CASE REPORT

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Fibrous Dysplasia in the Maxillary Bone: The Early Diagnosis of Progressive Pain with Facial Asymmetry

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ABSTRACT

Introduction: Fibrous dysplasia (FD) is a genetic, non-familial skeletal disorder, where in medullary bone is replaced by fibro-osseus tissue. Ninety percent of FD instances involve the craniofacial region, which usually involves one or more bones. The ethmoid, sphenoid, frontal, maxillary, and temporal bones are the most affected, but the maxilla is most affected. Misdiagnosis and inappropriate diversity in investigations and therapies are frequently caused by the disease's rarity and varying presentation to other specialties. This study aims to analyze the signs, symptoms, and laboratory-radiology examinations of suspected FD patients at Dr. Kariadi General Hospital Semarang so that it can increase the sensitivity of medical personnel in establishing this diagnosis based on the symptoms and signs complained of by the patient.

Case Presentation: A case series study assessing three patients diagnosed with FD of the maxillae between January and December 2021 at Dr. Kariadi General Hospital, Semarang. The study found 3 patients diagnosed with FD of the maxillae consisting of 2 males and 1 female. The initial complaints reported in all patients were progressive facial pain, thick sensation in the cheek area, and asymmetrical facial shape due to abnormal protrusions. Physical examination found an increase in facial volume with a hard consistency. CT scan evaluation obtained a sclerotic image with ground glass opacity in two patients and hyperostosis in one patient. Surgery with the Weber Ferguson technique was undergone by two patients, while one patient underwent the Weber Ferguson Technique + Zoledronic Acid for 6 cycles. Both patients who underwent Weber Ferguson technique surgery experienced recurrence related to symptoms in the form of pain and increased volume in the facial area within 2–4 weeks after surgery.

Conclusions: Patients with complaints of progressive pain and asymmetrical shape in the facial area and physical examination found an increase in facial volume with hard consistency should be able to increase the suspicion of medical personnel regarding the diagnosis of FD. Further radiological examination with tissue histopathology should be performed to rule out or confirm the diagnosis of FD.

INTRODUCTION

Fibrous dysplasia (FD) is a rare, non-hereditary bone disorder characterized by the chronic deposition of non-neoplastic fibrous tissue in areas of healthy bone. Abnormalities in cell-to-cell signaling in the tissue and the resulting responses result in disruption of tissue structure, and loss of the primary structural features of mineralized tissue is a factor that has been identified as playing a role in the induction of FD [1].

Although data estimate the incidence of FD worldwide at 1 in 5,000 to 10,000 cases, accurate evaluation of the incidence is generally difficult because of the asymptomatic clinical manifestations that often occur in FD patients. Approximately 2.5–5% of all instances of benign bone lesions are thought to be FD

cases, which is a very small amount overall [2]. The majority of FD diagnoses occur in youth or young people. For FD, there is no gender preference. Five percent of all benign bone lesions are FD. Approximately 75% to 80% of FD cases are of the monostotic kind, which is the most prevalent [3].

Bone pain is considered one of the most common immediate symptoms complained by FD patients. Clinical studies have shown differences in the degree of pain experienced by patients, from minimal to severe pain, by patients in various age groups and FD subtypes [4]. Bone pain associated with FD tends to worsen with age, with approximately 81% of adult FD patients reporting bone pain compared to 49% of pediatric FD patients. Bone remodeling is considered an important component of the pathology and biology of FD. The process of bone remodeling is physiologically important for maintaining skeletal integrity, regenerating, and repairing microdamage, and maintaining mineral homeostasis. However, in FD patients, this process occurs asynchronously at some focal bone sites. Trabecular bone typically exhibits higher levels of metabolic activity than cortical bone [5]. This process can ultimately cause asymmetrical changes in bone shape.

