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# Juvenile ossifying fibroma

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**Abstract**
Fibro-osseous lesions of the jaws, including juvenile ossifying fibroma, pose diagnostic and therapeutic difficulties due to their clinical, radiological and histological variability. Although juvenile ossifying fibroma is an uncommon clinical entity, its aggressive local behaviour and high recurrence rate means that it is important to make an early diagnosis, apply the appropriate treatment and, especially, followup the patient over the long-term.

**Keywords:** fibro-osseous, fibroma, juvenile

## 1. Introduction

Fibro-osseous lesions of the cranial and facial bones are usually benign and tend to grow slowly. Benign fibroosseous lesions have similar histopathological features with fibrous dysplasia, ossifying fibroma, and cementoossifiying dysplasia [1, 2].

Ossifying fibroma, a rare tumour entity, is a well demarcated benign fibro-osseous tumour with capsule composed of metaplastic bone, fibrous tissue and varying amounts of osteoid [3, 4, 5]. The ossifying fibromas are subdivided into conventional and juvenile clinicopathologic subtypes [3]. Conventional ossifying fibromas are usually slow growing and generally seen in the third and forth decades of life [6, 7]. They are treated with simple curettage and the recurrence is rare [8]. It affects people of all ages, but in contrast to the form seen at adults, the juvenile form is clinically more aggressive and tends to be recurrent [3].

According to the new edition of the classification of the World Health Organization [9], ossifying fibromas which appear as fast growing mass between 5 and 15 years of age, radiologically well bordered, and consistent with ossifying fibroma histologically, are referred as juvenile (aggressive) ossifying fibroma.

Juvenile ossifying fibroma (JOF) appears at an early age and in 79% of the patients are diagnosed before the age of 15 [2, 3, 10]. Males and females are equally affected [11]. JOF originates from periodontal ligament and ranges 2% of oral tumours in children [13]. The JOF is located mainly (85%) in facial bones, in some cases (12%) in calvarium and very seldom (3%) extracranially [2]. Ninety percent of the lesions located in the face region, involve the sinuses, mainly the maxillary antra [2]. Mandibular lesions are seen in 10% of the cases [2, 14]. The tumour is well circumscribed by a tiny sclerotic shell of bone. It appears locally aggressive with cortical disruption and involvement of many adjacent anatomical structures. This lesion has predominating soft tissue consistency with variable amounts of internal calcification and/or linear or irregular focal bone [2]. It usually shows a low density mass due to cystic changes on computed tomography (CT) scans. Following intravenous injection of iodinated contrast, the lesion may show diffuse appearance enhancement [2]. Magnetic resonance imaging (MRI) is important for the lesion extent evaluation, but is inadequate for bony components. It is isointense on T1-weighted images and hypointense on T2-weighted images. Following gadolinium injection, there is homogeneous tumour appearance enhancement [2]. Histologically, JOF is characterized by the presence of cellular fibrous stroma, garland like bony strands and cement particles [2, 6, 11, 13]. The JOFs are classified into two distinct Clinicopathological entities: the trabecular and the psammomatoid types. Trabecular JOF is distinguished by the presence of trabeculae of fibrillar osteoid and woven bone and psammomatoid JOF is characterised by the presence of small uniform spherical ossicles that resemble psammoma bodies [15]. Psammomatoid JOF is reported more commonly than trabecular JOF [14, 16]. Psammomatoid JOF occurs predominantly in the sinonasal and orbital bones, and trabecular JOF predominantly affects the jaws. Psammomatoid JOF has aggressive behaviour and it has a very strong tendency to recur [15-17]. An accurate diagnosis of JOF is made by correlating the clinical, CT scan, MRI and histopathological findings [2].

## 2. Discussion

Most benign fibro-osseous lesions of jaws are asymptomatic and slowly progressing. Moreover, an unusual clinical presentation with apparent aggressive and destructive growth may be expected when the lesion is encountered in a younger patient, especially below the age of 15 years [24, 26].

