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Case Report

A rare case report of fibrous dysplasia of maxillary bone causing progressive vision loss: Case report

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ABSTRACT

Fibrous dysplasia is a benign tumor of bone, uncommonly affecting the maxilla, congenital, recurrent character and etiology unknown, caused by a bone modeling disorder in which normal bone is replaced by immature fibrous tissue. The lesion may involve one or more bones being the maxilla the facial bone more affected. We report a case of monostotic variety fibrous dysplasia in a male patient complaining of progressive swelling in rt maxilla that compress the inferior orbital bone and finally produce significant loss of vision. In this case, Computed Tomography (CT) was the main radiographic examination to demonstrate the extension and radiodensity that the lesion assumes in the craniofacial bones, being therefore of fundamental importance in the surgical planning and in the longitudinal follow-up of the operated patient. The diagnosis was confirmed by anatopathology and the treatment of choice was removal of the tumor [total maxillectomy in this case followed by reconstruction of palate and defect. This article also aims to review the main clinical, radiological, histopathological, differential diagnosis and Fibrous dysplasia treatment.

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1. Introduction

Fibrous Dysplasia (FD) is described as a slowly growing, benign tumour disorder of bone development characterized by the replacement of normal bone by excessive proliferation of cellular fibrous connective tissue interspersed with irregular bone trabeculae.^{1,2}

In 1891, Von Recklinghausen denominated de “generalist fibrous osteitis” pathological conditions that characterized deformities and bone alterations.^{3,4} Therefore, only in 1938, Liechtenstein and Jaffe recognized these conditions described, previously, as a well-characterized disease and named it fibrous dysplasia.^{5,6}

The etiology of this entity is still unknown.^{7,8} Clinically it is classified as monostotic or polyostotic, the first being focal, limited to a single bone, and the second, multifocal,

involving several bones simultaneously.⁹

The monostotic form shows no predilection for sex and is found in about 70% of all cases of fibrous dysplasia, with gnathic bones being the most commonly affected sites.¹⁰ The cases of monostotic fibrous dysplasia are usually diagnosed during the second decade of life, with painless progressive hard swelling being the most common feature.

Polyostotic fibrous dysplasia is an uncommon condition, presenting a higher prevalence for females. The number of bones affected ranges from a few up to 75% of the entire skeleton and almost all patients with polyostotic fibrous dysplasia present craniofacial involvement with surprising predilection on one side of the body.¹¹ The involvement of two or more bones associated with brown-to-white pigmentation characterizes Jaffe-Lichtenstein Syndrome.

Clinically patient present with: slow growing progressive swelling with bulging of the involved region and facial

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asymmetry (when in great proportions).^{12–14} The soft tissue over the swelling is normal in appearance. Pain and paraesthesia are rare complaints, with slow growth of the lesion, and the patient often cannot remember when the lesion was initially perceived.

Diagnosis can be made by conventional radiography, scintigraphy, magnetic resonance imaging and Computed Tomography Scans (CT)¹⁵ although CT is considered the investigation of choice in the evaluation of craniofacial lesions.

Currently, the surgical treatment is resection of entire tumor [total maxillectomy with orbital reconstruction or osteoplasty with cosmetic reconstruction of palate and defect of maxilla by our prosthodontics colligues] is recommended when there is functional or aesthetic impairment caused by the lesion, however it should be postponed as much as possible due to the possibility of recurrence in cases where bone growth is active.^{7,16}

Thus, the present study aims to report a case of monostotic bone fibrous dysplasia, as well as the clinical, imaging, laboratory, histological and development of the case aspects will be discussed.



Fig. 1: Pre-operative image of patient.



Fig. 2: Healed weber ferguson incision with barbosa extension seen in the post-operative image.

2. Case Report

Our Patient Ratan Ji, 35-years-old, Hindu male, attended ENT OPD at SRG hospital Jhalawar on 01/07/2021 at 01:00PM with Complain of 1. Massive swelling over right Maxilla that pressing the right inferior orbital wall and causing loss of vision. 2. Pain since last 3 months 3. Bulging of right upper hard palate which causing difficulty in chewing and swallowing 4. Progressive loss of vision since last 2 months.