FD is typically asymptomatic and monostotic. Bowing of the bone in cases of severe bone deformities can cause musculoskeletal dysfunction or hasten the onset of osteoarthritis. Orbital asymmetry can be caused by facial involvement. Mandibular expansion, frontal prominence, and proptosis are further possible face problems. Craniofacial cases carry a risk of cranial nerve deficits including visual and hearing impairment [6]. The most often documented neurologic side effects of FD are hearing loss and visual problems. In 0.5–1% of patients, significant FD degeneration has been documented. This risk is elevated after radiation therapy and in polyostotic illness, particularly in McCune-Albright patients. Malignant transformation to sarcoma is rare but may occur with prior radiation therapy. Lesions in the vertebrae can predispose to scoliosis and limit the patient's functional abilities [3].

Reports of incidents related to FD are still rarely reported in Indonesia. Research related to FD in Indonesia based on journal reviews found 3 articles reporting FD in the spine at Hasan Sadikin Hospital, Bandung, FD in the Mandibula at Hasan Sadikin Hospital, Bandung, and FD in the Maxilla at the Hassanudin University Dental and Oral Hospital [7–9]. The purpose of writing a case report is to explain the rare occurrence of FD in hospitals so that it can increase the sensitivity of medical personnel in establishing this diagnosis based on the symptoms and signs complained of by the patient.

A case report study was conducted on three patients diagnosed with FD of the Maxillae based on evaluation of signs and symptoms, physical examination, radiological examination, and tissue histopathology. The study was conducted at Dr. Kariadi General Hospital, Semarang between January and December 2021. Patients were evaluated from their first visit to post-operative follow-up.

CASE PRESENTATION

There were three patients with signs, symptoms, and radiological examinations that suggested a diagnosis of recurrent maxillary FD (**Table 1**).

The first case is an 18-year-old boy with thickening and pain (VAS 3) in his left cheek in the last 6 months. He was diagnosed with FD and underwent hemi-maxillectomy 4 years ago. The physical examination showed unilateral deformity in the left cheek, in palpation, we found hard consistency without pain. There was no abnormality in the nasal and oral examination **(Figure 1A).** The paranasal sinuses CT scan showed a sclerotic appearance with ground glass opacity in the super-anteroposterior of the left maxillary bone **(Figure 1B).**

Table 1. Patient characteristics, signs, and symptoms

Variables	Case 1	Case 2	Case 3
Age	18 years	50 years	30 years
Gender	Male	Female	Male
Symptoms	Pain (VAS 3) and thickening on the left cheek, headache	Pain (VAS 6) and swelling on the right cheek	Pain (VAS 7) and progressive thickening of the right cheek, with blurry vision and strabismus of the right side of the eye
Physical examination	Increased facial volume Abnormal mass of hard consistency No tenderness pain	Right side facial asymmetry and increased anteroinferior volume Abnormal mass of hard consistency Tenderness pain	Minimal right-side facial asymmetry Vision: 6/60
Paranasal sinuses CT-Scan	Sclerotic appearance with ground glass opacity	Hyperostosis picture	Sclerotic appearance with ground glass opacity

VAS: Visual Analogue Scale; CT-Scan: Computed Tomography Scan

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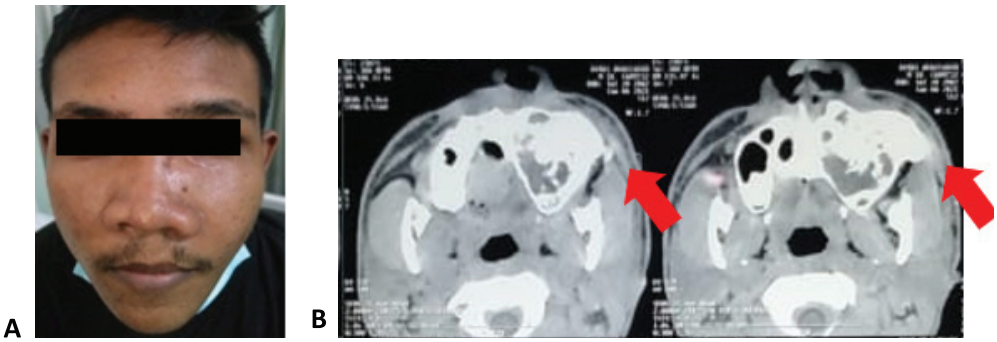

Figure 1. (A) Patient's profile; (B) Axial section of paranasal sinuses CT scan. The red arrows show the ground glass opacity in the supero-antero-posterior of the left maxillary bone.

