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Case report

A case of florid osseous dysplasia associated with chronic sclerosing osteomyelitis requiring segmental resection

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ABSTRACT

We report a case of florid osseous dysplasia (FOD) in the right mandible in a 40-year-old woman with communication difficulty due to mental retardation. Swelling and a mixture of radiolucency and radio-opacity were observed in the right mandible on an X-ray image. The mass was adjacent to the inferior border of the mandible and there was a radiolucent layer between the mass and the surrounding normal bone. A definitive diagnosis could not be made by biopsy and follow-up was performed. The mass increased during the follow-up period and segmental resection was performed. Clinical, X-ray, and histopathological findings indicated a diagnosis of FOD associated with chronic sclerosing osteomyelitis. An evaluation of the therapeutic regimen for FOD associated with secondary infection in this case suggested that aggressive surgical treatment and histopathological examination are required in such cases.

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1. Introduction

Florid osseous dysplasia (FOD) is classified into subtypes of osseous dysplasia, focal osseous dysplasia, familial gigantiform cementoma, and periapical osseous dysplasia in the 2005 WHO classification of odontogenic tumors [1]. Differential diagnoses include chronic sclerosing osteomyelitis (CSO) and ossifying fibroma. However, histopathology and X-ray images for these conditions may be similar to those of FOD, which may make diagnosis and treatment difficult. Here, we report a case of FOD with CSO associated with marked bone expansion in a patient with mental retardation.

2. Case report

The patient was a 40-year-old Japanese woman with a chief complaint of swelling in the right mandible. The patient had developed cerebral palsy and mental retardation after a triple vaccination for diphtheria, pertussis and tetanus when she was 3 months old. She underwent surgery for subarachnoid hemorrhage when she was 33 years old and had taken no oral medications at that time. Her family history was unremarkable.

[☆] AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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Her family had found a swollen mass in her right mandible 2 months before the initial examination. The mass had been observed for a while, but had increased in size prior to her first visit to our hospital. At the initial visit, the patient was obese and her body mass index (BMI) was 32. Her nutritional status was good. There were no other abnormal findings, but she could not communicate well because of mental retardation.

An extra-oral examination showed a swollen mass in the right mandible, but no other abnormalities in her lips and jaw movement (Fig. 1). An oral examination showed poor oral hygiene. A hard bone-like swelling with a normal surface was present from the distal part of 43 to the molars. There were many remaining stump teeth, including that of 44 at the swollen site (Fig. 2). Redness was observed in the gingiva around the stump, but with no pus discharge. The presence of pain and numbness could not be determined due to difficulty in communication.

X-ray findings showed a mixture of radiolucency and radio-opacity in the right mandible, which was suspected to be due to a fibro-osseous lesion. The lesion was separated from the surrounding normal bone by a radiolucent layer (Fig. 3). In a CT scan, the lesion was well-defined and heterogeneous with a mixture of high and low signal intensities. The mass was centered in the area of 44 and adjacent to the inferior border of the mandible, spreading from the area of 43 to the area of 46, with compression of the mandibular canal (Fig. 4). In blood tests, the white blood cell count (WBC) and C-reactive protein (CRP) level were slightly increased, but the levels of alkaline phosphatase (ALP) and other molecules were within their normal ranges.

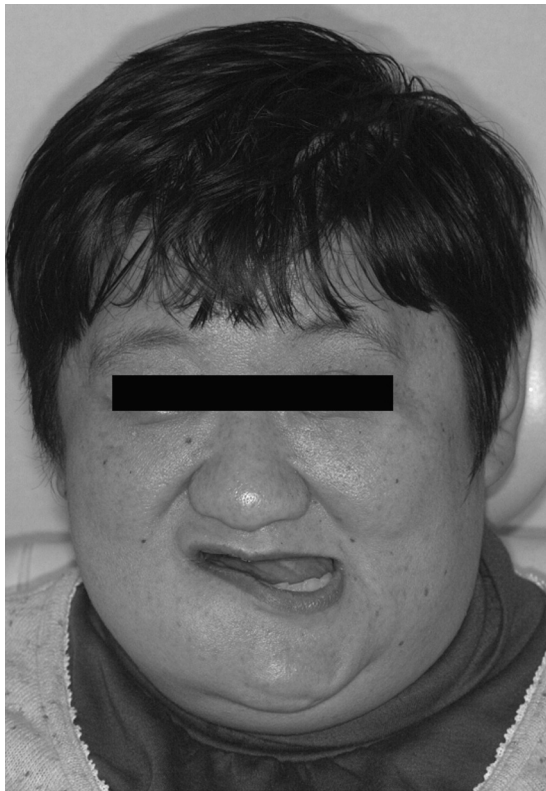


Fig. 1. Face at the initial examination: swelling in the right mandible was observed.