History of Present Illness:- Patient was apparently healthy 8 months back than he faced injury by bamboo stick over right maxillary region than he attend local primary health center and patient treated for 2 days by local antibiotics, analgesics, clean stitch and antiseptic dressing than patient discharged for home. After 10 days patient noticed a small swelling over right maxillary region that is gradual in onset, progressive in nature. And after 5 month patient experienced mild dull aching pain over right maxillary & frontal region. Than swelling is progressive day by day & pressing the right orbit causing the orbital compression and slowly progressive loss of vision. At the time of attending the ENT OPD at SRG Hospital swelling was very big 89×68×71mm [CT Findings] causing the orbital compression and loss of vision [Visual Acuity Right Eye 6/24 Left Eye 6/9] by the report given by ophthalmologist.

2.1. Past history

Patient attended multiple hospitals with above mentioned complains, No history of similar complain in the past, patient was hypertensive but doesn't have any treatment history. No history of Tuberculosis, Bronchial asthma & any other systemic disorders.

2.2. Personal history

Patient Belong to low socioeconomic status, Labourer by occupation, Vegetarian by diet, Chronic smoker since last 15 years, No history of snuffing of Nicotine & other snuffing agents. Patient doesn't have any history of occupational exposure.

2.3. Family history

No significant family history present in this case.

Than patient admitted in ENT Ward. Than advised for all routine Investigations CECT PNS with Oral cavity & Orbit & Neck, Bone Scan, Biopsy.

3. Biopsy

Biopsy already taken in our department under general anaesthesia by endonasal approach after reporting by the pathologist the histopathological features are suggestive of Fibrous dysplasia.

4. CECT Paranasal Sinuses

Imaging features are expansile heterogenous soft tissue density mass measuring approx. size 89×68×71 mm is noted in the right maxillary sinus with expansion of the region and mass effect over the inferior wall of orbit, medial wall of maxillary sinus as well as extending in to intraorally through the maxilla bone with obliteration of the left nasal cavity suggestive of bone tumors. This tumor pressing the inferior orbital wall and increase the intraocular pressure that deviate the eye balls upward and laterally and severe loss of vision in right eye in compare to the left eye.

4.1. Eye examination

Due to severe loss of vision that is noticed by patient itself patient refer to the our eye department for complete preoperative eye examinations.

1. Eye:- Right eye turned upward & Laterally, Left eye was normal in position
2. Eyelid: Right Eye lower eyelid edema, Left eye NAD
3. Conjunctiva: In right eye chemosed, hypertrophied & velvety, Left eye NAD
4. Cornea: In right eye not fully visible, hazy, left eye normal
5. Vision:- severely affected in right eye 6/24, Left eye is normal 6/9
6. Fundus Examination:- C:D ratio >0.3, mild glaucamatus changes present on disc & retina in right eye, Left eye is normal
7. Tonometry: In right eye intraocular pressure is raised 34 mm of hg with 5.5gram, Left eye 21 mm of hg.

4.2. Other examination

Before going to surgery we should evaluate the patient thoroughly by general physical examination, all essential routine investigation like [cbc, Lft, kft, ecg, x ray chest, bt, ct, hiv, Hbs,] other radiological investigation like NCCT PNS,

After complete work up with proper discussion with ophthalmologist we planned surgery total maxillectomy with orbital floor reconstruction and maxillary prosthesis with palate reconstruction by prosthodontics.

Than patient refer to anaesthesia department for preaneasthetic check up, than all the preoperative preparation is done one day befor surgery.

NBM. overnight Antianxiety medication Intravenous fluid RL Intravenous antibiotics.

Proper informed written consent.

4.3. OT notes

Total maxillectomy with orbital floor reconstruction by using locally available bones, cartilage, and muscle all are sutured nicely to each other hemostasis achived. Antibiotic

ointment soaked pack is put in rt maxillary sinus and nasal cavity. Weber furgusion incision is sutured in layer. clean and antiseptic dressing done. ryles tube no.16 is put in situ for feeding.

