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**MANAGEMENT FOR RECURRENT FIBROUS DYSPLASIA OF THE MAXILLA: A
CASE REPORT**

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ABSTRACT

Background: Fibrous dysplasia is a developmental disorder of the bone, characterized as a benign pathological condition, and its etiology is not yet known. Fibrous dysplasia can occur in the entire body frame, but is most often found in the skull, maxilla, and mandible. Management of fibrous dysplasia is observation and conservative surgery. **Objective:** To report the management of recurrent fibrous dysplasia of the maxilla. **Case:** A 33-year-old female patient came to Universitas Airlangga Dental Hospital with a chief complaint of a lump on her right cheekbone, asymmetrical face, painless, and had a history of maxillectomy in the right region in 2013. CT radiography was performed with a result of polyostotic fibrous dysplasia involving the maxillary bone, nasal, zygoma, skull base, sphenoid, and right frontal bones. **Case Management:** Osteotomy and surgical recontouring were performed under general anesthesia, along with the extraction of teeth 11, 12, 21, and 22. Postoperative recovery was uneventful. **Conclusion:** Immediate surgical treatment for recurrent FD of the maxilla is necessary, and regular radiographic follow-up is important to see its progression.

Keywords: Fibrous dysplasia, Maxilla, Non-communicable disease, Recurrent, Surgical recontouring

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INTRODUCTION

Fibrous dysplasia (FD) is a rare non-odontogenic genetic disorder in bones characterized by the replacement of bone with fibrous tissue that can occur in one bone only (monostotic) or in several bones (polyostotic) throughout the skeletal area of the human body. Fibrous dysplasia most commonly appears on long bones, followed by the jaw (the upper jaw is more common than the lower jaw), the base of the skull, and the ribs. This bone disorder is a congenital disease, not a hereditary disease. The incidence of fibrous dysplasia is reported to vary from 1:4000 to 1:10,000. This disorder accounts for about 2.5% of the category of lesions on the bone and about 7% of the category of benign bone tumors.^{1,2} The patient demographics were similar to previous reports with an equal gender distribution and diagnosis usually in early adulthood (mean age 32.6 years).³

Fibrous dysplasia consists of three major types, namely 1) monostotic, which involves one part of the bone; 2) polyostotic, in the form of multiple lesions and involving multiple bones; and 3) McCune Albright syndrome, polyostotic form of FD that also involves endocrine abnormalities (Kim, 2023). FD can involve the bones of the craniofacial system. Research by Feller et al states that craniofacial bones are involved in 10% of monostotic fibrous dysplasia cases, and 50% in 50-100 polyostotic FD. Fibrous dysplasia of the jaw more often involves the maxilla than the mandible. In the jaw, involvement of the maxilla may be accompanied by zygoma or sphenoid bones.^{1,3}

The most common clinical symptom of FD is a slow, painless enlargement of the involved bone. Lesions that grow slowly can cause painless expansion of the involved bones, resulting in visible facial asymmetry. Involvement of the maxilla and mandible can cause tooth dysplasia, malocclusion, and sometimes resorption of the tooth root. Sometimes there is also a headache or paresthesia on the face. Sensory disorders can also arise as a result of the pinching of sensory nerves around the lesion. Rapid growth of FD of the maxilla and mandible, on the other hand, may produce airway obstruction due to the posterior displacement of the tongue.^{4,5}

In the radiographic incision, fibrous dysplasia is described as the appearance of "ground glass" with unclear borders and visible cortical expansion. The pattern of radiographic imaging of fibrous dysplasia involving

maxillofacial bones, namely cystic (radiolucent or lytic: initial lesion), sclerotic (mid-phase lesion), and radiolucent/radiopaque (pagetoid: late lesion).^{5,6} Characteristic for FD is a homogeneous opacity of radiodense mixed in normal, cortical thin bone and bone expansion. Computed tomography (CT) is the most excellent imaging modality, and three-dimensional reconstructions and stereolithographic models are useful in determining the level of involvement and in planning operations.^{1,5,6}

As no curative treatment exists, the main treatment goals for craniofacial FD are to improve functional capacity, alleviate pain, and attain satisfactory cosmesis. Treatment of FD is done *by surgical contouring*, which is the removal of tumor masses (lesions) surgically by restoring the contour of the bone as before. Recurrence after surgical excision is not uncommon, especially in younger patients or in cases where complete resection is not feasible due to proximity to vital structures.^{1,3,7}

This case highlights the importance of long-term follow-up and management in patients with FD, as well as the potential for recurrence even after a decade, emphasizing the need for timely intervention in progressive or symptomatic lesions.

