CASE REPORT

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Polyostotic fibrous dysplasia of the craniofacial bones: a case report

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ABSTRACT

Objectives: This case report is aimed to show the radiographic features of fibrous dysplasia, a rare benign condition of abnormal bone growth that progresses slowly and is marked by uneven bone trabeculae interspersed with excessive proliferation of cellular fibrous connective tissue.

Case Report: An 18-year-old female patient with a chief complaint of swelling on the right upper cheek was referred to the Oral and Maxillofacial Department of Universitas Indonesia Hospital. The patient is then referred to the Radiology Department to undergo a panoramic and CT scan examination. On panoramic and CT examination, signs typical of polyostotic fibrous dysplasia were found, namely "ground glass appearance" with ill-defined borders, which extended unilaterally and extended from the maxilla and mandible to the frontal bone.

Conclusion: Polyostotic fibrous dysplasia can be identified on panoramic radiographs, a reliable and widely available diagnostic tool. Computed tomography (CT) is an advanced imaging modality for further evaluation of the lesion extension and tissue involvement. Management requires careful consideration of multiple clinical and radiographic factors.

Keywords: *Fibrous dysplasia, panoramic radiograph, CT-scan*
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INTRODUCTION

Fibrous Dysplasia is a benign but expansive bone growth that progresses slowly. It is marked by uneven bone trabeculae interspersed with excessive proliferation of cellular fibrous connective tissue, which replaces normal bone.[1] The etiology of fibrous dysplasia is still unknown.[2] It might appear as a single lesion, known as monostotic, or as several lesions affecting numerous bones, known as polyostotic.[3]

Given its 1:4000–1:10,000 incidence, it appears to be a rare condition.[4] The monostotic form, which accounts for roughly 70% of all occurrences of fibrous dysplasia, has no sex-specific preference.[5] The most common symptom of monostotic fibrous dysplasia is painless swelling, which is typically identified in the second decade of life.[1] Polyostotic fibrous dysplasia is a rare condition that is more common in females. Nearly all individuals with polyostotic fibrous dysplasia exhibit craniofacial involvement, with a startling predominance on one side of the body. The number of damaged bones ranges from a few to 75% of the complete skeleton.[6]

Sclerosis, thickening, and enlargement are typically visible in the afflicted bones. The mandible and maxilla are the bones most frequently affected. Ethmoid, sphenoid, frontal, and temporal bones are rarely involved. Patients may experience tooth displacement, facial asymmetry, hearing problems, and vision anomalies, depending on the bones involved.[7]

Computed tomography (CT), magnetic resonance imaging (MRI), and panoramic radiography can all be used to assess fibrous dysplasia.[8] Panoramic radiography offers the advantage of a two-dimensional imaging technique with broad coverage of both the maxilla and mandible, enabling visualization of extensive jaw lesions such as fibrous dysplasia. Computed tomography (CT), as an advanced three-dimensional imaging modality, is required to assess lesion expansion in the third dimension and to evaluate both osseous and soft tissue involvement.[24] While magnetic resonance imaging (MRI), also a three-dimensional modality, is preferred since it has superior soft tissue contrast,

making it more suitable for the characterization of lesions involving soft tissues.[25] This paper presents a radiographic feature of polyostotic fibrous dysplasia in an 18-year-old female.

CASE REPORT

An 18-year-old female patient with a chief complaint of swelling on the right upper cheek was referred to the Oral and Maxillofacial Surgery Department in Universitas Indonesia Hospital. The patient had been aware of the swelling of the right cheek since the age of 10.

The patient was then referred to the Radiology Department for panoramic examination. On panoramic examination, the right maxillary region appeared to be enlarged. There were mixed radiopaque and radiolucent images extending from the right maxillary tuberosity region involving the entire alveolar bone to the periapical region of tooth 21, expansion involving the entire right maxillary cavity and right cavum nasi, “ground glass appearance” in internal structures, ill-defined borders, and loss of lamina dura in teeth 18 to 11. The right mandibular region also appeared to be enlarged, with mixed radiopaque and radiolucent images extending from the right mandibular ramus, corpus, symphisis, to the left mandibular quadrant involving the entire alveolar bone up to the periapical region of tooth 36, with “ground glass” internal structures and ill defined borders, loss of lamina dura in all teeth of the right mandibular region up to symphisis region. There was a loss of right mandibular canal borders.