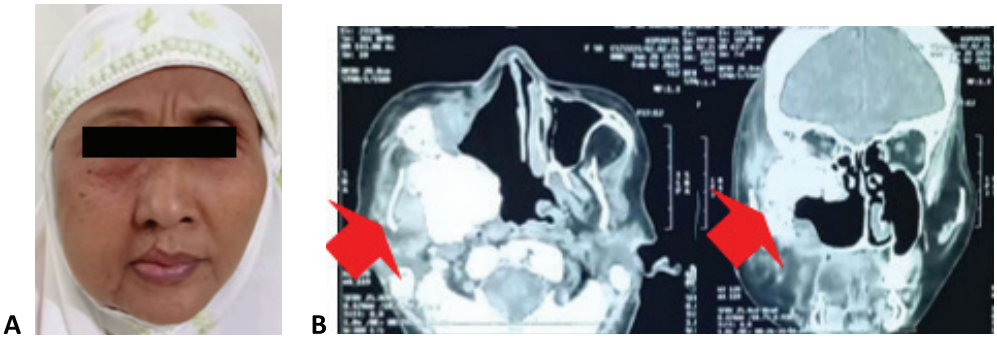

Figure 2. (A) Patient's profile; (B) Axial and coronal section of paranasal sinuses CT Scan. The red arrows show the hyperostosis appearance in the supero-postero-lateral of the right maxillary bone.

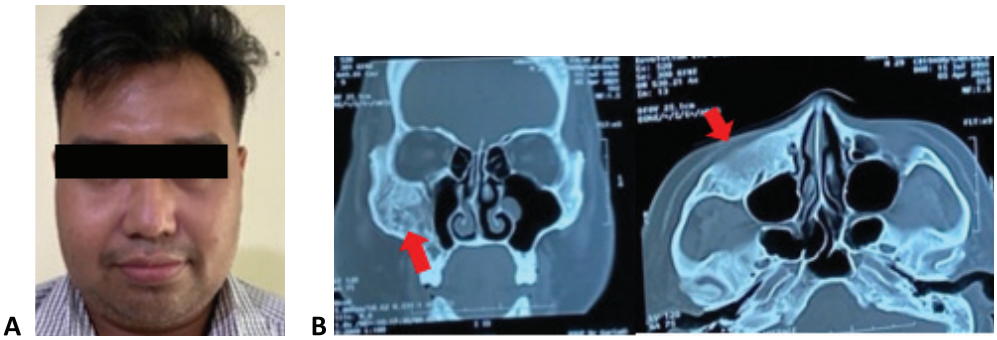

Figure 3. (A) Patient's profile; (B) Coronal and axial section of paranasal sinuses CT Scan. The red arrows show the ground glass opacity in the supero-antero-lateral of the right maxillary bone with an infiltrated periantral aspect of the maxilla.

This patient underwent a contouring operation with a Weber Ferguson incision. The tumor had a macroscopic appearance solid, white, and hard surface. One month after surgery patient reported pain (VAS 3) and increased volume in the same place for 2 weeks after surgery.

The second case is a 50-year-old woman who complained of pain (VAS 6) and swelling in the right cheek for 5 months. She was diagnosed with FD and had 8 operations in 6 years. The physical examination showed unilateral deformity due to the increased volume of the right cheek, with hard consistency and pain of pressure (**Figure 2A**). There was no abnormality in nasal and oral examination. The paranasal sinuses CT scan showed a hyperostosis appearance in the supero-postero-lateral of the right maxillary bone (**Figure 2B**).