4.4. Postoperative care

Immediate post operative care using intravenous broad antibiotics, intramuscular analgesic, clean and antiseptic dressing, and ryles tube feeding, than patients refered to the prosthodontics for maxillary prosthesis and palate reconstruction.

4.5. Postoperative eye examination

Eye:- rt eye return in normal position, left eye normal
Eyelids:- both eye lids return to normal condition
Conjunctiva:- slightly chemosed

Cornea:- normal Vision:-6/9 in both eye.

Fundus:- show reversible changes.

Tonometry:- normal intraocular pressure 20 mm of hg.

At the extra-oral clinical examination, in an inferior-superior view, facial asymmetry was observed, presenting a volumetric increase in the zygomatic region, right maxilla, deletion of the right nasogenian sulcus, no alteration of skin color, lymphadenopathy or any other alteration.

At the intraoral examination, there was an increase in volume in the region of end of sulcus from the canine to right maxillary tuberosity, with erasure of the vestibular sulcus. Of hard consistency to the palpation, with oral mucosa of normal appearance, smooth surface, regular and without ulcerations.

Computed tomography revealed a hyperdense image, an expansive mass in the maxilla palate and with compression of the right nasal cavity.

Face CT revealed an insufflating lesion, with dense glass density in the maxillary bone, partially occupying the right maxillary sinus and the ipsilateral nasal cavity and lowering of the hard palate with involvement of the dental alveoli. 3D reconstruction offers a better three-dimensional understanding of the lesion, being more accurate in relation to the volume, density and involvement of adjacent anatomical structures, which facilitates the treatment plan.

Patient prepared for surgery, patients submitted to a surgical procedure under general anesthesia, by weber furgusion incision approach.

Macroscopically we can see a hard, -looking bone lesion. All material was collected and sent for histopathology examination in our department of pathology in jhalawar medical college for histopathological analysis, which confirmed the diagnosis of fibrous dysplasia.

Patient referred for post-anesthetic recovery under anesthesiologist care, evolving postoperative without intercurrance.

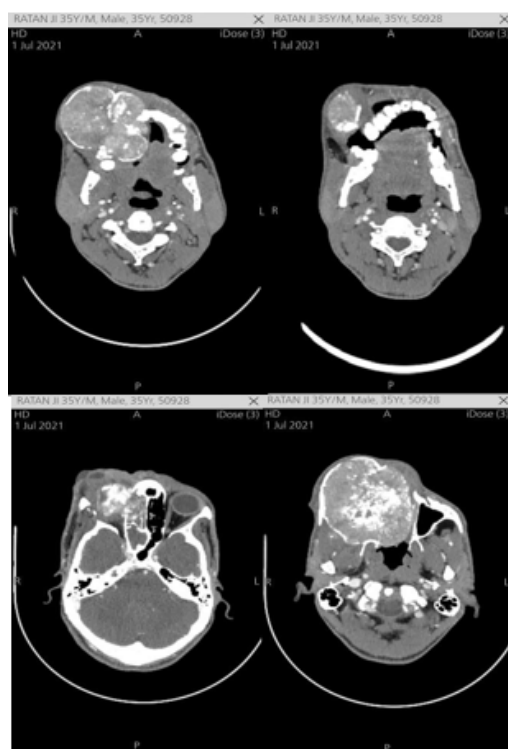


Fig. 3: Selected axial section for analysis.

At the moment the patient is with a year and a half of postoperative follow-up without clinical signs and satisfactory aesthetic result.

5. Discussion

The fibrous dysplasia is a uncommon disease, congenital, benign, characterized by defective bone modeling, with gradual replacement of normal bone by fibrosis with irregularly mineralized osteoids. Some believe to be a congenital anomaly of mesenchymal tissue development.¹⁷

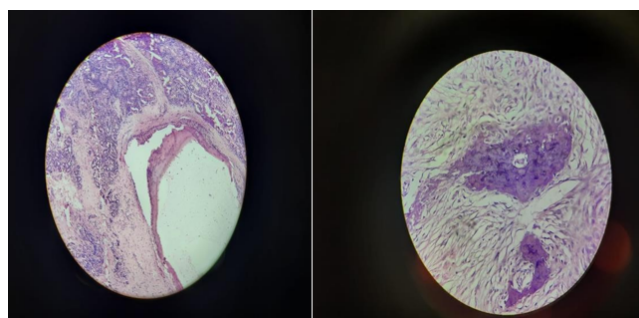


Fig. 4: Anatomopathological.