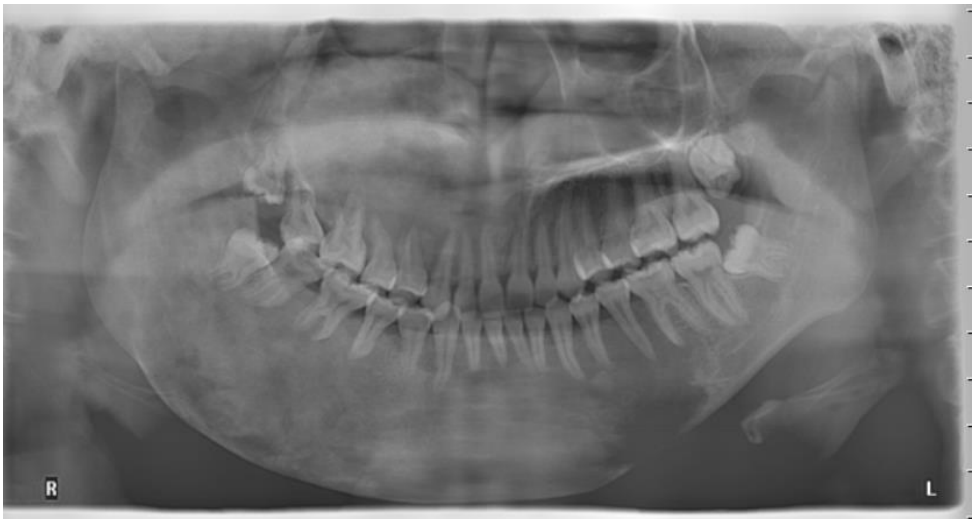

Figure 1. Panoramic radiography shows a mixed radiolucent and radiopaque lesion with a ground-glass appearance

The patient was advised to have a CT examination to get a more detailed diagnosis and expansion of the lesion. On CT examination, an irregularly shaped radiopaque mass was visible with ill-defined borders, internal structures of mixed radiopaque and radiolucent resembling “ground glass” extending from the right mandibular ramus, corpus, symphysis, reaching the left mandibular quadrant, involving the entire alveolar bone, resulting in narrowing of the right mandibular canal. In the right maxillary region, it appeared to extend from the right maxillary tuberosity involving all alveolar bone to the region of tooth 21, appearing to extend to the right frontal bone, right sphenoid-temporal, clivus, medial-lateral wall of the right orbita, paranasal sinus wall, sphenoid, ethmoid, right pterygoid, cribifrom plate, and lamina papyracea, as well as the right intracavitary bone structure of right nasi, appearing to protrude the right orbital cavity.

