# **Original Research Article**

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# Fibro-osseous lesions of the paranasal sinuses and the skull base

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## **ABSTRACT**

**Background:** Osteomas, ossifying fibromas, and fibrous dysplasia comprise a set of benign neoplasms known as fibro-osseous lesions (FO), which can arise in the paranasal sinuses. The vast majority of benign FO lesions (BFOL) are incidental findings on radiographs. They tend to be slow-growing tumors, and are infrequently symptomatic. The management strategy can vary significantly for theses lesions. This can range from serial, observations to aggressive surgical resection, based on symptomatology, size, and location of the lesion.

**Methods:** Retrospective analysis of all the data of different symptomatic FO of the paranasal sinuses and the skull base was performed at King Fahad specialist hospital, Dammam, KSA (2006 to 2017).

**Results:** A total of 21 patients were identified; 10 (46.7%) patients were diagnosed with osteoma, 8 (38.1%) with fibrous dysplasia, 2 (9.5%) with ossifying fibroma and one (4.8%) with juvenile active ossifying (JAOF), 17 were adults and 4 were children, the range of f/u (3-60) months. 19 patients were managed by the endonasal endoscopic approach. They continue to be followed with no evidence of recurrence. Two cases required the revision combined endoscopic open approach to ensure adequate removal of the disease (residual and recurrence).

**Conclusions:** Diagnostic dilemma of BFOL can be overcome with a combination of clinical, radiological and pathological criteria. Correct diagnosis of fibro-osseous tumors is crucial for adequate therapy as their treatment, prognosis, clinical aggressiveness and long term complications of individual entities vary significantly. Endoscopic computer-assisted surgery is the treatment of choice. Endoscopic resection remains a technical challenge.

**Keywords:** Fibro-osseous lesion, Osteoma, Fibrous dysplasia, Ossifying fibrom

## INTRODUCTION

Osteomas, ossifying fibromas, and fibrous dysplasia comprise a set of benign neoplasms known as benign fibro-osseous lesions (BFOL) that, and can arise in the paranasal sinuses. Osteomas, are the most common benign tumors occurring in the paranasal sinuses. They arise mostly at the frontoethmoid region. There are three histologic types of osteomas: 1) an ivory type composed of dense cortical bone, 2) a spongy form composed of cancellous bone, and 3) a mixed type that contains

features of both.<sup>1-3</sup> On the other hand, fibrous dysplasia and ossifying fibroma (OF) are less common lesions.<sup>1</sup>

The vast majority of fibro-osseous lesions are incidental findings on radiographs. <sup>1</sup> They are slow-growing tumors, and are infrequently symptomatic. <sup>1</sup> frontal headache is the most common complaint, followed by drainage and nasal obstruction associated with secondary sinusitis. however, large lesion may extend intracranially and produce pneumocephalus, meningitis, cerebrospinal fluid leak, or mental status and neurologic changes. A tumor

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with orbital extension can lead to proptosis, exophthalmos, or diplopia, and reduced vision. 1

The management strategy can vary significantly for these lesions. This can range from serial, observations to aggressive surgical resection, depend on symptomatology, size, and location of the lesion.<sup>4</sup>

In general, osteomas may be observed by serial imaging, unless symptomatic, characterized by rapid growth, associated with mucocele formation, or compression of vital orbital or intracranial structures.<sup>4</sup>

Treatment for fibrous dysplasia includes observation with serial imaging, with surgical intervention being reserved for cases resulting in cosmetic deformity, paresthesia, trismus, or optic nerve involvement.<sup>4</sup>

The aim of this study is to present our experience in the management of symptomatic fibro-osseous lesions of the paranasal sinuses.

## **METHODS**

A retrospective cohort study was done over the period of two months (June-July, 2018) to include all different cases of fibrosseous lesions managed at King Fahad specialist tertiary care hospital, Dammam (KFSHD), Saudi Arabia throughout the period 2006-2017.

## Inclusion criteria

Inclusion criteria were cases with symptomatic fibrosseous lesions.