CASE

A 33-year-old woman came to the Oral Surgery clinic of Universitas Airlangga Dental Hospital with the chief complaint that there was a lump on the right cheekbone. The lump is painless, relatively fixed in size, and there is a history of surgery to remove the right maxillary tumor (*Maxillectomy Dextra*) in 2013. On clinical examination, the patient's general condition was good, facial asymmetry was visible, the zygoma dextra regional mass appeared, the diffuse border, the size was approximately 4x3x2 cm, the color was the same as the surrounding tissue, minimal nasal deviation in the sinistra direction, the solid consistency was hard, and there was no tenderness. On intra-oral examination, the anterior maxilla sinistra prognati appears, the maxillary defect dextra region 13-18 appears, not oedematous, not hyperemic, and no tenderness.



Figure 1. Clinical picture of oral extra oral with facial asymmetry and anterior maxilla sinistra prognathus

CT scan shows *polyostotic craniofacial fibrous dysplasia* affecting the maxilla, nasal, zygoma, skull base, sphenoid bone, and right frontal. *Normal brain parenchyma* and thickening of the mucosa in the ethmoidal sinuses.



Figure 2. Axial CT scan shows ground glass image

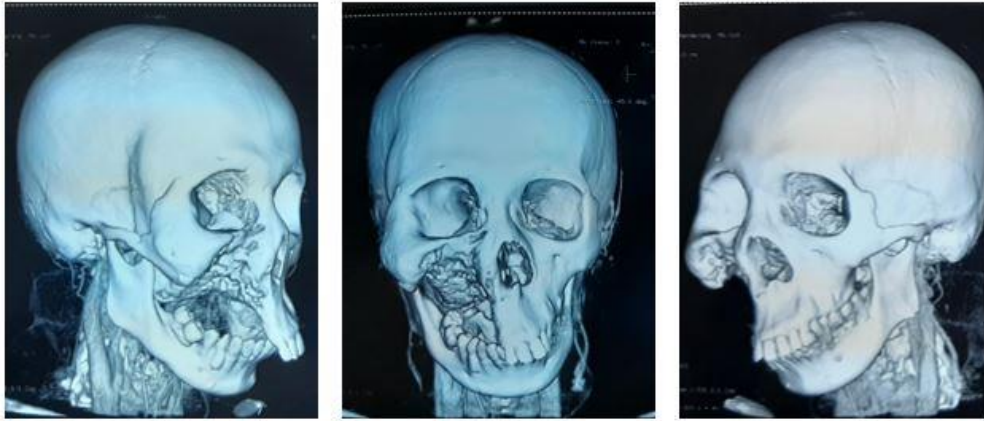


Figure 3. Reconstructed 3D image showing bone thickening in maxilla, nasal, zygoma, skull base, sphenoid and right frontal

CASE MANAGEMENT

The treatment plan in this case is *surgical recontouring* and *ostectomy* of the anterior maxillary region and extraction of teeth 11,12,21,22 with general anesthesia. The anesthesia procedure begins with orotracheal intubation using the endotracheal tube. The operation begins with aseptic action on the area of extra surgery and intra-oral. Inject lidocaine plus adrenaline diluted 1:200,000 at the incision site. In extra oral fills with Weber Ferguson design.

The flaps are opened layer by layer and bleeding control is carried out using a *cauter*. The next stage is *ostectomy* of the zygoma dextra and maxilla anterior using chisel and mallet, then the flap is continued intra oral and then *surgical recontouring* of the zygoma dextra bone and anterior maxilla using bur cutter cross until the bone matches the anatomical shape. Tooth extraction 11, 12, 21, 22 and flap closure by suturing are performed.