The fibrous dysplasia is a pathology responsible for about 2.5% of bone tumours, and more than 7% of all non-malignant bone tumours.¹⁸ In yours variety forms, affects one in every 4,000 to 10,000 individuals, don't having



Fig. 5:

relation with hereditary factor.¹

Approximately 70% of the cases are manifested in the first decade of life, with slow growth, stabilizes at puberty, along with the skeleton. It's more frequent in females, recurrent in about 37% of the cases in adults.¹⁷

Fibrous dysplasia remained with your indefinite etiology, with authors,^{19,20} describing embryological, neurological and congenital theories to explain their etiology. We now know that fibrous dysplasia is a sporadic condition that results from a post zygomatic mutation in the GNAS1 gene (protein bound to the guanine nucleotide α -stimulating activity of polypeptide1). We can affirm that fibrous dysplasia is a pathology that causes a disorganization in the bone tissue, generating an exaggerated and disorganized growth of this disease.

It's classified according to the number of bones affected and the presence or not of extra-skeletal abnormalities. The monostotic form affects only one bone and corresponds to 70-80% of FD cases. The polyostotic form, in which multiple bones are affected, can be divided in three subtypes: Craniofacial, in which only bones of the craniofacial are involved including the mandible and maxilla; Lichtenstein-Jaffe, in which besides the involvement of multiple bones of the skeleton are brown-to-brown pigmentations in the skin; Albright syndrome, characterized by the involvement of several bones, brown-to-brown pigmentation on the skin and endocrine impairment, with prominence for early puberty in girls. The polyostotic form responds for 20-30% of the cases.²¹ Referring to tomographic examinations of the presented case, it was evidenced compromise of the palatine bones, maxilla, zygomatic complex right side, sphenoid, as well as region of nasal cavity.

Initially the disease is asymptomatic. The signs and symptoms in FD are dependent on the location of the lesion(s) and the compressive effect on neighbouring structures as the tumour progresses slowly; Asymmetry and facial deformity; Pathological fractures; Obstruction of the paranasal sinuses generating recurrent infections, cysts and mucocoeles; Anosmia, headache, loss of visual acuity

by compression of the optic nerve; Alteration of ocular movements, ptosis, exophthalmos, strabismus; Conductive hearing loss^{18,22–24}.

The clinical signs and symptoms observed in this case were also found by several authors,^{13,14,20,25–27} such as: Volume increase in the region, functional impairment, facial asymmetry, slow and asymptomatic evolution, mucosa adjacent to lesion without changes, hard consistency on palpation, normal-appearing mucosa with smooth surface and no ulcerations. The main symptoms reported by patients, when present, are: Throbbing or discomfort, some heat at the local of injury, diplopia, decreased visual acuity and may even lead to vision loss, deafness, nasal obstruction and dizziness^{28,28}. Another very common feature of facial asymmetry is leontiasis ossea, caused by the expansion of maxilla, with loss of the nasomaxillary angle, generating a feline facial appearance. In the report, the patient only complained about localized asymmetry, especially in the zygomatic-maxillary region.

CT is the test of choice for the study of lesion(s), avaluation of its extension and surgical preparation.^{23,29} Basically, three radiographic patterns in the fibrous dysplasia of the skull and facial bones are described: Pategoid, alternating radiodense and radiotransparent areas; Sclerotic, homogeneously dense; and cystic pattern with spherical or ovoid radiolucent area surrounded by dense boundaries. In the case reported, tomographic images of the lesion assumed a hyperdense pattern interspersed with hypodense areas of imprecise limits, resulting in the classic aspect of “ground glass”. The definitive diagnosis of FD is made by the correlation of clinical, radiological and anatomopathological findings. It is of great importance that computed tomography should always be used before the surgical procedure to evaluate the actual size of the pathology, anatomical structures and their compromise, and mainly, to guide in the planning of the extent of osteotomy and osteoplasty that will be performed. Besides that, can be divided into three patterns of involvement: the sclera, the cystic or the mixed.