# Exclusion criteria

Exclusion criteria were asymptomatic cases with incidental radiological findings

A data collection sheet was prepared specially for this study. It included information regarding patient demographic characteristics, location of the lesion at presentation, CT scan findings, management approach, details of surgery performed, and outcomes. Research

proposal was approved by the regional Research and Ethics committee at KFSHD. Statistical analysis was performed using the Statistical Package for Social Science (SPSS), version 23. Categorical variables were described in the form of frequency and percentage while contiguous variables were presented as mean, standard deviation and range.

## **RESULTS**

Twenty-one patients with symptomatic fibro-osseous lesions were identified for the study. Their demographics and findings are summarized in (Table 1). The diagnosed was osteoma in 10 patients (47.6%), fibrous dysplasia (FD) in 8 patients (38.1%), and OF in 2 patients (9.5%) and one patient with juvenile active OF (4.8%). Seventeen patients were adults (80.5%) and 4 were children (19.5%). The age ranged from 3 to 40 years. Mean age±Standard deviation was 23.7±9.2 years. The male to female ratio was 1.6:1.

The range of follow up was from 6 to 60 months. Two cases required revision combined approach (endoscopic and open) surgery to ensure adequate removal of the disease which represents only (9.5%). However, 19 cases (90.5%) managed completely via the endonasl endoscopic approach with no recurrence along the follow-up period.

#### **Osteomas**

Ten patients presented to our hospital for management of symptomatic paranasal sinus osteomas (Table 2 and 3). The most common symptoms at presentation were frontal headaches followed by nasal obstruction. The osteomas involved the frontal sinuses in 4 cases (40%), sphenoid sinuses in 2 cases (20%), frontoethmoid in 2 cases (20%), one case involved the ethmoid sinus (10%) and one case for a patient known to have osteopetrosis with extensive craniofacial osteoma (10%). The male to female ratio was 4: 1. All patients with osteoma were managed via the endonasl endoscopic approach for removal. No reported surgical complications and none of the patients had orbital injury or CSF leak. The patients continued to be followed-up with no evidence of recurrence of their osteomas (Figure 1).

Table 1: Demographic characteristics of patients (N=21).

	Osteoma	FD	OF	JAOF
Number of patient (%)	10 (47.6)	8 (38.1)	2 (9.5)	1 (4.8)
Gender				
Male (%)	8 (80.0)	4 (50.0)	1 (50.0)	10 (0.0)
Female (%)	2 (20.0)	4 50.0)	1 (50.0)	1 (100)
Age (years)				
Mean	25.4	23.3	21.0	
SD	9.7	10.3	1.4	15
Range	3-40	5-37	20-22	

F: Female; M: Male; FD: Fibrous dysplasia; OF: Ossifying fibroma; JAOF: Juvenile active ossifying fibroma; SD: Standard deviation.

Table 2: Clinical characteristics of patients with osteomas.

	Frequency	Percentage (%)
Lesion site		
Sphenoid	2	20.0
Ethmoid	1	10.0
Frontal	3	30.0
Fronto-ethmoid	2	20.0
Craniofacial	1	10.0
Frontal and skull base	1	1.0
Procedure		
ESS	9	90.0
Debulking	1	10.0
Complications		•
Yes	0	0.0
No	10	100
Outcome		
No recurrence, improve	10	100
Recurrence	0	0.0

ESS: Endoscopic sinus surgery.

Table 3: Summary of data for patients with fibrous dysplasia/ossifying fibroma/JAOF.

	FD N=8 N (%)	OF N=2 N (%)	JAOF N=1 N (%)	
Lesion site	-			
Sphenoid	2 (25.0)			
Maxillary	1 (12.5)			
Fronto-ethmoid and skull base	2 (25.0)			
Fronto-ethmoid	2 (25.0)			
Craniofacial	1 (12.5)			
Ethmoid, orbit, skull base		1 (50.0)	1 (100)	
Ethmoid, orbit, middle turbinate		1 (50.0)		
Procedure	-	-		
FESS	8 (100)	1 (50.0)		
Fess+lateralrhinotomy +craniotomy	0 (0.0)	1 (50.0)		
Fess, craniotomy			1 (100)	
Complications				
Yes, temporary				
diplopia,	0(0.0)	1 (50.0)		
enophthalmos				
No	8 (100)	1 (50.0)	1 (100)	
Outcome				
No recurrence	8 (100)	1 (50.0		
Recurrence	0 (0.0)	1 (50.0)	1 (50.0)	

FD: Fibrous dysplasia; OF: Ossifying fibroma; JAOF: Juvenile active ossifying fibroma; FESS: Functional endoscopic sinus surgery.



