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Case Series

Management of juvenile ossifying fibroma: insights from three cases and a review of literature

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ABSTRACT

Juvenile ossifying fibroma (JOF) is a rare, benign fibro-osseous neoplasm exhibiting locally aggressive behaviour predominantly localized within the craniofacial bones. This distinct entity is distinguished from ossifying fibroma (OF) by its earlier age of onset and an increased propensity for recurrence. A comprehensive review of existing literature, supplemented by a case series, is presented to elucidate the clinical, radiological, and histopathological characteristics of JOF. Diagnostic challenges associated with JOF are often encountered due to its variable presentation and potential overlap with other fibro-osseous lesions. Treatment is by surgical resection, with a focus on obtaining total excision to reduce the chance of recurrence. Long-term follow-up is crucial for early detection of recurrence. This combined literature review and case series presentation aims to improve knowledge of JOF, enabling more accurate diagnosis and effective therapy.

Keywords: Juvenile ossifying fibroma, Fibrous dysplasia, Sinonasal bony lesions

INTRODUCTION

Juvenile ossifying fibroma (JOF) is a rare, benign, locally aggressive fibro-osseous neoplasm primarily affecting the craniofacial bones.[1] It has distinct clinical behaviour characterized by rapid growth and a higher recurrence rate. JOF predominantly occurs in children and young adults. JOF is histologically categorized into trabecular (JTOF) and psammomatoid (JPOF) subtypes. This classification cannot accurately predict the biological behaviour or prognosis.[2] The clinical presentation of JOF varies, often manifesting as a painless swelling in the maxilla or mandible. However, depending on the location and extent of the lesion, it can lead to significant facial deformity and functional impairment.[1] Accurate diagnosis requires clinical, radiological, and histopathological evaluation.[1,3]

Imaging studies, including computed tomography (CT) and magnetic resonance imaging (MRI), play a crucial role in determining the extent of the lesion, its relationship to surrounding vital structures, and surgical planning.[4]

The optimal management of JOF is by complete surgical excision.[5] However, the extent of resection is based upon factors such as lesion size, location, and accessibility. So, the resection is individualized, from enucleation to radical excision. The high recurrence rate necessitates long-term follow-up to ensure early detection and prompt management of recurrent disease.[6]

This case series highlights the clinical presentations and management approaches employed. Furthermore, a comprehensive review of current literature is provided to expand existing knowledge on JOF and offer insights into its diagnosis, treatment, and prognosis. This combined approach of case series and literature review aims to enhance understanding of this rare entity and contribute to improved clinical practice.

CASE SERIES

Case 1

A 10-year-old boy presented to ENT OPD of PGIMS Rohtak with complaints of right eye protrusion and heaviness of right face that had progressed slowly for six months (Figure 1). He had a history of facial trauma one year ago by bicycle handle. Ophthalmologic examination was normal except right eye protrusion. His right eye movements and eye closure were normal. On diagnostic nasal endoscopy, a bony hard globular mass was present in the right middle meatus with normal overlying mucosa, pushing middle turbinate medially and causing buckling of septum towards the left side. Oral cavity and oropharynx examination was normal.

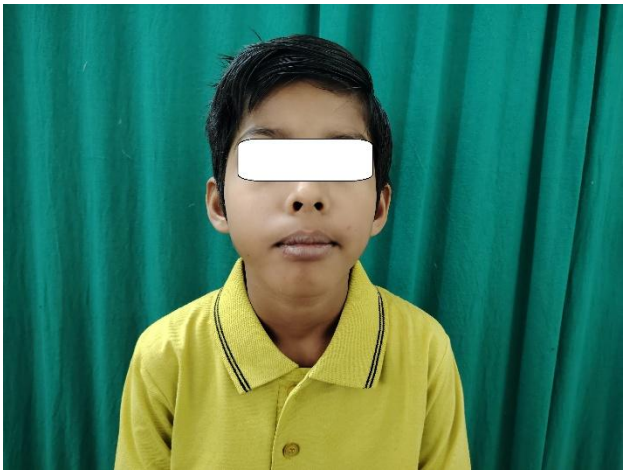

Figure 1: Right facial swelling and right eye protrusion.