The JOF is a fibro-osseous lesion that occurs in the facial bones [1, 2]. It is also called aggressive ossifying fibroma due to its aggressiveness and the high tendency to recur, unlike other fibro-osseous lesions, such as cemento-ossifying fibroma, which may resemble radiographically [18]. Due to its distinct histological features, JOF has been recognized as a separate histopathological entity among the fibro-osseous group of lesions [9].

JOF is a relatively rare fibro-osseous lesion of the jaws characterized by the early age of onset i.e., under 15 years of age, the location of tumour, and the radiological appearance and the tendency to recur [28].

JOF affects both males and females equally without any significant gender predilection. However some researches showed that it is more common among men [19]. In contrast, Johnson *et al*. [20] stated that mandibular tumours are more frequently common in girls between the age of 5 - 11 or during the second to fourth decades of life [6].

A few cases of facial trauma have been suggested as a possible etiologic factor in the JOF development [10]. Noffke [21] after 8 year follow-up of a juvenile ossifying fibroma in the left mandible of a 4 year old boy demonstrated initial lack of radiological evidence of demarcation and subsequent eccentric enlargement, selective tooth displacement and a multilocular appearance in areas of active growth. Additionally, an aneurysmal bone cyst and a decrease in the bone content were presented in the excision specimen. Furthermore, osteblastoma, osteosarcoma and odontogenic tumours should be considered in the differential diagnosis of JOF. While the osteoblastoma is radiologically seen as cystic bone lesion with sclerotic boundary, abnormal soft tissue mass and aggressive bone destruction is seen in the osteosarcoma, and cystic lesion connected to premolar or molar teeth is seen in odontogenic tumours [3]. The JOF is characterized as expansive, having defined sclerotic borders, locally aggressive and destructive at cortex on CT scan. This lesion is observed as a soft tissue mass with internal calcification, linear or irregular bone focuses [2, 22]. An increase in diffuse contrast is seen after intravenous injection [22]. Contrast increase is seen on adventitia on MRI [14]. While aggressive cortical changes are seen in juvenile form, sclerotic changes are more common in adult form [2]. JOF is isointense in T1-weighted images and hypointense or isointense in T2-weighted images. Cystic areas can be identified. Following gadolinium injection, a slight increase in contrasting is seen [2, 22].

Ong and Siar [23] presented JOF as a progressively growing lesion that can attain an enormous size with resultant deformity if left untreated. They presented a case of large cemento-ossifying fibroma involving the left mandible in a 15 year old male patient. The long lasting history of untreated JOF resulted to spontaneous fracture of mandible. Furthermore, if JOF do not have adequate surgical treatment, it may have high rate of recurrence [4, 24]. The recurrences are generally seen at early stage and they are more aggressive when compared to primary lesions [4].

There is no consensus on the treatment of JOF cases. Radical resection, local excision conservatively or enucleation with curettage are among the treatment alternatives [4, 13, 25]. Slootweg and Müller [10] suggested that there were no differences between the cases that have limited surgical treatment and those with major surgery in terms of results, and they recommended conservative surgery. On the other hand, Waldron *et al*. [26] suggested that local excision and curettage should be a more preferable method and added that local surgical excision can be applied for recurrent tumour treatment. However, rate of recurrence after conservative treatment was reported in 30 - 58% of cases [4, 27, 28]. Incomplete resection causes recurrence in aggressive tumours. Therefore, some authors were recommended en block resection as an adequate treatment [12, 28]. Curettage together with peripheral osteotomy or sometimes segmental mandibular resection and mandibular reconstruction are suggested in prevalent or recurrent cases [4]. Sarcomatous degeneration is reported to develop in lesions that have recurrence in long term [18]. In contrast, Espinosa *et al*. [4] reported a case of unusual bone regeneration after resection of JOF. Secondary mandibular reconstruction with autogenous grafts was delayed due to the rapid bone formation. Zama *et al*. [8] in a similar case performed resection and reconstruction keeping the mandibular tissue around the temporomandibular joint.

## 3. Conclusion

Although juvenile ossifying fibroma is an uncommon clinical entity, its aggressive local behaviour and high recurrence rate mean that it is important to make an early diagnosis, apply the appropriate treatment and, especially, follow-up the patient over the long-term.

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