Figure 1: CT sinuses showing right homogenous hyperdense mass (osteoma) involving right nasal cavity, middle turbinate and ethmoid sinus.

# Fibrous dysplasia (FD)

FD involved more than one sinus in 4 cases (50%), the sphenoid sinus in 2 cases (25%), skull base in 2 cases (25%), and maxillary sinus in one case (12.5%). The common symptoms were facial pressure and pains. Male to female ratio was 3:5. Seven cases (87.5%) were managed by the endonasal endoscopic approach and one pediatric case (12.5%) was managed by the navigation assisted endoscopic approach. None of the cases required an open surgical approach. The follow up ranges from 12 to 60 months (Figure 2).



Figure 2: Axial CT scan showing the ground glass appearance of fibrous dysplasia.

## Ossifying fibroma/JAOF

Two cases (66.7%) were diagnosed with OFa involving the ethmoid and skull base and one case (33.3%) of juvenile active ossifying fibroma (JAOF). The female to male ratio was 2:1. The clinical presentations were nasal obstruction, headache, facial pressure, proptosis and recurrent sinusitis. The follow up ranges from 24 to 60 months. All cases were managed initially with debulking via the endonasal endoscopic approach; two cases (66.6%) had recurrence of the disease and managed by an open surgical approach.

We had a patient with an ethmoidal of causing progressive right proptosis, severe pain and nasal obstruction over 3 months (Figures 3 a) that was resected via an endoscopic sinus surgery (ESS)/debulking. Most of the lesion was resected; the histopathology confirmed the diagnosis of OF. One year later, the patient presented with clinical and radiological recurrence which was managed by the neurosurgery team with Frontal Craniotomy and skull base reconstruction. The surgery was complicated by temporary diplopia, mild enophthalmos. The patient continued follow on both otolaryngology and neurosurgery clinics with no evidence of recurrence clinically nor radiologically (Figure 3b), diplopia resolved and vision improved.

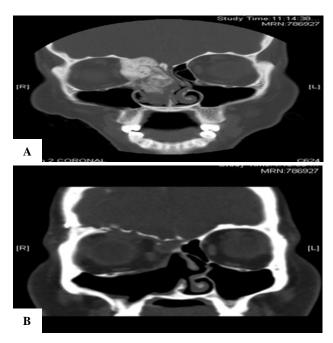


Figure 3: (A) Coronal CT-scan showing a heterogeneous lesion involving right nasal cavity and right ethmoid sinus pushing septum to left side encroaching the orbit and skull base; (B): postoperative CT sinuses with clear sinuses, orbit and skull base.

We had a child with progressive right nasal obstruction, hyposmia and right proptosis for 10 months duration. No visual disturbances, diplopia or nasal discharge, on examination a large fleshy mass filling the right nasal cavity pushing the septum to the other side and right eye proptosis CT scan (Figure 4a). The tumor was resected via the navigation assisted endonasal endoscopic approach and histopathology confirmed the diagnosis of JAOF (Figures 4 b, 4c). On follow up, the patient started to complain of right sided headache and hyposmia with no nasal obstruction. The patient had an extensive skull base recurrence after 6 months and underwent an open craniotomy for tumor removal and skull base reconstruction, with no complication and No evidence of recurrence endoscopically nor radiologically for 3 years postoperatively.

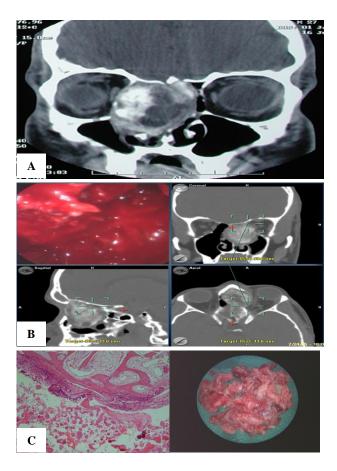


Figure 4: (A): CT sinuses showing a heterogamous expansible lesion involving right ethmoid pushing septum to left side and abutting the right eye; (B): endoscopic navigation assisted removal from the orbit and skull base; (C): Gross and histopathological diagnosis of juvenile ossifying fibroma which are composed of cellular stroma of spindled to stellate like cell, bony trabecular with osteoblastic rimming.