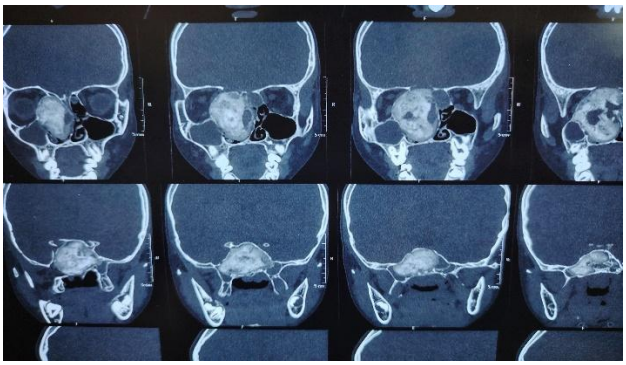

Figure 2: Black arrow showing bony lesion with ground glass matrix in the right sinonasal cavity, obliterating right nasal cavity.

Non-contrast computed tomography (NCCT) demonstrated an expansile bony lesion with ground glass matrix in the right sinonasal cavity, obliterating the nasal cavity and buckling nasal septum towards the left side (Figure 2). Magnetic resonance imaging (MRI) showed marked bulging of right lamina papyracea into orbit by fibro-osseous lesion and displacement of the right medial rectus and posterior intraorbital segment of the right optic nerve. Superiorly, it caused a bulging of the planum sphenoidal, with thinning of the cortex and abutting of the right internal carotid artery (Figure 3). No intraorbital lesion was noted. Duramater was grossly maintained.

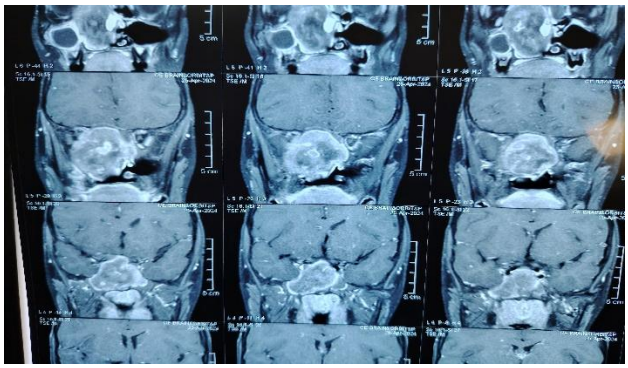

Figure 3: Black arrow showing fibro-osseous lesion abutting the right internal carotid artery.

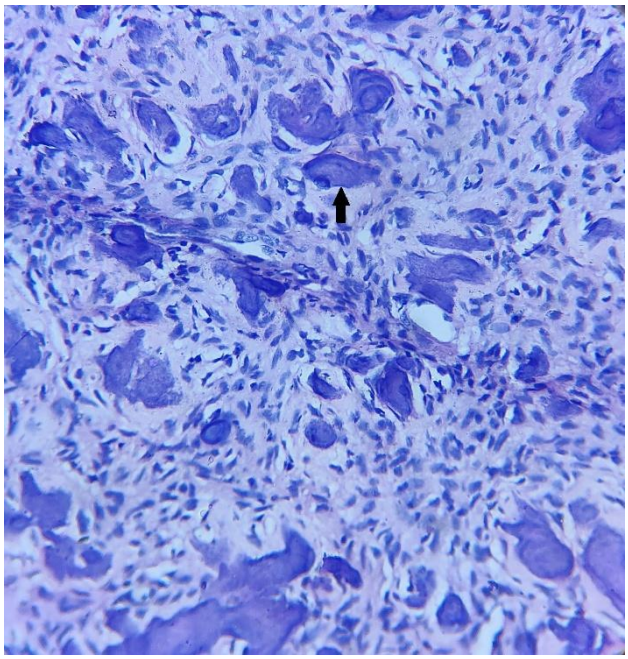

Figure 4: Microsection showing variable sized cementum-like ossicles embedded in fibrous stroma (black arrow) (H&E; 40X).

