Ossifying fibromas of the jaws and craniofacial bones

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Abstract

Fibro-osseous lesions include a large group of pathologic processes in which normal bone is replaced by fibrous tissue containing variable amounts of mineralised material. Due to considerable similarity of features, definitive diagnosis of these lesions requires an accurate correlation of the clinical, radiographic and histopathological findings.

Ossifying fibroma is a fibro-osseous neoplasm that commonly affects the jaw bones and comprises three distinct entities, all with overlapping features. We review the clinicopathologic and radiographic features of cemento-ossifying fibroma (COF), juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF).

Keywords cemento-ossifying fibroma; fibro-osseous lesions; juvenile ossifying fibroma; juvenile psammomatoid ossifying fibroma; juvenile trabecular ossifying fibroma; ossifying fibroma

Introduction

Benign fibro-osseous lesions of the jaws and craniofacial bones are a group of intraosseous pathological conditions characterised by replacement of normal bone by fibrous tissue containing foci of mineralisation.[1] The group includes developmental, dysplastic and neoplastic lesions.[2] Clinically, the fibro-osseous lesions of the jaws show variable features ranging from localised asymptomatic lesions, detected only by routine radiographic examination, to significant lesions with cosmetic and functional disturbances. Although some of these lesions can be diagnosed by histological analysis, in most cases the overlap in the histological features makes the diagnosis challenging for the pathologist, and a final diagnosis can only be confirmed by reviewing and correlating the clinical, histological and radiological features.[1]

There is no generally accepted classification for fibro-osseous lesions of the jaws, although many attempts have been made. The reasons behind this may be due to lack of consensus on the diagnostic terminology for these lesions, the overlap in the microscopic features and the existence of rare lesions which may not be specific to the jaw bones. In the most recent classification of head and neck tumours by the World Health Organization,[3] the classification of odontogenic tumours includes three different clinicopathological groups of benign fibro-osseous lesions: fibrous dysplasia, osseous dysplasias and ossifying fibroma (Table 1).

Suggestions have been made that fibro-osseous lesions can be classified based on radiographic appearance and growth pattern.[4] Poorly-delineated lesions which involve large expanses of bone are most likely to be fibrous dysplasia while the main diagnostic feature of ossifying fibroma is its well-defined growth pattern. This may be radiolucent with some degree of associated radiopacity in the absence of other lesions. Osseous dysplasias are in the tooth bearing areas and are often associated with the root of a tooth. However, the actual diagnosis is rarely this straightforward, and a definitive diagnosis must be based on clinicopathological features as well as the radiology.[1–4]

In this review we will present the clinicopathological variants of ossifying fibroma, and highlight the key features that can be used to assist in diagnosis.

Ossifying fibroma

Ossifying fibroma (OF) is a benign fibro-osseous neoplasm characterised by progressive growth with bony expansion and well demarcated margins radiologically. Microscopically they contain cementum-like material or bone in a fibrous connective tissue stroma.

The current classification of ossifying fibroma comprises three distinct entities.[1–3] First, the conventional ossifying fibroma of the jaw, which has an odontogenic origin and variable morphology. Several names have been suggested in the literature including ossifying fibroma, cementifying fibroma, cemento-ossifying fibroma, ossifying odontogenic fibroma and periodontoma. In the 1992 WHO classification of odontogenic tumours the favoured term was 'cemento-ossifying fibroma',[5] but this terminology was used loosely. In part it was preferred because of the association of these lesions with teeth, but also because of the presence of "cementum-like" material, even in

WHO Classification of fibro-osseous lesions of the maxillofacial region (from Barnes et al., 2005.[3])

1. **Fibrous dysplasia**
   - Monostotic fibrous dysplasia
   - Polyostotic fibrous dysplasia
   - Craniofacial fibrous dysplasia
2. **Osseous dysplasias**
   - Periapical osseous dysplasia
   - Focal osseous dysplasia
   - Florid osseous dysplasia
   - Familial gigantiform cementoma
3. **Ossifying fibroma**
   - Conventional ossifying fibroma
   - Juvenile trabecular ossifying fibroma
   - Juvenile psammomatoid ossifying fibroma

