CASE REPORT

Rapidly maturing juvenile ossifying fibroma: a case report

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This report describes an unusual case of juvenile ossifying fibroma (JOF) in a 16-year-old female patient with a clinical presentation of a single large, well-circumscribed swelling on left side of the palate. Ossifying fibromas are normally slow growing benign lesions. Radiographically they can present as radiolucent, mixed or radiopaque lesions depending on the degree of maturity. A period of at least 6 years is required for the lesion to pass from the radiolucent to radiopaque stage. All previously reported cases of JOF have been either radiolucent or mixed density lesions, but our case is unusual because the lesion had shown apparently rapid transformation into an almost complete radiopaque stage at a very early age.

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A 16-year-old female reported to the Department of Oral Medicine and Radiology, Sharad Pawar Dental College and Hospital, Wardha, India, with the chief complaint of swelling on left side of the palate, which had been present for 1 year. The swelling was insidious in onset and gradually increased to its present size over a period of 1 year. The swelling was initially painless but pain had developed in the last 15 days. The pain was gradual in onset, moderate, localized and intermittent in nature. Pain was aggravated by touch, chewing and swallowing. As a result of the swelling the patient had difficulty chewing, swallowing and speaking. The patient had no history of trauma or discharge from the swelling. The patient was of moderate build and a healthy weight. Dental history was not significant. The general systemic examination did not reveal any major illness.

Intraoral examination revealed a single large ovoid and well-circumscribed swelling of approximately 3 × 4 cm on the left side of the hard palate. It extended anteroposteriorly from the palatal surface of the left canine to the mesial surface of the left first molar and mediolaterally from the midline of palate to the

palatine gingival margins of the upper left canine, premolars and first molar (Figure 1). The overlying mucosa was smooth and intact. The swelling was hard in consistency, tender on palpation and was fixed to underlying structures. There was no evidence of mobility, caries or tenderness of the teeth in the affected region of the jaw.

Intraoral periapical radiographs of the upper left canine and premolar region showed a single large radiopaque lesion at the periapical region of the upper left canine, and first and second premolars of approximately 2.0 × 2.5 cm in size. The lamina dura was intact. A panoramic radiograph (Figure 2) and a lateral maxillary occlusal view (Figure 3) revealed a single large radiopaque lesion in the upper left posterior region extending anteroposteriorly from the upper left central incisor to the first molar and superoinferiorly 0.5 cm away from the cervical line of the upper left lateral incisor, canine, first and second premolars to approximately 2.5 cm away from the apices, causing the displacement and elevation of the floor of the maxillary sinus. The lesion was well defined with a hyperostotic border and surrounded by the radiolucent rim. Internal structures were amorphous, dense and almost completely radiopaque. A Water's view showed haziness in the left maxillary antrum. On the basis of age, site, clinical behaviour and radiographic findings, a provisional diagnosis of juvenile ossifying fibroma (JOF) was made. Benign cementoblastoma was

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Figure 1 Intraoral view showing palatal swelling

considered as a differential diagnosis. The lesion was subjected to surgical enucleation under general anaesthesia. It was found to be encapsulated and was removed in a single piece. Post-operative healing was uneventful and an obturator was given to the patient for the management of surgical defect. The specimen was sent for histopathological examination. The histological haematoxylin and eosin section showed irregularly interconnecting bony trabeculae in the cellular stroma. Bony trabeculae showed osteoblastic rimming in places (Figure 4). These histopathological findings confirmed the provisional diagnosis of JOF.

Discussion

Benign fibro-osseous lesions of maxillofacial bones comprise a diverse group of pathological conditions that include developmental lesions, reactive or dysplastic disease and neoplasms.¹ The diagnosis of fibro-osseous lesions is often arbitrary and subjective, since both droplet cementum and bone formation are found

Figure 2 Panoramic radiograph showing a single large radiopaque lesion in the upper left posterior region with well-defined hyperostotic border surrounding the radiolucent rim

