CASE REPORT

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Florid osseous dysplasia with chronic suppurative osteomyelitis in Asian female: an incidental finding on Panoramic and CBCT examination

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ABSTRACT

Objectives: In a previous case study, we reported an incidental finding of florid osseous dysplasia with osteomyelitis, which was identified on panoramic and CBCT imaging.

Case Report: A 53-year-old female patient came to the Radiology Department of Dental Hospital Padjadjaran Bandung with the chief complaint of recurrent pain one year ago; there was swelling from the cheek to the left mandible; the swelling reappeared one week ago with clinical pus mixed with blood in the swelling area. The patient had previously seen an oral surgeon and was referred for panoramic and CBCT radiographs. The results of the panoramic radiographs were that there was an irregular radiopaque lesion with ill-defined borders in the posterior region affecting 4 quadrants; in the sinister mandibular region, there was cortical destruction, root resorption, and the lesion appeared to extend to the canalis mandibular area. The CBCT examination showed multiple globular radiopaque lesions affecting 4 quadrants in the posterior area. There were sequestra with lytic lesions on the sinistra mandible resulting in buccal and lingual cortical bone discontinuity and buccal-lingual expansion. Expansion of the lesion area resulted in discontinuity of the canals mandibular sinistra and decreased bone density of the trabeculae up to the mandibular ramus; there was apical resorption trabecula bone density. From the Panoramic examination and CBCT results, the suspect radiodiagnosis was Florid osseous dysplasia associated with osteomyelitis.

Conclusion: An examination of panoramic and CBCT images revealed FOD that had been secondarily infected with chronic diffuse osteomyelitis.

Keywords: *Chronic osteomyelitis, florid osseous dysplasia, panoramic, CBCT examination*
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INTRODUCTION

Osseous dysplasia (OD) is an idiopathic condition that affects the periapical area of the upper and lower jaws. Metaplastic and normal bone are replaced by fibrous tissue, which indicates the condition.[1] In 2005, the World Health Organization divided osseous dysplasia into three categories: florid dysplasia, which occurs when lesions are found bilaterally in the mandible and may be seen in all four quadrants; periapical bone dysplasia, which occurs when lesions are found in the anterior region of the mandible; and focal dysplasia, which occurs when lesions are found to be limited to the posterior mandibular quadrant.[1,2] Melrose et al. introduced florid cement-osseous dysplasia (FCOD) in 1976. It is identified by large multi-quadrant masses of cementum and bone in both jaws and simple bone cavity-like lesions in the affected quadrant.[2,3] Florid osseous dysplasia (FOD) was an exuberant form of OD characterized by multiple, diffuse, lobular, or irregularly shaped radiopacities throughout the alveolar processes but not restricted to the root apices. Cortical bone expansion may be present. However, external visible facial asymmetry is usually unobserved. Middle-aged women (40–50 years old) are the most common age group for FOD cases. This condition often affects the mandible bilaterally and symmetrically, but can also affect the maxilla. In other situations, this pathological condition is asymptomatic, and the lesion is "only discovered" during routine radiographs.[2,3]

Osteomyelitis is a progressive spread of inflammation in the bone and bone marrow, more commonly affecting the mandible than the maxilla. It is more common in the mandible than the maxilla because of the dense, poorly vascularised cortical plates and the inferior alveolar neurovascular bundle blood supply. Local factors like periapical infection, pericoronitis, acute periodontal lesions, trauma, or extractions are typically the cause.[4–6] Osteomyelitis (OM) of the jaws is a disease that is often difficult to treat. To diagnose, acute and