# **DISCUSSION**

This report presents our experience in the management of symptomatic fibro-osseous lesions of the paranasal sinuses and the skull base over a period of 11 years. The outcome was favorable with the majority of patients' symptoms improved after surgical treatment. The endonasal endoscopic approach with and without navigation was the main approach performed to confirm the diagnosis and to remove the tumors from the sinuses and the skull.

## Osteoma

The majority of osteomas in our study arise from the frontal sinus, which is similar to other reported series. There is a male predominance in most series. In general, osteoma of the paranasal sinuses are asymptomatic and discovered incidentally during radiographic evaluation for unrelated problems. Symptoms related to osteoma generally arise from a "mass effect" as the lesion impinges on normal structures. Extra sinus complications

of osteoma can be either orbital or intracranial. Osteomas may expand slowly into the orbital vault, displacing the orbital contents. This may lead to diplopia, epiphora, facial distortion, and even blindness. <sup>6,7</sup> Intracranial complications occur when an osteoma penetrates the dura resulting in mucoceles, meningitis, frontal abscess, cerebrospinal fluid (CSF) leak, or pneumatocele. <sup>6,8-10</sup> On computed tomography (CT) scan, it appears as a very dense, homogeneous, well-circumscribed mass attached with an apparent range of broad to narrow pedicle to an adjacent bone. The surrounding bone is normal and does not have a lytic or moth-eaten appearance. <sup>6,11,12</sup>

Where there is any question of extra sinus extension or intracranial involvement, MRI is recommended. <sup>6,13,14</sup>

Histologically, there are three types of osteomas. The eburnated type, also known as the ivory or compact type, which is very dense and lacks haversian canals. It may arise from membranous elements. 6,12,15 The second is the mature type, or osteomaspongiosum, which is composed of softer bone that is more similar to cancellous bone. It is thought to arise from cartilaginous elements. 6,11 The last type is the mixed type of osteoma which contains elements of both the eburnated and mature forms.6 The primary dilemma in patients with osteoma is determining if surgical removal is indicated. The management of asymptomatic incidental osteomas is conservative, with periodic imaging to follow the growth, and intervention is reserved for cases of progressive growth prior to the development of complications. 4,16 Initially, a 3 to 6 month follow-up and an annual followup thereafter are recommended if no growth is detectable on CT and the patient remains asymptomatic. <sup>6</sup> Rapid growths, infection, compression of vital structures, severe pain, and facial deformity are indications for surgical resection.4,6,17

Multiple surgical approaches have been described for removal of this neoplasm from the paranasal sinus including, endonasal endoscopic, external, or combined. These approaches were done according to the size and location of the tumor. Regardless of the technique chosen, the primary goal is complete resection with minimal trauma to the sinonasal mucosa and the surrounding normal structures. If untreated, osteomas will continue to grow slowly during the life of the patient, but they are always benign with no reported malignant degeneration or metastasis. Recurrence after complete resection is extremely rare, as observed also in our series of cases. Recurrence after

## Fibrous dysplasia (FD)

It is an abnormal proliferation and differentiation of bone marrow stromal cells, producing excessive fibrous tissue within the bone marrow.<sup>4</sup> The classification system subdivide the disease into three types: type 1, is monostotic and characterized by unique or multiple lesions in a single bone, type 2, is polyostotic and,

characterized by multiple lesions involving different bones and type 3, is characterized by dissemination of pathological bone changes connected to other disturbances such as McCune–Albright syndrome (polyostotic fibrous dysplasia, cutaneous pigmentation, and endocrine abnormalities).<sup>20</sup>

Monostotic fibrous dysplasia is the most common form (80%), 20% of cases are localized in the head and neck region. <sup>20,21</sup> Multiple areas of the skeleton are involved in polyostotic fibrous dysplasia, which accounts for 20%, craniofacial involvement 50% to 100% of the cases. <sup>20,22</sup>