Figure 3 Maxillary lateral occlusal view showing a single large radiopaque lesion in the upper left posterior region with internal structure amorphous, dense and almost completely radiopaque

in many of these tumours. The diagnosis requires correlation of the histological appearance of the lesion with clinical, radiographic and intraoperative findings as well as the viewer's interpretation of each one's relative significance.^{1,2} Ossifying fibromas are considered to be true benign fibro-osseous neoplasms that develop mainly within the jawbones.^{1,3,4} The updated World Health Organization's (WHO) classification for head and neck tumours^{1,5} does not differentiate ossifying fibroma from cementifying fibroma because the distinction between them is equivocal and there is no clinical relevance. Although the growth of ossifying fibromas is described as concentric with outward expansion approximately equal all in directions,⁶ overall the growth potential of ossifying fibromas is unpredictable and there is no clinical or microscopic features

Figure 4 Photomicrograph (haematoxylin and eosin section; ×10 magnification) showing irregularly interconnecting bony trabeculae in a cellular stroma. Bony trabeculae show osteoblastic rimming in places

that can be used effectively to predict behaviour.¹ Ossifying fibromas are normally slow-growing benign lesions and radiographically can present as a radiolucent, mixed or radiopaque lesion depending on the degree of maturity.⁶⁻⁸ A period of at least 6 years is required for the lesion to pass from the radiolucent to the radiopaque stage.⁴

JOF is also referred to as juvenile active ossifying fibroma or juvenile aggressive ossifying fibroma. It is a benign bone-forming neoplasm and is defined as a variant of ossifying fibroma in the craniofacial skeleton of young patients.^{1,5} JOF is a controversial lesion that has been distinguished from the adult variant of ossifying fibroma on the basis of site, clinical behaviour and microscopic appearance. It is seen in patients at a young age, the most common site of involvement is the maxilla, the clinical behaviour is rapid growth and it is well circumscribed.³ Williams *et al*⁹ analysed 8 cases of JOF, of which 5 were male and 3 were female (age range 5–15 years). Six cases involved the mandible and two involved the maxilla. Noffke¹⁰ reported a case of JOF with an 8-year radiological follow up in a 4-year-old boy involving the mandible. Smith *et al*¹¹ reported a case of JOF, in a 12-year-old female patient involving the mandible. In the present case there was involvement of the maxilla in a 16-year-old girl. Although the histopathological definition of JOF is controversial, the radiographic appearance has similarities with ossifying fibroma.⁶ The term JOF has been used for an aggressive lesion affecting individuals under the age of 15–20 years.^{3,6} The case presented in this article falls in this age group, demonstrating radiographic evidence of a rapidly maturing lesion. The term “mature” has been used to denote the transformation of the lesion from the radiolucent to radiopaque stage.¹² Most cases of cemento-ossifying fibroma (ossifying fibroma according to the WHO classification) studied by Sciubba and Younai¹³ were radiolucent because the patients in their report were younger, whereas the majority of cases reported by

MacDonald-Jankowski⁷ were mixed density lesions because the patients were comparatively older. However, in the present case the lesion became almost completely radiopaque at a young age.

JOF is known to have two distinct histological subtypes, that is, juvenile trabecular ossifying fibroma and juvenile psammomatoid ossifying fibroma. The psammomatoid type mainly involves the bones of the orbit and paranasal sinuses whereas the trabecular type commonly involves the jaws.^{5,14} The histological criteria for accepting any case as JOF was defined by the WHO¹⁵ and Slootweg *et al*.¹⁴ According to them, well-demarcated lesions showing a fibrocellular stroma containing trabeculae of osteoid and woven bone with or without osteoblastic rimming with paucity of cementicles and containing giant cells can be considered as JOF. A fibrocellular stroma containing trabeculae of osteoid and woven bone was present in all cases discussed above. Osteoblastic rimming was present in all cases of JOF reported by William *et al*,⁹ in some cases of JOF studied by Slootweg *et al*¹⁴ and in the present case.

All cases of JOF previously reported were either radiolucent or mixed-density lesions but our case was unusual because the lesion showed apparently rapid transformation into the almost complete radiopaque stage at a very early age. In conclusion, the present case can be distinguished from the adult variant of ossifying fibroma on the basis of age, site and clinical behaviour, as well as radiographic and microscopic appearance.

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