Figure 4. *Ostectomy* procedure of the zygoma dextra and maxillary anterior using chisel and mallet, and *surgical recontouring* of the zygoma dextra bone and anterior maxilla using bur cutter cross

The patient was treated for 4 days postoperatively with fluid therapy in the form of intravenous fluids and a liquid diet, antibiotics, analgesics, and antiemetics. Postoperative take-home medications are antibiotics, analgesics, and mouthwashes. Postoperative instructions for maintaining oral hygiene and control on the 10th day post-operative for suture collection



Figure 5. Post 10th day of operation

DISCUSSION

Fibrous dysplasia is a chronic developmental tumorlike condition, characterized by replacement of normal bone by a proliferation of cellular fibrous connective tissue with irregular bony trabeculae. This sporadic condition results from postzygotic, activating mutations in the *GNAS* gene, which encodes the alpha subunit of a stimulatory G protein. The extent of disease depends on when the *GNAS* mutation occurs. Its onset is frequently associated with rapid skeletal growth and exhibits a self-limiting pattern, because once bone growth ceases, FD becomes more quiescent or static.^{1,2,8}

For all patients, diagnosis was primarily based on imaging findings.³ The preferred imaging examination is cone-beam CT, since it offers a broader view of the extent and location of the lesion, which helps in planning the surgical procedure. Radiographically, the FD picture varies from radiopaque with indefinite boundaries to mixed lesions in the form of bone radiolucent involved, depending on the stage and development of the disease. The most frequent X-ray characteristic of craniofacial FD is “ground glass” appearance with a fine cortex and no defined borders; this was the kind of image found in examination of our patient. Ogunsalu et al. propose the following categories of X-ray appearance: (1) orange peel/ground glass appearance; (2) opacification of the antrum; (3) radiolucent lesion of the mandible; and (4) varying degrees of opacification (not ground glass and not orange peel).^{1,5,6,9} This is consistent with radiographic images found in this case, where the bone appears to be pressed against higher-density fibrous tissue with lower trabeculation. It also appears that the right maxillary sinus is filled with bone mass.

The treatment of fibrous dysplasia depends on clinical findings and symptoms, so treatment can vary. If the lesion does not develop and the patient does not complain about changes in facial shape, then the treatment is sufficient by observation and monitoring of the changes in lesions that occur, and regular examinations are carried out. However, if there are complaints from patients and there is a rapid development of lesions, then surgical methods should be done for the removal of FD and also consideration of post-enhancement facial reconstruction.¹ In thinner bones such as the maxilla, the bone cortex will develop faster and larger than in thicker bones. This causes lesions in the craniofacial area capable of causing complications, such as pathological fractures, facial paralysis, malignant transformations, rapid growth of FD of the maxilla and mandible that may produce airway obstruction due to the posterior displacement of the tongue, involvement of the skull base that may affect the optic canal and cause subsequent blindness.^{5,10} Lesion not showing rapid growth, no new onset of pain or paresthesia, and visual or hearing changes, such as in our study warrants immediate surgical treatment.^{11,12}

In this case, the treatment method chosen is surgical applying *osteotomy* followed by *recountouring*. This is done because the patient complains of a lump that results in facial asymmetry so that recontouring is done to restore the shape of the bone to symmetry with the sinistra side. For the maxilla, by an upper gingivobuccal incision, burring is performed with caution regarding the dental roots and infraorbital nerve.^{1,7} Post-operative control was carried out on the 10th day and periodical radiography was performed every six months to see the progression of fibrous lesions in the dysplasia.

It is stated that it is impossible to prognosticate whether the FD will recur or not. However, Kim (2023) mentioned that *resection with reconstruction* had shown a lower rate of recurrence than recontouring procedures only (45% vs. 82%, respectively) despite the greater morbidity.¹ Some authors also report that the growth of FD lesions tends to stop around puberty; however, other studies indicate that their progression is continuous into adulthood. Another important fact is that approximately 20–25% of these tumours continue to grow between 1-2 years after surgery, regardless of the type of therapy applied.^{7,9} Therefore, routine assessment and radiological follow-up of the patient are always recommended due to the possibility of recurrence. Follow-up visit was advised every three months or whenever the patient exhibits symptoms.⁷

CONCLUSION

Immediate surgical treatment for recurrent FD of the maxilla is necessary, and regular radiographic follow-up is important to see its progression.

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