The patient performed a contouring operation with a Weber Ferguson incision at the same place as the last operation. During surgery, we found a macroscopically spongiotic and yellowish appearance. One month after surgery patient underwent evaluation. A month after surgery patient reported pain (VAS 5) and increased volume of the right cheek.

The third cases are a 30-year-old man with symptoms of pain (VAS 7) and progressive thickening of the right cheek since 5 years ago. Three months lately he reported blurry vision in the right eye. He performed two previous surgeries in the last 2 years. The physical examination showed unilateral deformity of the right cheek and minimal scars were found (**Figure 3A**). The right eye examination found his vision was 6/60, with lateral strabismus, and limitation of mobility to lateral. The paranasal sinuses of the CT scan showed a sclerotic appearance with ground glass opacity in supero-antero-posterior of the right maxilla (**Figure 3B**).

The patient had surgery with Weber Ferguson Technique and completed 6 cycles of Zoledronic Acid medication. A week after therapy, the patient reported decreased pain (VAS 2) and no increased volume of the right cheek.

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Table 2. Management and evaluation post-management

Variables	Case 1	Case 2	Case 3
Management	Operation with Weber Ferguson Technique	Operation with Weber Ferguson Technique	Operation with Weber Ferguson Technique + Zoledronic Acid for 6 cycles
Evaluation	Increased pain (VAS 3) and increased facial volume 2 weeks after surgery	Increased pain (VAS 5) and increased facial volume 4 weeks after surgery	Decreased pain (VAS 2) and no increase in facial volume after surgery
Recurrency	Yes	Yes	No

VAS: Visual Analogue Scale

DISCUSSION

The etiology of fibrous dysplasia (FD), a benign lesion that is congenital and recurring, remains unknown. It is caused by missense mutations in the GNASl gene, which is found on chromosome 20. A bone modeling condition in which immature fibrous tissue continuously replaces regular bone. Lichtenstein made the initial discovery of this illness in 1938, and he and Jaffe worked together to report it in the medical literature for the first time in 1942 [10,11].

Typically, monostotic FD appears in the second or third decade. It is the most prevalent type of the disease and is distinguished by the involvement of only one bone without any systemic symptoms [12]. Around 75–80% of FD cases are of the monostotic kind, which is the most common [10,13]. Affects one in every 4,000 to 10,000 individuals [14].

Ninety percent of FD instances involve the craniofacial region, which usually involves one or more bones. The maxilla is the most afflicted, followed by the ethmoid, sphenoid, frontal, maxillary, and temporal bones [11]. Pregnancy can raise the risk of discomfort and pathologic fracture by increasing disease activity [13].

Most patients (90%) with FD are asymptomatic [10]. The lesion's location and the compressive force on the surrounding structures determine the symptoms and indicators. Clinical signs of maxillary FD typically include facial asymmetry, bulging of the afflicted area, and sluggish growth or volume rise. Clinical signs and symptoms include facial deformity and asymmetry, pathological fractures, obstruction of the paranasal sinuses that results in recurrent infections, cysts, and mucoceles, headaches, anosmia, changes in ocular movements, ptosis, exophthalmos, strabismus, conductive hearing loss, and loss of visual acuity due to compression of the optic nerve [15]. Speaking and chewing may become difficult when the lesion spreads into the oral cavity. In order to rectify the malposition of the affected teeth and jaw, this disorder may also involve periodontal and occlusal abnormalities, necessitating orthodontic treatment and orthognathic surgery [13]. In our cases the symptoms are dominant with pain and thickening of their cheek, one patient with expansion to eyes (blurry vision)

Image examination of computed tomography (CT), magnetic resonance imaging (MRI), or conventional radiographs, as well as biopsy, are used to confirm craniofacial FD [11]. An enlarged osseous lesion with a poorly defined boundary encased in an eggshell-thin cortex is the usual radiography result. On radiography, FD might also show up as a sclerotic or pagetoid lesion. Radiographic scans reveal a calcified, thick, expanded sinus edge and a mass inside the sinus that looks like ground glass when the paranasal sinuses are affected [10,11]. Treatment planning is facilitated by 3D reconstruction, which provides a more realistic three-dimensional picture of the lesion in terms of volume, density, and involvement of nearby anatomical structures [14].