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chronic clinical courses must be distinguished. OM may present radiographically as suppurative, sclerosing, and with periostitis (Garre´), tuberculous, and osteoradionecrosis. Osteomyelitis can be acute or chronic, depending on the progress made after the onset of symptoms. An acute process occurs up to one month after the onset of symptoms, and a chronic process occurs for longer than one month.[7,8] Disease incidence has decreased significantly following broad-spectrum antibiotics, advances in restorative dentistry, increased awareness of oral hygiene, and better dental health care. When antimicrobial agents prove ineffective, acute osteomyelitis may progress to the chronic stage, becoming a refractory condition that is difficult to treat. Hence, the correct diagnosis of osteomyelitis is decisive. Healthcare practitioners must thoroughly know the signs and symptoms for proper evaluation and effective management.[7] Keystones leading to the diagnosis are the characteristic, but not pathognomonic, radiological signs: single or multiple irregular radiolucencies, ill-defined cortical border, sequestra, osteosclerotic changes, bone enlargement, periosteal reaction (onion peel appearance or Single line). The radiological findings were described as osteolytic areas within sclerotic zones and cortical destruction, dissolution, loss of trabecular pattern, moth-eaten appearance, and shortening of tooth roots. Only periosteal bone formation and sequestra are pathologic expressions directly linked to OM.[5,8]

First-line imaging methods, such as intraoral radiographs or panoramic images, can help make a diagnosis. CBCT imaging is reliable for identifying common alterations, including soft tissue, periosteal reactions, and bone involvement. It also helps to determine the extent of the damaged bone and its relationship to neighboring anatomical structures.[4,8] In this case report, we presented a case of non-familial Florid Osseous Dysplasia associated with chronic suppurative osteomyelitis in a 53-year-old Asian female, which was an incidental finding on Panoramic and CBCT examination.

CASE REPORT

A 53-year-old female patient came to the Radiology Department of Dental Hospital Padjadjaran Bandung with the chief complaint of recurrent pain one year ago; there was swelling from the cheek to the left mandible; the swelling reappeared one week ago with clinical pus mixed with blood in the swelling area (Fig 1). The patient

Figure 1. Clinical examination shows a swelling from the cheek to the left mandible

Figure 2. Intraoral examination shows a fistula that had developed in the edentulous region and exposed bone

Figure 3. Panoramic radiograph shows multiple irregular radiopaque lesions bilaterally with ill-defined borders in the posterior mandible and also in the posterior maxilla, in the sinister mandibular region, there was cortical destruction, root resorption, and the lesion appeared to extend to the canalis mandibular area

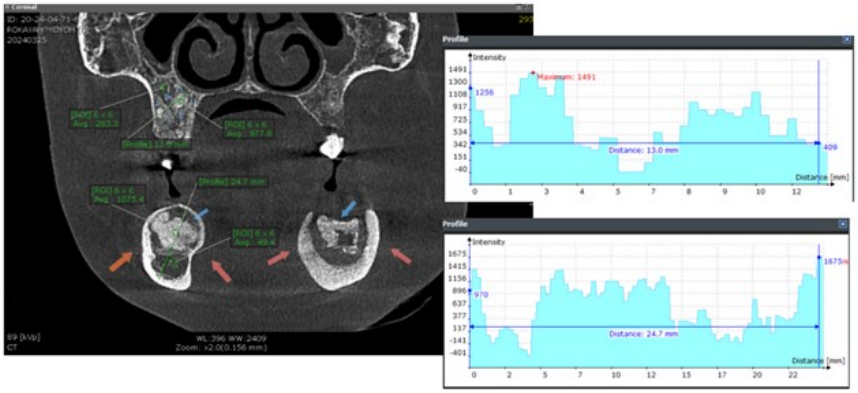


Figure 4. The coronal slice shows a mixed-density lesion with an amorphous shape, an ill-defined border in the mandible, and a radiopaque lesion in the maxilla. On profile examination, the lesion density is irregular in the maxilla and mandible. On ROI examination, the densities of the lesion cores are higher than those of the surrounding tissues.

had a history of extraction of teeth 36 and 37 two years ago. A fistula that had developed in the edentulous region and exposed bone was discovered during the clinical examination (Fig 2).

