar bone, osteoid, and cementum-like

materials[12].

CBCT is useful to facilitate diagnosis in a case when COD lesions mimic apical periodontitis or other tumors, so it prevents unnecessary dental treatment and secondary infection[7,15]. CBCT can provide information about the lesion's extent and radiodensity. Micro-opacities that are not seen in the x-ray can be detected. Moreover, COD lesions have self-limiting nature, so they usually do not invade the mandibular canal or other anatomic structures, which is one of the most definite natures of COD lesions seen in CBCT. But fibro-osseous diseases such as ossifying fibroma show a more expansive and aggressive nature. Therefore, differentiating COD from similar lesions is easier and more confident with an aid of CBCT, while diagnosis gets more difficult in case the COD lesion is associated with a root canal treated tooth, especially in case of inadequate or failed outcome[11,16].

In this case, the COD lesion of the patient was initially misdiagnosed as a periapical lesion in need of endodontic treatment or other dental procedure, which is a common mistake in the similar radiographic findings of the osteolytic phase of COD. But the unnecessary endodontic treatment was not engaged because the tooth exhibited a normal response to pulp vitality tests. However, some clinical situations, such as large restorations, pulp sclerosis, or recent trauma might not lead to normal pulpal response. Clinicians should also search for any possibilities of initiating endodontic procedures to deal with authentic pulpal disease if there are deep caries, large restorations, or previous root canal treatment[7].

COD is a disease of the jaws associated with vital teeth, and there is no need for treatment in general except in case of complications. The differential diagnosis between the early stage of this lesion and the periapical lesion is hard to make because they look similar[12]. But it is essential for avoiding unnecessary endodontic treatment due to improper diagnosis. Careful and thorough clinical and radiographic examinations including pulp vitality tests are necessary for clinicians to differentiate this disease and prevent iatrogenic dental treatment. On the other hand, the mixed or radiopaque stage of COD should be differentiated from chronic osteomyelitis, Paget's disease, cementoblastoma, hypercementosis, osteoma, and fibro-osseous lesions such as ossifying fibroma, cemento-ossifying fibroma (COF) and fibrous dysplasia[3,7,11,16,17]. Paget's disease is bilateral and affects the maxilla more than the mandible. In the active stage of Paget's disease, the blood alkaline phosphatase level is elevated. Ossifying fibroma, COF, and fibrous dysplasia have greater expanding potency, and chronic osteomyelitis is not confined to tooth-bearing areas and is normally related to the infection process. COF appears at a younger age and does not show a female predilection[3]. When the lesions get more radiopaque, the periodontal ligament may become impossible to detect in the x-ray, compromising the use of electronic apex locators, and the radiopacity could veil the apical radiolucency. Therefore, in some clinical situations, these might disturb endodontic procedure[18].

As COD is a benign, self-limiting disease, no fur-

ther investigation or treatment is needed except for simple monitoring with a long-term radiographic follow-up[17]. However, patients diagnosed with florid COD may become symptomatic and develop osteomyelitis, which may require surgical treatment[19]. Complications are rare, but secondary infection can occur after exposure of the lesion due to surgical procedures such as biopsy, and tooth extraction[9,16]. A case series study of infected COD showed a female/male ratio of 21:1, mean age of 57 years, mostly in the mandible by tooth extraction, and the florid type was the most frequently infected type[2]. Surgical resection and antibiotic treatment can be carried out for such infections, but the management is challenging being aware of the avascular nature of COD lesions. Removal of COD lesion may not help eliminate the patient's subjective symptom[9].

In conclusion, COD showing various radiological features depending on its stage is a challenging task, and differential diagnosis with other radiolucent and/or mixed radiopaque/radiolucent apical lesions is essential for prompt management. Clinicians should suspect COD when a lesion is located in the apex of a tooth, especially if a tooth is asymptomatic but is still vital.

COD might be the proper diagnosis even if the susceptive tooth is symptomatic because of other odontogenic reasons such as caries or crack like in this case. And the patients should be regularly monitored with long-term clinical and radiographic evaluation. Improving oral hygiene to prevent tooth loss is also recommended and avoiding trauma in the affected region is necessary not to expose and infect COD.

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