In this case, a CT scan sinus paranasal (SPN) was the main radiographic examination to demonstrate ground glass opacity and hyperostosis in the maxillary. From it, we make a fundamental decision the surgical planning. The diagnosis was confirmed by pathologically and the treatment of choice was contouring. Histopathological evaluation showed irregular, immature bone trabeculae scattered within a fibrous tissue.

Histologically, the FD lesion reveals normal marrow-space bone replaced by irregular spindle-shaped mesenchymal cells. The mesenchymal cells form whorled patterns, and the bony trabeculae are poorly developed. One differentiating feature of FD is the lack of osteoblastic rimming, in which osteoblast and rim the fibro-osseous tissue [10]. There are three histological types of FD, according to research. A "Chinese writing pattern" is the term used to describe the classical form of irregularly shaped trabeculae of juvenile woven bone in a loosely structured, cellular fibrous stroma. The sclerotic/ pagetoid, which is connected to the cranial bones, is distinguished by dense, sclerotic trabecular bone with intricate cement line systems. The sclerotic/ hypercellular type, discontinuous bony trabeculae distributed in an ordered "often parallel pattern", is associated with gnathic and skull base bones [14,16]. In this case report, all three subjects had a sclerotic/pagetoid type of picture.

The location and radiographic features of the lesion, such as ossifying fibroma, FD, and osteomyelitis, frequently determine the differential diagnosis of FD [11]. Clinically, the FD lesion is easily distinguishable, even

though it may resemble a pagetoid on radiography. Since malignant degeneration occurs in 1 in 200 cases, osteosarcoma must always be taken into consideration. If the diagnosis is unclear or malignant degeneration is suspected, a biopsy should be done [10].

The best way to treat this illness is still up for dispute. Surgery is still the cornerstone of treatment, even if medicinal therapy plays a part in symptom control [16]. According to numerous clinical investigations, the location, function, and aesthetics of this lesion all influence the best course of treatment. The buccogingival or bicoronal route, the Weber-Ferguson incision, and the midfacial degloving method are some of the surgical approaches that have been suggested thus far for the treatment of FD in the midface [13]. Partial resection and bone remodeling are the best options. When the dentition is impacted or hearing is compromised, aggressive operations are performed. Surgery aims to restore normal face aesthetics by removing connective tissue, however, it frequently cannot correct orbital, nasal, and temporal issues [11]. Surgical treatment for decompression of cranial nerves (typically the optic, cochleovestibular, or facial nerves) helps restore function but should not be performed prophylactically because of the risk of iatrogenic injury. Surgical curettage or contouring can help with diplopia and relieve sinus obstruction. Although with this procedure. Recurrence rates reach 20% to 30%. Studies report that post-operative recurrence rates occur between 1-2 years after surgery. [17] In recent years, refinements in surgical instrumentation and craniofacial surgical techniques have made more aggressive non-disabling procedures possible. Additionally, bisphosphonates may reduce bony pain and the risk of fractures [10]. In these cases, the patients have done the contouring to reduce the pain and make a symmetrical face, as the lesion was not found to be aggressive.

The standard medical management for FD includes systemic bisphosphonate therapy. The efficacy of systemic bisphosphonate is however limited with minimal functional improvement, pain relief, and reduce the rate of the growth lesion of FD. Systemic zoledronate at a recommended dose of 4 mg or 0.1 mg/kg IV infusion over 1 h, is followed by intralesional bisphosphonate. At 6 months follow-up the improvement in the pain scores [18,19].