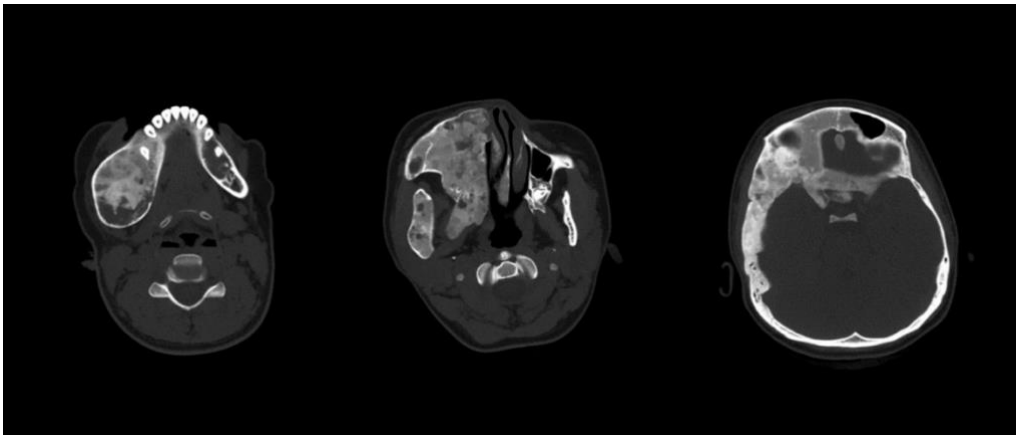

Figure 2. Selected axial section for analysis

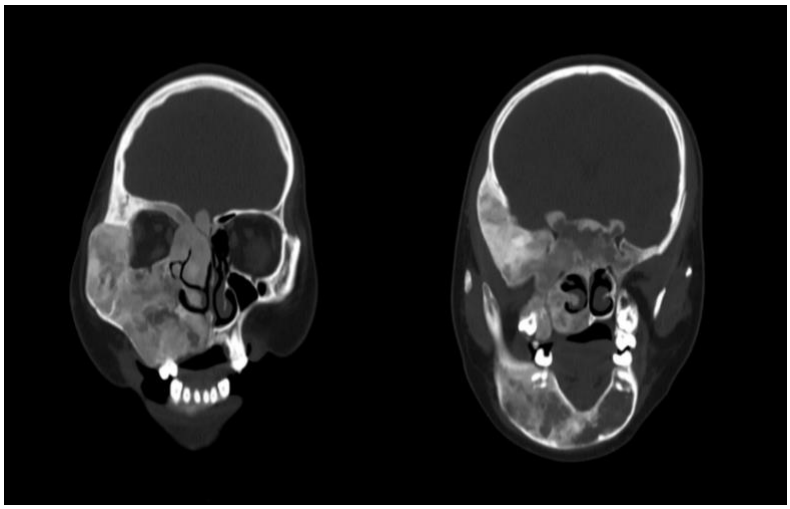

Figure 3. Selected coronal section for analysis

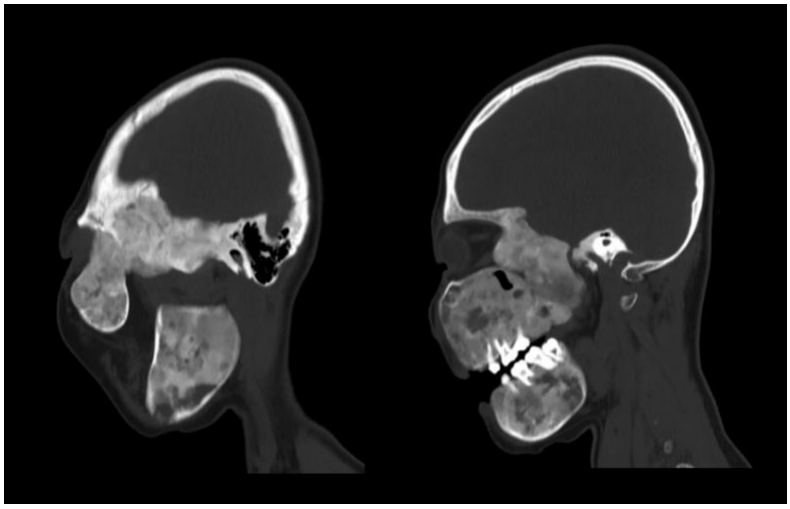

Figure 4. Selected sagittal section for analysis

Based on the results of the panoramic examination and CT scan, the radiodiagnosis of the patient leads to fibrous dysplasia. Periodic monitoring is planned to monitor changes in the lesion and growth of the patient. Orthodontic treatment and cosmetic surgery will be deferred until after the maturation of the bone.