An incomplete surgical resection was performed by sublabial midfacial degloving approach with medial maxillectomy because of the proximity of lesion to cavernous part of internal carotid artery. The fibro-osseous lesion abutting the internal carotid artery was not removed. Excised specimen was sent for histopathological examination (HPE) and was reported as JOF (Figure 4). Patient had decreased protrusion of right eye in immediate postoperative period. Patient was followed up regularly. Repeat CT was performed after six months. It showed the recurrence of the lesion (Figure 5).

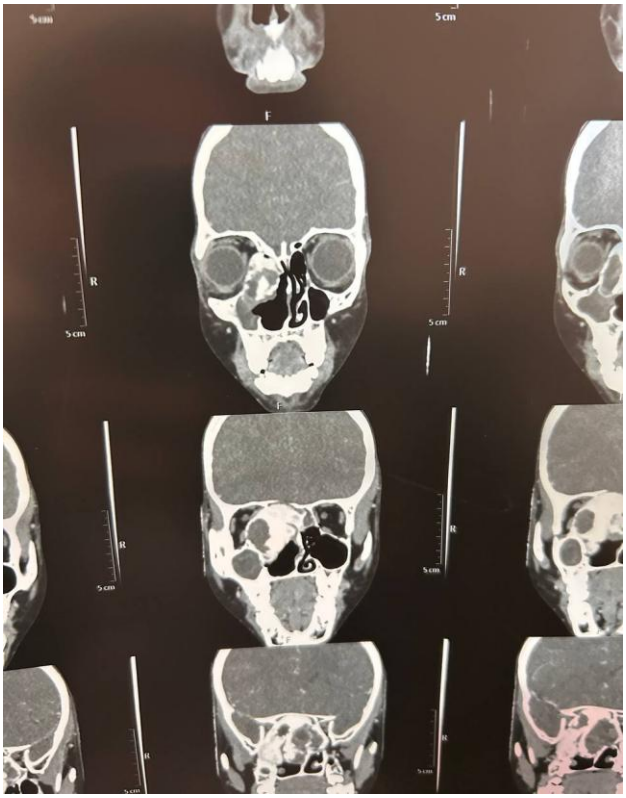

Figure 5: Black arrow showing recurrence of the fibro-osseous lesion with post op changes.

Case 2

A 12-year-old girl presented to ENT OPD of PGIMS Rohtak with left nasal obstruction and left facial asymmetry that had progressed slowly for one year (Figure 6). She had associated anosmia. She had a history of trauma six months ago. On nasal examination, a bony hard globular mass was present in the left nasal cavity reaching up to the vestibule. Ocular examination was normal with normal visual acuity and normal ocular mobility. Oral cavity and oropharynx examination was normal.

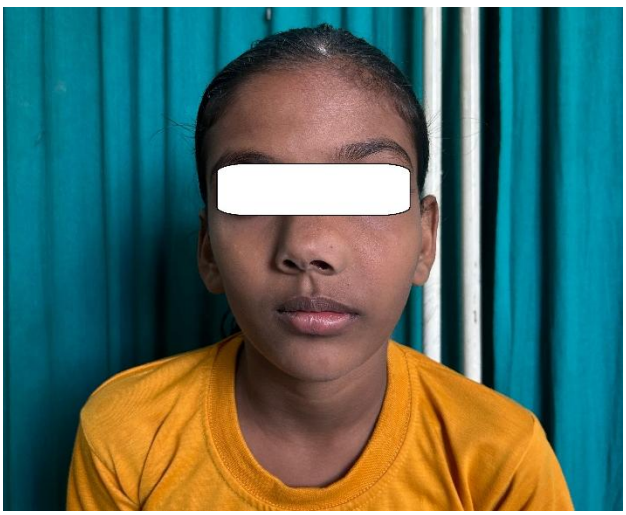

Figure 6: Left facial swelling.