It has been demonstrated that locally acting bisphosphonates inhibit osteoclast function at the bone-implant interface while simultaneously stimulating osteoblast activity, hence facilitating bone-implant integration. Because only 50% of the intravenously administered drug can be incorporated into the bone matrix, systemic bisphosphonates given to a patient with FD are concentrated at the sites of the lesions; therefore, the concentration is likely to be significantly lower than that of the locally injected drug. As seen by our index patient's left tibia, local bone turnover at the FD lesion is only slightly inhibited at such low quantities. When given locally instead of systemically, the two may work in concert to suppress local bone turnover more deeply, which lowers uptake values and pain scores. Additionally, the higher drug concentrations obtained locally through intralesional administration may enable smaller and less frequent dosages. By increasing the drug's biological permanence at the intended site of action, drug-coated scaffolds or carriers can further boost the effectiveness of locally given bisphosphonate [18]. Fever and myalgia are the adverse effects that occur two to three days after zoledronic acid is used. An injection of calcium gluconate was an excellent treatment for a toddler who exhibited symptoms of hypocalcemia. Five patients had many obvious physeal growth arrest lines [19].

Radiotherapy is contraindicated as it leads to malignant transformation of the lesion, due to the high risk of being sarcomatous [14,15]. Observation with periodic follow-ups is used in the small and asymptomatic lesions [16]. The follow-up is of fundamental importance to detect relapses or a possible, malignant change at an early stage [14].

Face paraesthesia and mild transnasal crusting are the most frequent side effects of the midfacial degloving procedure. Although it happens in about 0.5% of patients, people with the polyostotic form are more likely to have malignant degeneration. Rapid tumor growth, discomfort, intralesional necrosis, bleeding, and an increase in the blood alkaline phosphatase level can all be signs of malignant alterations [12]. Craniofacial cases may have associated cranial nerve deficits including vision and hearing loss. Malignant transformation to sarcoma is rare but can occur with a prior history of radiation therapy. Patients with maxillary disease should receive coaching on how to keep an eye out for developing cranial nerve abnormalities, such as hearing and vision loss [13]. Routine assessment and radiological follow-up of the patient are always recommended due to the possibility of recurrence. Every three months or whenever the patient exhibits symptoms, we advise control.

Dalle Carbonare & Manisali [20] found that for both syndromic (84%) and non-syndromic (26%) patients, conservative surgery had a greater recurrence rate than radical resection ($p < 0.001$, CI 70.9 to 92.8 and $p < 0.001$, CI 21.8 to 30.6). Valentini et al. [21] recommended the only way to achieve total remission (no recurrence after 70 radical resections) for monocytic FD involving the maxilla and mandible is through radical surgery. In contrast, individuals with polystotic FD and McCune-Albright syndrome, particularly in zone 4, have a significant recurrence incidence (23%) following conservative surgery. Similar recurrence rates have also been evaluated by other researchers, and some have advocated for more aggressive zygomaticomaxillary therapy. Valentini et al. [21] also recommend putting

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off drastic resection in order to wait for the lesion to reach a static phase and skeletal development. Fattah et al. [22], found that compared to prior surgery, which had a 50% recurrence rate, radical excision following bone maturation had a 14% rate. Kim [23] also mentioned that to minimize recurrence, elective surgery should be postponed until bone maturation occurs, when FD lesions tend to become quieter. However, cases of aggressive disease with significant growth potential, especially in conditions of endocrinopathy and excess GH, are candidates for surgery, in which complete excision should be performed in patients of all ages.

CONCLUSIONS

One condition that can cause both functional and aesthetic harm is fibrous dysplasia. The patient's age, whether or not they have facial asymmetry, the severity of their pain, the involvement of their face, and their prospects for rehabilitation should all be taken into consideration when treating them. Restoring the patient's appearance, boosting their self-esteem, minimizing functional impairments, and relieving their discomfort are all crucial. When there is a considerable deformity, severe discomfort, or a pathological fracture, surgery is recommended. A crucial case for detecting recurrence or malignancy resolution is follow-up.

DECLARATIONS

Ethics approval and consent to participate

The research has obtained ethical permission from the Health Research Ethics Commission of Dr. Kariadi Semarang General Hospital.

Competing interest

The author(s) declare no competing interest in this study.

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