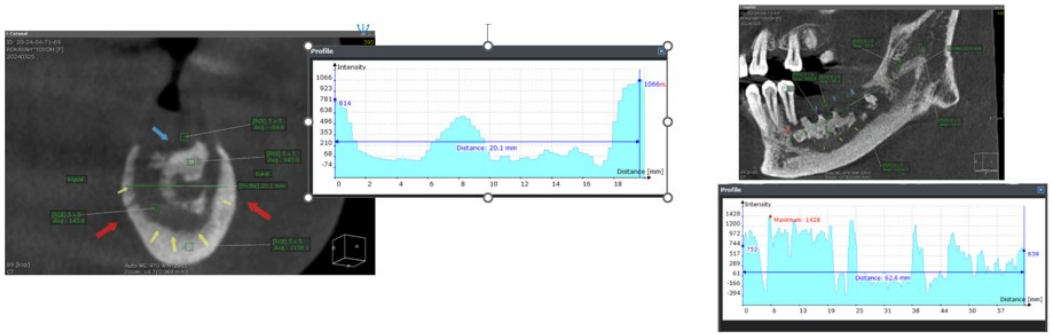
The patient had previously seen an oral surgeon and was referred for panoramic and CBCT radiographs. The results of the panoramic radiographs were that there was an irregular radiopaque lesion with ill-defined borders in the posterior region affecting four quadrants; in the sinister mandibular region, there was cortical destruction, root resorption, and the lesion appeared to extend to the canalis mandibular area (Fig 3). The CBCT examination showed multiple globular radiopaque lesions affecting four quadrants in the posterior region (Fig 4). There were sequestra with lytic lesions on the sinistra mandible, resulting in buccal and lingual cortical bone discontinuity and buccal-lingual expansion. Expansion of the lesion area resulted in discontinuity of the canals mandibular sinistra and decreased trabeculae bone density up to the mandibular ramus; there was apical resorption 35 (Fig 5, 6) . No histopathologic examination has been performed, and the patient has only received treatment in the form of antibiotics, anti-inflammatory drugs, painkillers, and mouthwash. From the panoramic examination and CBCT results, the suspect radiodiagnosis was Florid osseous dysplasia that had been secondarily infected with osteomyelitis.

DISCUSSION

Cemental-osseous dysplasia is a well-known term used to describe a group of disorders. Within this classification, periapical cemental dysplasia (PCOD) specifically affects middle-aged black women.[1,3,9–11] Research indicates that 59% of reported cases are in black individuals, 37% in Asians (including Japanese, Chinese, and Korean), and 3% in Caucasians, including Indians. According to Zegarelli et al., approximately 2-3 in 1000 people in the general population have PCOD.[9] There are 1:2 males to females. It is currently unknown what

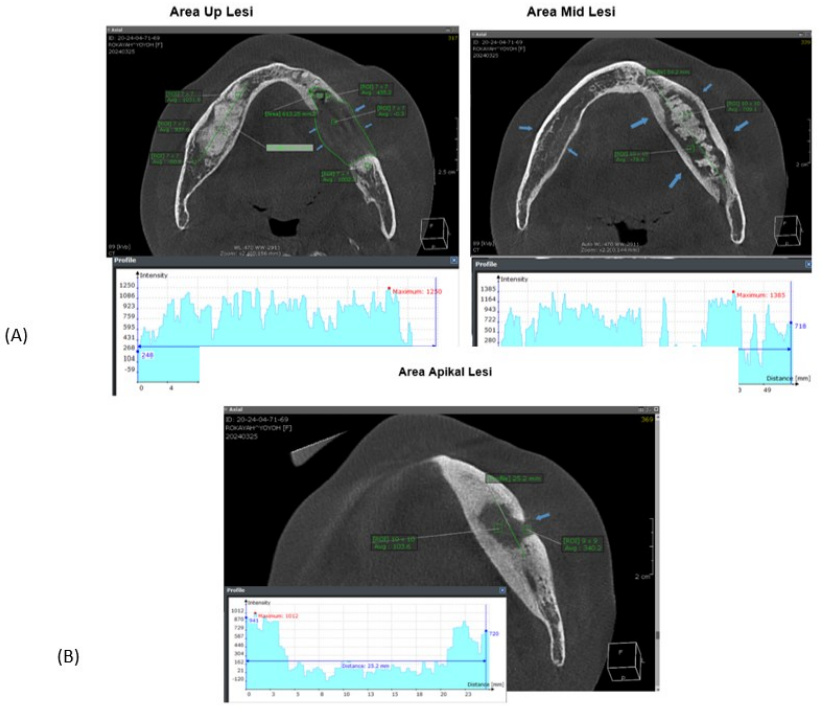
causes this illness and why the prevalence varies among racial and gender groups. According to numerous studies, the disease's histological resemblance and localized occurrence prove that it originated in the periodontal ligament. Studies have shown that it might result from the cementum staying in the socket following an extraction.[1]