CT demonstrated a large expansile homogenous mass centered around left ethmoidal air cells with focal erosion of sinus walls extending into left nasal cavity with posterior extension in choana and nasopharynx.

A complete surgical excision was achieved by left medial maxillectomy via sublabial midfacial degloving approach. HPE confirmed the diagnosis as JOF. Immediately postoperative, facial swelling was reduced (Figure 7). The patient was regularly followed up to look for recurrence for two years. To date, there are no clinical or radiological signs of recurrence of disease.

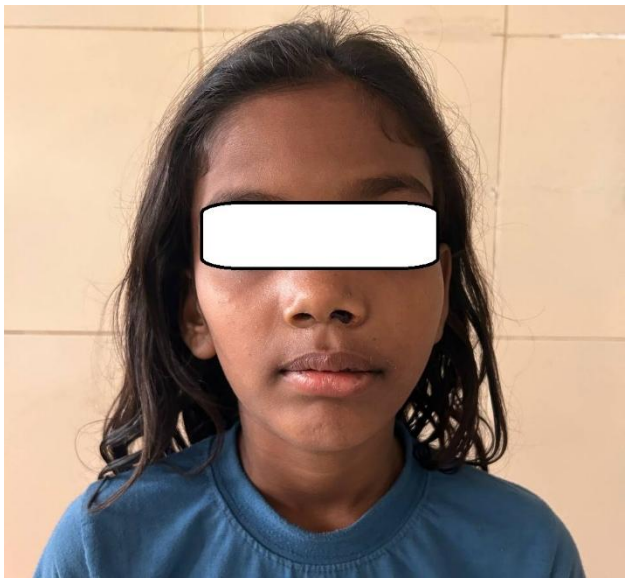

Figure 7: Post-operative picture showing decreased facial swelling.

Case 3

A 16-year-old female patient presented to ENT OPD of PGIMS Rohtak with left facial swelling and left nasal obstruction for 10 months (Figure 8). She had a history of trauma to face two years ago. The facial swelling had a bony, hard consistency. On nasal examination, there was a hard globular mass in the left nasal cavity and nasal septum was grossly deviated towards the right side. Ocular examination was normal with normal visual acuity.

CT demonstrated a fibro-osseous lesion with ground glass matrix centered around anterior wall of left maxillary sinus. The bony mass was extending medially into left nasal cavity pushing the nasal septum towards opposite side. Ethmoid air cells showed mucosal thickening due to retained secretions. Left maxillary mass excision was planned via midfacial degloving approach with medial maxillectomy (Figures 9). The HPE of the excised mass showed OF. Immediately after the operation, left facial swelling was significantly reduced (Figure 10). The patient was followed up at regular interval to look for recurrence. At six-month post op, there were no signs of recurrence of the disease.

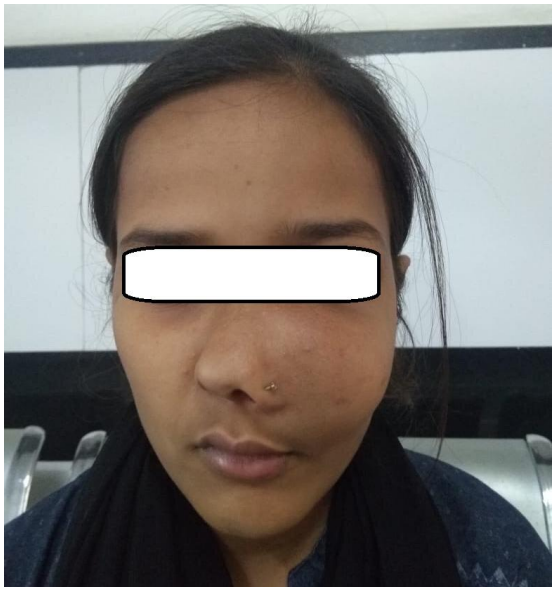

Figure 8: Left facial swelling.