A fibro-osseous lesion was suspected and biopsy and extraction of the remaining teeth (except for 44) were performed under general anesthesia. The biopsy showed an increase of bone tissue with inflammation. However, neither growth of fibrous tissue nor a possible tumor was observed. Oral hygiene instructions and anti-inflammatory therapy with antibiotics were given to the patient at the outpatient department. The oral environment was improved, but the WBC count and CRP level remained high. Comparison of the mass using 3D models (Z printer™ 3D Systems) made from CT images obtained at the initial examination and 2 months later showed that the width, mesiodistal diameter, and distance from the inferior border of the mandible had increased by 4, 3, and 2 mm, respectively (Fig. 5).



Fig. 2. Intraoral conditions at the initial examination: a hard bone-like swelling with a normal mucosal color was present from the distal part of 43 to the molars. Swelling was observed on the remaining stump of 44.

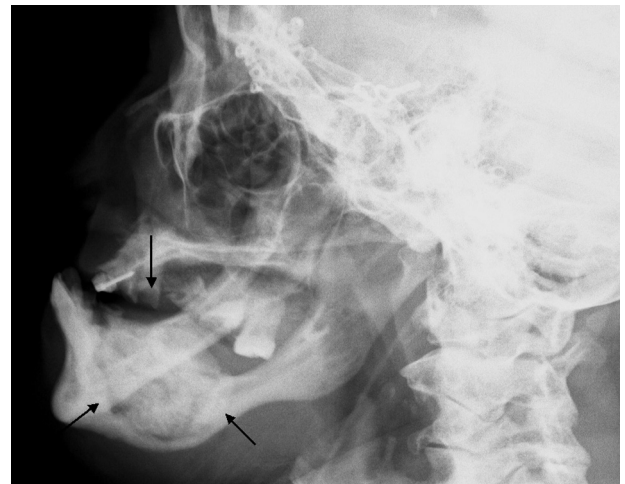


Fig. 3. X-ray image showing an oblique view of the mandible: a well-defined mixture of radiolucency and radio-opacity was apparent in the right mandible with a radiolucent layer between the mass and the surrounding normal bone.

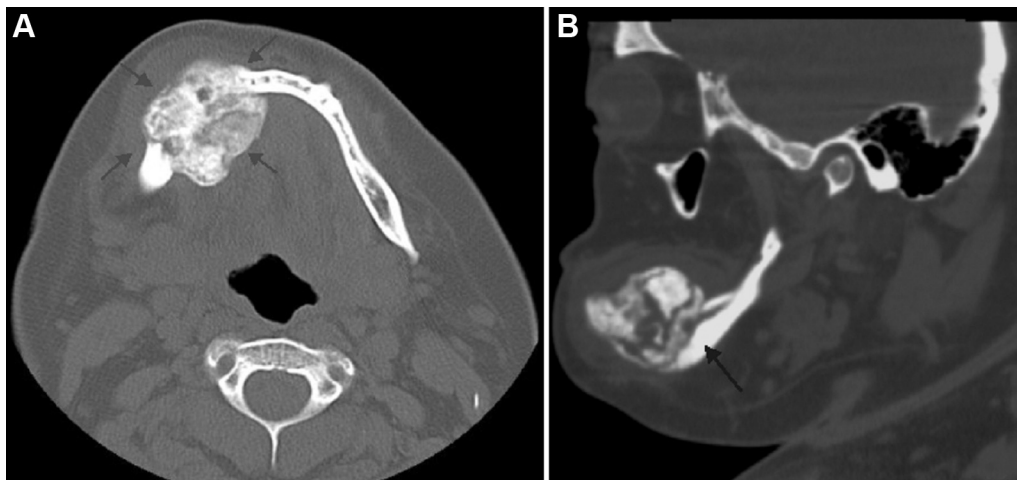


Fig. 4. CT images: a well-defined and heterogeneous lesion that gave a mixture of high and low signal intensities was observed (a). The mass was located adjacent to the inferior border of the mandible and compressed the mandibular canal (b).

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