In accordance with findings of other studies, the frontoethmoid and sphenoid sinus were the most common areas involved with FD in the present study. We had 3 cases of extensive FD which involved more than one sinus. The most common clinical presentation of fibrous dysplasia in the head and neck area is facial asymmetry, followed by ocular symptoms, headache, and hearing loss. <sup>20,23,24</sup>

FD has a low rate of malignant transformation which, occurs in 0.5% of polyostotic forms and in 4% of lesions in patients with McCune-Albright syndrome. Most reported cases of malignant degeneration of FD have occurred after radiation therapy. Sa.25,26 Osteosarcoma is the most common histologic type of malignant transformation, followed by fibrosarcoma, chondrosarcoma, and malignant fibrohistiocytoma. These malignancies are most commonly found in the maxilla and mandible, with rare involvement of the calvarium.

Diagnosis can be accomplished via radiographic appearance, although some investigators recommend biopsy of FD lesions.<sup>5,6</sup> On CT scan, the FD lesions are characterized by hazy borders and a fairly homogeneous "ground glass" appearance representing the disorganized spicules of bone that characterize FD's histology. As the lesion scleroses, "cotton wool" areas arise.<sup>6</sup>

On MRI, FD lesion appears intermediately in T1 and hypo intense in T2. Conservative management with serial scans and observation for FD patients with minimal symptoms or mild cosmetic deformity is generally recommended.<sup>4</sup> Surgery is indicated in cases of significant cosmetic deformity or complications such as visual compromise.<sup>2</sup> This is usually accomplished via an endonasal endoscopic debulking technique. Radical or complete resection is not usually necessary. Patients should be followed up with periodic imaging to guide management of any regrowth.<sup>6</sup>

# Ossifying fibroma (OF)

It is the most concerning of the fibro-osseous lesions. It is known as cemento-ossifying fibroma, psammomatoid, OF, and JAOF. OF have been more frequently observed in the third and fourth decades of life and has a higher incidence in the black population. <sup>29-31</sup> In our series, we had two cases aged 20 and 22 years.

The molar and premolar periapical regions of the mandible and maxilla are the most frequently involved sites by OF.<sup>29</sup> There are two main histological variants of the classic OF; (trabecular) and cementiform or psammomatoid. The latter is characterized by round calcific masses resembling psammomatoid bodies of meningiomas.<sup>29, 32</sup>

A clinical variant of OF, called juvenile-aggressive ossifying fibroma, is more frequently observed in the sinonasal tract, where it predominantly affects male subjects. <sup>29,33,34</sup> It usually belongs to the cementiform type and is considered to have an aggressive biological behavior mimicking a malignant neoplasm. <sup>29,35</sup> Even though painless facial and skull deformities are the most frequently observed signs, symptoms such as nasal obstruction, headache, epistaxis, anosmia, loosening of teeth, facial paralysis, hearing loss, trigeminal neuralgialike pain, and recurrent rhinosinusitis due to drainage impairment may develop. <sup>29</sup> Radiographic ally, it appears on CT scan as a sharply circumscribed round or oval lesion with an eggshell rim and a central radiolucency. <sup>5,6</sup>

OF requires radical excision, which can be obtained by the endonasal endoscopic approach or if large lesion by open anterior craniofacial resection. Aggressive surgery is justified by the high percentage of recurrences which has been estimated to range between 30% and 44%. <sup>29,37</sup> In our series, we had one case with recurrent course after surgery.

## **CONCLUSION**

Diagnostic dilemma of benign fibro-osseous lesions (BFOL) of the paranasal sinuses and skull base can be resolved with a combination of clinical, radiological and pathological criteria. Correct diagnosis of fibroosseous tumors is crucial for adequate therapy as their treatment, prognosis, clinical aggressiveness and outcomes of individual entities vary significantly. If indicated, resection via the endonasal endoscopic navigation-assisted approach is the treatment of choice. Long follow up is recommended to detect and manage early recurrence or malignant transformation.

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Ethical approval: The study was approved by the

Institutional Ethics Committee

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