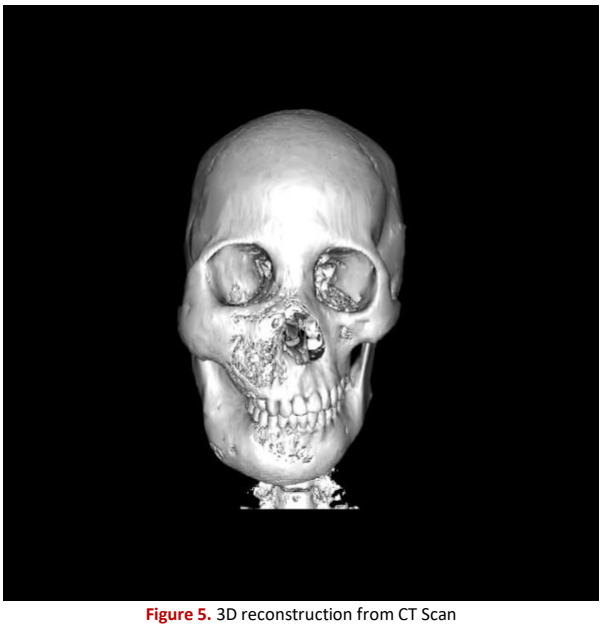

Figure 5. 3D reconstruction from CT Scan

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DISCUSSION

Fibrous dysplasia is a type of fibro-osseous lesion that is defined by the intermix of osseous and fibrous tissue, which results in a developmental hamartoma. Osteoblasts do not mature properly, therefore the resulting secondary bony metaplasia produces immature, newly produced, and weakly calcified bone. 10%–25% of monostotic cases and 50% of polyostotic cases involve the craniofacial bones.[9] Fibrous dysplasia of the jaws more commonly presents as the monostotic (solitary) type, involving a single bone location. Polyostotic fibrous dysplasia, in contrast, affects multiple skeletal sites. Radiographically, both forms demonstrate variable imaging features, including alterations in bone density and diverse trabecular patterns. A small group of polyostotic fibrous dysplasia can also be a part of McCune-Albright syndrome, a multisystem developmental condition that is linked to café au lait cutaneous macules and endocrine hyperfunction.[3] In this patient, signs of McCune-Albright syndrome were not observed.

The overall incidence of fibrous dysplasia is 3.6 per 1,000,000 person-years.[10] In the craniofacial area, fibrous dysplasia is quite uncommon, occurring in approximately 10%–25% of monostotic cases and 50% of polyostotic cases.[11] Any age can be affected by fibrous dysplasia. However, 75% of patients present before the age of 30, and it is typically seen in children and young adults.[12] Some studies say that the predilection of polyostotic fibrous dysplasia in females is higher than in males.[6,8] As in this case, which occurred in an 18-year-old female patient.

In polyostotic fibrous dysplasia, the maxilla is more frequently affected than the mandible in the craniomaxillofacial region, and it typically affects other bones like the sphenoid, zygoma, and frontal bone. It also often affects the femur, tibia, and pelvis, but may involve nearly any bone in the body. Patients frequently present with a hard, gradually increasing, painless swelling in the afflicted bones. Depending mostly on the anatomical sites and structure involved, its overgrowth tends to cause facial asymmetry, deformity, malocclusion, orbital dystopia, exophthalmos, and paresthesia or anesthesia.[13] Most bony lesions become clinically significant and non-silent by the age of ten, and hardly any new lesions develop after the age of fifteen.[14] Polyostotic fibrous dysplasia may be unilateral or bilateral, but often shows asymmetric distribution, and in craniofacial fibrous dysplasia, bilateral craniofacial fibrous dysplasia is very rare. Therefore, the patient's onset and symptoms were consistent with the typical course of fibrous dysplasia. However, despite mild involvement of the orbital bones, the patient had no complaint related to the eye or sight.