The differential diagnosis includes benign lesions: Solitary unilocular cyst, non-ossifying fibroma, eosinophilic granuloma, cholesteatoma, meningioma, Paget’s disease, osteochondroma, ossifying fibroma, giant cell reparative granuloma, exostoses, aneurysmal bone cyst, cystic fibrous osteitis; and malignancies, such as: Sarcoma and metastatic osteoblastic lesions. The clinical aspects, in the case reported, along with the laboratory and imaging findings, were fibrosis dysplasia, but it is indispensable to perform histopathological examination to confirm the diagnosis.

Treatment is still debatable, especially in relation to lesions in the middle third of the face, which are difficult to access, when, after surgical intervention, esthetic and functional deficits can be found. Therefore, the best choice for approach in this region is through partial resection

or bone remodeling. Aggressive surgeries are reserved for other cases in which vision or dentition is impaired. The patient of the exposed clinical case had an important impairment of the right third of the face on the right side, but without nasal obstruction or malocclusion and with preserved visual acuity. The proposed surgery was to perform an intraoral access in the upper vestibule, with aesthetic purpose, and the high rate of recurrence of the lesion. The option was made for facial remodeling through osteoplasty, agreeing with the authors previously mentioned.

At anatomopathological examination, two forms can be distinguished macroscopically: The compact form, characterized by the presence of osteoid tissue, which progressively ossifies and assumes the appearance of mature bone and the cystic form, which presents one or more cavities surrounded by the changes described above.

Microscopically, the appearance of these two forms is unique. There is proliferation of fibrous tissue, with progressive ossification and destruction of the affected bone.

A specimen was sent for histopathological examination for microscopic findings of the lesions resemble those described in the literature and were: Young trabeculae interspersed by fibrous conjunctive tissue compatible with diagnosis of fibrous dysplasia.

Radiotherapy is contraindicated due to the possibility of malignant transformation of the post-irradiation lesion, chemotherapy is also contraindicated, since it is not effective in curing the disease.

Some authors defend the need for a clinical, endocrinological and scintigraphic study with computed tomography in patients diagnosed with fibrous dysplasia in search of other lesions in other sites and extra-skeletal involvement.

Clinical and radiological follow-up by CT is fundamental in patients with Fibrous dysplasia due to the moderate rate of recurrence of the lesion(s), and may reach 37% according to some authors.

Huge fibrous dysplasia of rt maxilla not only causes facial asymmetry but also produces significant loss of vision [6/24 in our cases] that is significantly improvement of vision [6/9 in our cases] 4 to 5 days after removal of tumor.

Facing the clinical case, FD is considered a pathology, which may present functional and aesthetic impairment. In the moment of decision to treat fibrous dysplasia, consideration should be given to the patient’s age, presence or absence of facial asymmetry, facial involvement and future rehabilitation. Because it is a tumour with no precise and relapsing limits, it is important to remove as much tissue as possible without causing mutilations to the patient, functional deficits or lesions of noble structures. Surgical treatment is indicated in case of significant deformity, significant pain or pathological fracture. Radiation therapy

Table 1:

S.No.	Examination	Preoperative	Postoperative
1	Swelling	Huge swelling	No swelling
2	Nose	Rt nasal obstruction	No obstruction
3	Ephiphora	Present in rt eye	Abscent
4	Eye	Turned upward & laterally [rt]	Normal position
5	Eye lids	Swelling in lower eye lids[rt]	Normal
6	Conjunctiva	Chemosed[rt]	Normal
7	Cornea	Hazy[rt]	Normal
8	Vision	6/24	6/9
9	Fundus		
10	Tonometry	Iop-34 mm of hg in rt eye, 21 mm of hg in left eye	Rt 20 mm of hg Lt 21 mm of hg

is contraindicated due to the high risk of sarcomatous transformation. The follow-up is of fundamental importance (time) in order to detect relapses or a possible, malignant change at an early stage.

6. Source of Funding

None.

7. Conflict of Interest

None.

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