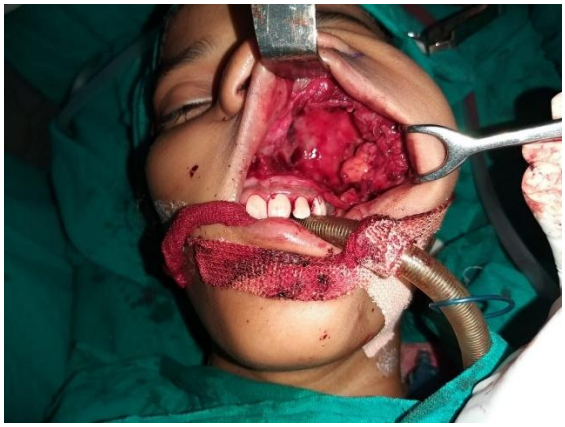

Figure 9: Intraoperative picture showing midfacial degloving via sublabial incision.

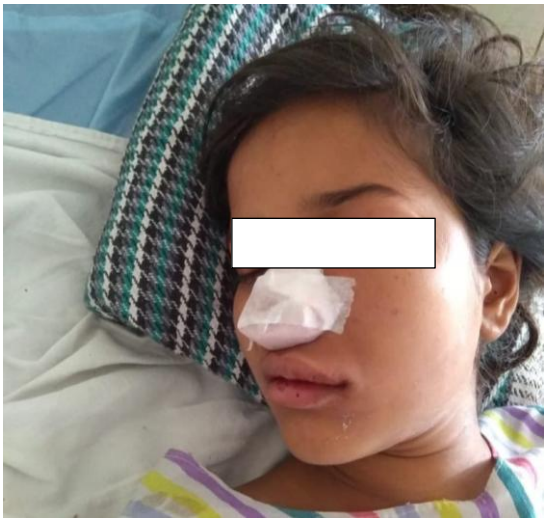

Figure 10: Post-operative picture showing decreased facial swelling.

DISCUSSION

JOF is a rare, benign but locally aggressive fibro-osseous neoplasm that primarily affects the craniofacial bones of children and young adults.[7-9] While histologically similar to ossifying fibroma, JOF distinguishes itself through its earlier onset, more aggressive behavior, and higher recurrence rate.[10] It typically arises in the maxilla and mandible, with 79% of cases diagnosed before the age of 15.[7,8,11] JOF is categorized into two subtypes: trabecular and psammomatoid.[2,12] The trabecular variant is characterized by osteoid trabeculae and woven bone, while the psammomatoid type exhibits small, round ossicles resembling psammoma bodies.[12,13] The histological appearance, however, does not predict the growth rate or prognosis.[14] Treatment decisions are based on the clinical and biological behavior of the tumor, often involving radical excision to prevent recurrence.[15]

JOF involves craniofacial bones primarily, mandible being the commonest as highlighted in several case studies.[9,16,17] A case report by Rinaggio et al described a 14-year-old boy with a juvenile ossifying fibroma of the mandible, presenting with congenitally missing teeth.[1] This case highlights the potential association between JOF and other developmental anomalies, adding another layer of complexity to diagnosis and treatment planning.

A retrospective study of 15 cases conducted by Han et al in 2015 analyzes the clinical and radiological features, treatment modalities, and recurrence rates of JOF in 15 patients.[3] The study found a slight female predilection and a mean age of onset of 10.9 years. The mandible was more frequently affected than the maxilla. The study also categorized the cases based on histopathological subtypes (trabecular and psammomatoid) and correlated these with radiographic findings. Importantly, the authors explored the relationship between different treatment approaches and patient prognosis, contributing valuable insights into the management of this condition.