Table 1

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lesions away from the tooth-bearing areas. This was changed in the 2005 WHO classification[3] so that all variants of "cemento-ossifying fibroma" were regarded as ossifying fibroma. This is due to the assertion that cementum and bone are essentially the same tissue with the same chemical composition, with no distinction between them except their relationship to the tooth root.[6] When the cementum is not associated with the tooth root as it is normally, it is no longer considered as a specific tissue. However, there is a general consensus that the conventional ossifying fibroma originates in the periodontal ligament of the tooth-bearing areas of the jaws and is of odontogenic origin.[7] The periodontal precursor cells may give rise to fibroblasts, bone or cementum[8] and it therefore seems that *cemento-ossifying fibroma* is the best descriptive name for this tumour and should be revived.[9] Furthermore, the term cemento-ossifying fibroma is suitably descriptive and indicates that these lesions are specific to the tooth bearing areas of the jaws and clearly distinguishes it from the juvenile variants.

The two further variants of ossifying fibroma are both termed juvenile ossifying fibroma. Although not always arising in children they do have a predilection for young people. These lesions have also had a number of changes in terminology. They have previously been designated as "active" or "aggressive" which refers to their purported aggressive nature. However, although they often show rapid growth and may recur, this probably reflects their occurrence in young people and their sites of origin. Very few lesions display true aggressive behaviour and there are no reports of malignant change. The two types do, however, show distinct clinical and histopathological features and are now recognised as two distinct entities; the *juvenile trabecular ossifying fibroma* and *juvenile psammomatoid ossifying fibroma*. The key features of the three variants of ossifying fibroma are summarised in Table 2.

Cemento-ossifying fibroma (conventional ossifying fibroma)

Cemento-ossifying fibroma (COF) is considered one of the most common benign fibro-osseous lesions to occur in the jaws. Although the overall prevalence has not been reported, they represent about 2–4% of jaw lesions, which is similar to fibrous dysplasia. COFs tend to appear over a wide age range between 8 and 53 years with the majority of patients in the second to fourth decades.[1] With regard to gender variance, COF shows a predilection for females with a male to female ratio of 1:5.[3] COF are most commonly encountered in the mandible with a predilection for the molar and premolar regions. A review of 64 cases reported that 90% arose in the mandible and that of these, 52% were found in the molar region followed by the premolar (25%), incisor (12%) and canine (11%) regions.[10] This predilection in the mandible has also been reported in many other studies.[1,11] Maxillary lesions have been reported both in anterior and posterior regions. In most cases, COF of the jaws has been described as a slow growing mass but occasionally locally destructive lesions have been reported. Although bone expansion of the buccal and lingual cortical plates was the most frequent clinical presentation, rarely, painful swelling and buccal perforation has been observed. Overall, there are no identifiable clinical or microscopic features to predict the behaviour of this tumour.

Radiographic features

COF shows variable radiographic features. Some lesions are completely radiolucent, whilst others are completely radiopaque, with most cases being mixed-density[7] (Figure 1). Many tumours

Summary of the key clinical, radiographic and histological features of the three ossifying fibroma variants

	Cemento-ossifying fibroma (COF)	Juvenile trabecular ossifying fibroma (JTOF)	Juvenile psammomatoid ossifying fibroma (JPOF)
Origin	Odontogenic	Non-odontogenic	Non-odontogenic
Peak age	2nd–4th decades	1st and 2nd decades	2nd–3rd decades
Sex	Female predominance (M:F 1:5)	No sex predilection (M = F)	Slight male predominance (M:F, 1.2:1)
Location	Mandible Tooth bearing areas Molar/premolar region	Mandible Non-tooth bearing areas Angle and ramus. Paranasal sinuses	Craniofacial bones Paranasal sinuses
Clinical features	Slow growing mass with buccal and lingual expansion	Bony swelling, facial deformity	Bony expansion of orbital and paranasal bones
Radiology	Well demarcated, variable radiopacity. Tooth displacement with minimal root resorption	Well-circumscribed uni- or multi-locular radiolucency. Variable radiopacity. Root resorption and displacement	Well-circumscribed. Corticated border. Expansion of paranasal bones. Filling of nose and sinuses
Histology	Variable. Encapsulated with border. Hypercellular Well formed bone trabeculae and "cementicles"	Well-demarcated but not encapsulated. Osteoid trabeculae within variably cellular stroma. Multinucleated giant cells and aneurysmal bone cyst formation may be seen	Well-demarcated but not encapsulated. Multiple concentric 'psammomatoid' ossicles. Occasional cystic change
Recurrence	Low recurrence	High recurrence (30–50%)	High recurrence (30–56%)

Table 2