FOD is a widespread form of Periapical osseous dysplasia (POD). If POD is identified in three or four quadrants or when it extends along one jaw, it is usually considered FOD. Often, FOD causes no symptoms. The teeth of the involved bone are still vital.[4,12] Florid osseous dysplasia (FOD) has synonyms florid cemental-osseous dysplasia, Gigantiform cementoma, and familial multiple cementomas. Florid osseous dysplasia (FOD) was the term first proposed by Melrose et al. In Cral[3], to describe a clustered form of OD characterized by multiple, diffuse, lobular, or irregular radiopaque lesions located along the processus alveolar, but not limited to the apices of the teeth.[1,4,12]FOD progresses without symptoms in most situations, but in severe cases, infection-related discomfort and localized swelling might result in facial asymmetry. The condition, which affects only the jaws, does not affect blood levels. FOD is usually diagnosed accidentally at routine dental radiographic examinations.[1,12,13]



(A) (B)

Figure 5. A: The lesion at the mandibular sinistra is classified as a mixed-density lesion, indicating a lower density inside the lesion than outside. The lesion appears to result in cortical bone discontinuity (blue arrow) surrounded by sclerotic trabeculae, which in turn results in lingual and buccal cortical bone expansion (red arrow). B: The sagittal slice shows multiple globular radiopaque lesions, sequestra with lytic lesions (yellow arrows), and resorption at the apex of tooth 35 (red arrow). Multiple globular radiopaque lesions with irregular lesion density were seen, resulting in discontinuity of the cortical bone and vital anatomy of the mandibular canal (blue arrow). In the 8x8 ROI measurement, there was a decrease in trabecular bone density in the ramus area of the mandibular sinistra



(A)

(B)

Figure 6. A & B: The axial slice shows a lesion on the mandibular sinistra with an area of ± 613.25 mm2, resulting in discontinuity of the buccal and lingual cortical bone (blue arrow). Multiple rounds and dense masses with irregular densities are present, causing the expansion of the buccal and lingual cortical bone in the mandible sinistra. However, there is no cortical bone expansion in the mandibular dextra. In region of interest (ROI) measurements of 10x10 and 9x9, there was a decrease in the density of the trabecular bone and discontinuity in the buccal cortical bone in the mandibula sinistra.

Radiographically, the lesion varies depending on the stage. FOD opacifies progressively as it becomes more mature. The classic appearance includes diffuse, lobular, irregular-shaped radioopacities throughout the alveolar process of the maxilla and mandible, which was seen in our case. The lesion is apical to the tooth, from the alveolar process, and usually posterior to the canine. The lesion is above the inferior alveolar canal if it occurs in the mandible. The external border is clear and has a sclerotic border that varies in width, similar to POD. The internal structure has a density ranging from equal area between radiolucent and radiopaque areas to full radiopacity.[3,4] Radiographically, these lesions become increasingly radiopaque as they mature over time, progressing through three stages as described[14]: Stage 1 – osteolytic stage: radiolucent lesions, Stage 2 – mixed stage: radiolucent and radiopaque lesions, and Stage 3 – osteogenic stage: radiopaque lesions.