Leimola-Virtanen et al presented two cases of JOF of the mandible, further highlighting the diagnostic and management challenges associated with this condition.[17] The authors emphasize the importance of distinguishing JOF from other fibro-osseous lesions, particularly fibrous dysplasia. They note the well-defined radiographic demarcation of JAOF from surrounding bone, a key feature in differential diagnosis. The report also discusses the surgical management of these cases, emphasizing the potential for recurrence and the need for long-term follow-up.

Chrcanovic and Gomez conducted a systematic review of 405 JOF cases reported in the literature, focusing on lesions in the jaws and paranasal sinuses.[5] Their analysis provides valuable insights into the demographic and clinical features of JOF, including age distribution, anatomical location, and histopathological subtypes. Both variants, psammomatoid and trabecular, showed painless

bone expansion. Radiologically, both variants had a mixed unilocular radiodensity appearance and well-defined limits. However, patients with JPOF were older than those with JTOF. Recurrence was 20.7% and was frequently associated with enucleation and curettage, regardless of the anatomical location or variant type of the lesion. Enucleation followed by either curettage or peripheral osteotomy showed lower recurrence rates than enucleation only. When resection was performed, only one case of JTOF presented recurrence. The review highlights the predilection of JOF for the craniofacial region.

Another updated systematic review on JOF done by Gautier et al in September 2024. This review highlighted on the patient characteristics and histopathological subtypes. The mean age of diagnosis of JPOF was older (19.5 years) than that of JTOF (11.9 years). They reported female predilection of JOF with swelling being the commonest symptom. Conservative surgery was the most performed procedure in 80.1% of patients. The mean recurrence rate reported was 17%.[18] Similarly, Dominguete et al reported a case of recurrent JOF in the jaw of an 18-year-old patient. She had a conservative resection at the age of 14 (tumor curettage and removal of all right inferior molar and premolar teeth).[19] These case studies highlight the importance of early recognition and aggressive surgical management of this rare but challenging lesion.

A case of JOF in a 7-year-old Nigerian boy was reported by Animasahun et al, in which the challenges in diagnosis and management within a resource-constrained setting were highlighted. The importance of considering JOF in the differential diagnosis of jaw masses, even in regions where other pathologies like Burkitt lymphoma are more common, was emphasized in this case. The clinical presentation, including a large facial mass, proptosis, and hypertelorism, was described by the authors, and the diagnostic workup and treatment challenges were discussed.[20]

The available case studies and retrospective analyses provide a comprehensive overview of the clinical, radiographic, and histopathological features of JOF.

CONCLUSION

JOF is a rare, clinically aggressive fibro-osseous neoplasm primarily affecting the craniofacial bones of young individuals, often under the age of 15. The mandible is a common site, and JOF can present with associated anomalies like congenitally missing teeth, adding complexity to diagnosis and treatment. These cases illustrate the diverse presentations of juvenile ossifying fibroma and underscore the importance of complete surgical resection. Case 1, with incomplete resection due to the lesion's proximity to the internal carotid artery, experienced recurrence, highlighting the critical need for thorough removal when feasible. Cases 2 and 3 demonstrated successful outcomes following complete excision. While previous trauma was noted in each case's history, a causal link remains unclear and requires further investigation. These findings emphasize the necessity of prompt diagnosis and aggressive surgical management of JOF to minimize morbidity and prevent recurrence. Long-term follow-up is essential to monitor for potential regrowth, even after complete resection.

In summary, the case studies reviewed provide valuable insights into the clinical, radiographic, and histopathological features of juvenile ossifying fibroma. The reports underscore the importance of early recognition and aggressive surgical management of this rare but potentially aggressive lesion to prevent recurrence and minimize morbidity.

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