While panoramic radiography can be used as a main diagnostic tool, CT imaging is necessary to assess the disease's severity, as CT scans can identify bone destruction, soft tissue masses, and malignant transformation.[15,16] Compared to MRI, CT is a better modality due to its ability to clearly define bone morphology, whereas MRI is complementary, used when there is suspicion of non-osseous involvement or complications. CT scan is also generally considered a better imaging modality than CBCT (Cone Beam CT) for evaluating fibrous dysplasia due to its ability to show the ground-glass matrix more clearly, provide better soft tissue evaluation, cover larger anatomical areas, and support more accurate quantitative assessment. Ground glass appearance with a thin cortex and no clear borders is one of the characteristics of craniofacial fibrous dysplasia in panoramic radiography.[17] A ground-glass pattern (56%), a homogenously dense pattern (23%), and a cystic pattern are the three types of fibrous dysplasia that can be identified by CT.[18] As the disease advances, it may change from having a homogeneous appearance to having a mixed radiopaque and radiolucent lesion.[15] It begins with radiolucency and progresses to a more mixed stage with a structure that is both radiolucent and radiopaque before becoming radiopaque.[19] As in this case, a panoramic radiography was done for initial diagnosis, then a CT scan was done to see a more detailed and clearer expansion of the lesion. In this case, the CT helped show a clearer image of a mixed radiopaque and radiolucent lesion with a definite ground glass appearance, diffused shape, and borders. The CT also helped identify extension of the lesion to the right side of the mandible, maxilla, and adjacent craniofacial bones such as the frontal, sphenoid, and temporal bones in a unilateral fashion. Additionally, expansion to the bony spaces such as the maxillary sinus, cavum nasi, sphenoid sinus, orbita, and mandibular canal was also seen. These radiographical appearances were highly indicative of polyostotic fibrous dysplasia.

Restoring normal facial cosmetics and correcting the related abnormalities is the primary treatment objective and challenge for these patients with craniomaxillofacial fibrous dysplasia.[20] When these conditions clearly impact a patient's facial appearance and functionality, early surgical intervention becomes both necessary and rational. The only method to fully resolve fibrous dysplasia is radical resection, but often times is not possible in polyostotic craniofacial fibrous dysplasia. Therefore, some authors recommended conservative shaving and curettage as the primary therapy of choice.[21,22] Surgical contouring of the bone is typically recommended after skeletal maturation ceases and bone enlargement stabilises, unless important anatomical structures are disturbed during active bony expansion. Until a patient has stable lesions that stop growing after puberty, wait-and-see is recommended. It is possible to achieve satisfactory functional and aesthetic outcomes with reconstructive procedures, and regrowth is possible. More extreme therapies are saved for relapse or after skeletal maturity.[20] Orthognathic surgery is also possible, as fibrous dysplasia still exhibits normal bony healing. Bisphosphonates are often used as medical treatments to reduce

increased bone resorption. They could reduce bone pain in fibrous dysplasia, but it's unclear how medical treatment would affect skeletal degeneration.[23] As per the above statement, the ideal treatment for this patient may be surgical contouring to restore the aesthetic and functional state.

However, urgency for surgical contouring is currently considered low because the patient was not functionally impaired and was still aesthetically tolerable. Based on those reasons, the patient is currently resistant to surgical approaches. The patient was also considered to be still in the puberty period, so it was recommended to observe any future changes in swelling or symptoms and conduct periodic controls.

CONCLUSION

Polyostotic craniofacial fibrous dysplasia is a fairly rare case. In this present case, the fibrous lesion was observed to extend across both the maxilla and mandible, which was clearly demonstrated on the panoramic radiograph. In the maxilla, the lesion had expanded to involve critical anatomical structures, including the maxillary sinus and the inferior orbital rim, thereby requiring further evaluation using advanced three-dimensional imaging modalities. Additionally, in this case, the characteristic variations in bone density and trabecular patterns were also clearly delineated on computed tomography (CT) imaging. For the treatment of polyostotic fibrous dysplasia, many things need to be considered, such as the age of the patient and the extent of the lesion. The most important thing in treatment is to restore cosmetics and function.

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FOOTNOTES

All authors have no conflict of interest to declare for this article. The authors certify that they have obtained all appropriate patient consent forms.

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