The rare clinical symptoms observed include pain, swelling, and local drainage. However, these are only encountered in cases of secondary infection when the calcified masses are exposed in the oral cavity.[14] Osteomyelitis is a severe condition characterized by bone and bone marrow inflammation. It requires prompt and thorough medical attention. It typically occurs after physical trauma or chronic infection of nearby tissue. Based on the symptoms, osteomyelitis can be classified as acute, subacute, or chronic.

One key difference between acute and chronic osteomyelitis is the level of pain experienced by the patient. In chronic cases, the patient may experience mild or no discomfort. Osteomyelitis is more common in the mandible due to its thick cortical bone and limited blood supply. Osteomyelitis can occur acutely or chronically depending on the infecting organism's virulence and the patient's resistance. Clinical symptoms of acute osteomyelitis are rapid onset, pain, swelling in surrounding tissues, fever, lymphadenopathy, and leukocytosis. Symptoms of chronic osteomyelitis are generally milder than those of acute osteomyelitis. Symptoms include recurrent swelling, pain, fever, and lymphadenopathy. Clinical findings in chronic mandibular osteomyelitis include local pain, fever, swelling, purulent discharge, intraoral and skin fistula, unhealed soft tissue in the oral cavity, neuropathy in the involved area, pathologic fracture, and trismus.[5] Our patient presented to us with dull, aching pain, foul odor, and a skin fistula. Pain tends to be mild or even absent in some cases of osteomyelitis. This type of infection is caused by multiple microbes, with the primary bacteria being Staphylococcus aureus and Staphylococcus epidermidis. The decline in prevalence may be due to the widespread availability of antibiotics and improved oral and dental health standards.[4,15,16]

Acute osteomyelitis may progress to a chronic stage when antimicrobial agents prove ineffective. This chronic osteomyelitis may further become a refractory condition that is difficult to treat. Hence, the correct diagnosis of osteomyelitis is decisive, and healthcare practitioners must have a thorough knowledge of the signs and symptoms for accurate evaluation and effective management.[7] Various methods can correctly diagnose chronic osteomyelitis, including microbial culture, bone biopsy, conventional radiography, computerized tomography, magnetic resonance imaging, and radioisotope bone scanning.[7] A radiological feature mainly associated with chronic osteomyelitis is the formation of a bony sequestrum.[17] Sequestration and laminations of periosteal new bone are crucial distinguishing features of osteomyelitis. Positive radiographic findings are delayed or secondary unless accompanied by a fracture.[5,18]

CBCT has been used in osteomyelitis (OM) cases to confirm the suspected diagnosis by identifying small osteolytic or sclerotic osteoblastic lesions and determining the extent of the affected area. It has emphasized that early detection of subtle bone changes, especially after radiotherapy, is essential. Orpe et al. described a series of 13 patients with chronic sclerosing OM of the mandible, noting that periosteal new bone formation, sclerosis, and bone enlargement are the most commonly detected characteristics.[8] In this case report, the FOD area experienced a decrease in bone density, and its internal part underwent lysis due to osteomyelitis.

Management of osteomyelitis entails a course of antibiotics in combination with surgical debridement (sequestrectomy). Surgical decortication is more effective than traditional surgical debridement for improving local vascularization in osteomyelitis of the jaws. This process removes poorly vascularized (infected) bone and introduces well-vascularized tissue, promoting healing and enhancing antibiotic delivery. Therefore, surgery and antibiotics are the main treatments for this condition.[6,15,19]

CONCLUSION

Based on clinical and supporting examination (Panoramic and CBCT), we concluded that the suspected radiodiagnosis is Florid Osseous Dysplasia with chronic suppurative osteomyelitis. While the patient is under observation and the therapy is antibiotic, anti-inflammatory, and anti-pain, a histopathology examination is needed to confirm the diagnosis and determine further treatment.

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FOOTNOTES

All authors have no potential conflict of interest to declare for this article. Informed consent was obtained from the patient for being included in this case report.

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