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Case report

Familial ossifying fibromas: report of two cases

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Abstract: Ossifying fibroma is a benign fibro-osseous lesion of the jaw containing varying amounts of calcified deposits such as bone, cementum or both. This type of lesion is referred to as dysplastic or neoplastic in nature. In 2000, a 52-year-old male patient was referred to our clinic complaining of a giant swelling in the mandibular premolar-molar region. A histopathological diagnosis of ossifying fibroma was made. Three months later, his daughter was admitted with a swelling on her mandible. Following biopsy, this patient was also diagnosed as having ossifying fibroma. The present report describes these two cases of familial and multiple ossifying fibromas. (J Oral Sci. 46, 61-64, 2004)

Key words: cemento-ossifying fibroma; familial; fibro-osseous lesion; multiple; ossifying fibroma.

Introduction

Ossifying fibroma is a benign fibro-osseous lesion that demonstrates a well demarcated or rarely encapsulated proliferation of cellular fibrous tissue with varying amounts of osseous products including bone, cementum or a mixture of both. Traditionally, this type of lesion was subclassified histologically into ossifying fibroma and cementifying fibroma according to the hard tissues formed, but both types are now known by the unifying term ossifying fibroma. It is generally accepted that the histological subclassification of these two lesions is of academic interest only since differential diagnosis is often arbitrary and their biological behavior seems to be identical (1). Ossifying fibroma develops from the multipotential mesenchymal cells of periodontal ligament origin which are able to form both bone and cementum (2,3). Although the precise pathogenesis is still unknown, Wenig et al. (4) has suggested that trauma-induced stimulation may play a role.

We report an unusual presentation of ossifying fibromas with multiple and familial occurrences.

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Case 1

In 2000, a 52-year-old man was referred to our clinic with the chief complaint of a giant painless, firm swelling on the left mandibular premolar-molar region. There was no significant medical or dental history. The swelling had first been noticed two years previously. Clinically there was a large buccal swelling approximately 80 mm in diameter with a well defined border. The tumor extended from the left mandibular premolar region to the posterior ramus. The swelling was non-tender, non-fluctuant and firm, and covered by intact mucosa. There was no paresthesia or hypoesthesia. Laboratory findings were within normal limits.

On panoramic radiography and CT, a large, well defined, very dense lesion extending from the distal of the canine to the posterior ramus was observed. On the left mandible there was cortical expansion and the inferior border of the mandible was thinned but remained intact. A similar but smaller lesion was also present in the maxillary molar region (Fig. 1a, 1b).

After incisional biopsy, both lesions were histopathologically diagnosed as ossifying fibromas. Microscopic examination revealed a tumor composed of bundles of

spindle fibroblastic cells containing spherical and small bone spicules. Around the bone spicules, osteoblasts were present. No atypical cells or mitosis were observed (Fig. 1c).

A left hemimandibulectomy was performed just beyond the periphery of the mass. This process was followed by immediate reconstruction using a titanium plate and an iliac bone graft. The lesion in the maxilla was also totally excised at the same time. The surgical specimens were submitted for histopathological examination and the diagnosis of ossifying fibroma was confirmed.

Case 2

Three months later, a 20-year-old woman, the daughter of the Case 1 patient, was admitted to our department with a swelling on her right mandible. Symptoms had been present for 6 months. On examination, a large buccal swelling extending from mandibular canine to the 1st molar was noted. The overlying mucosa was intact. The lesion was firm, non-fluctuant and the patient reported mild tenderness. On panoramic radiography and CT, a well circumscribed, non-homogeneous lesion approximately 45 mm in diameter and with radiopaque foci was found (Fig. 2a, 2b). The buccal surface of the cortical bone was thinned and the lingual cortical plate was destroyed. The lesion had caused root resorption of the first molar and displaced the second premolar. The involved teeth were vital except the second premolar. Laboratory findings were within normal limits.

Histopathological examination revealed a tumor composed of bundles of spindle fibroblastic cells, which were arranged in different directions. In the stroma, cementum-like hard tissues fused with small bone spicules were observed. Around these masses, cementoblast-like

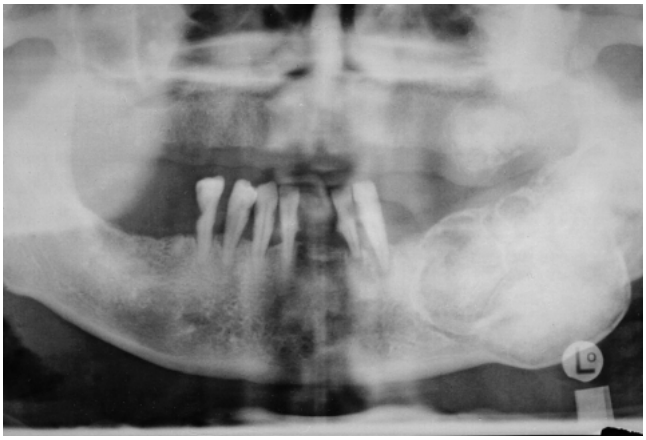

Fig. 1a Panoramic radiograph showing well defined, dense lesions in the mandible and maxilla.

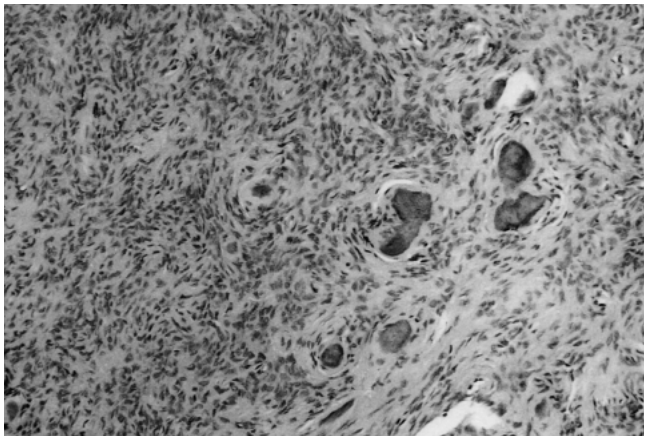

Fig. 1c Photomicrograph revealing a tumor composed of spindle fibroblastic cells and a few calcified deposits. (HE ×200)

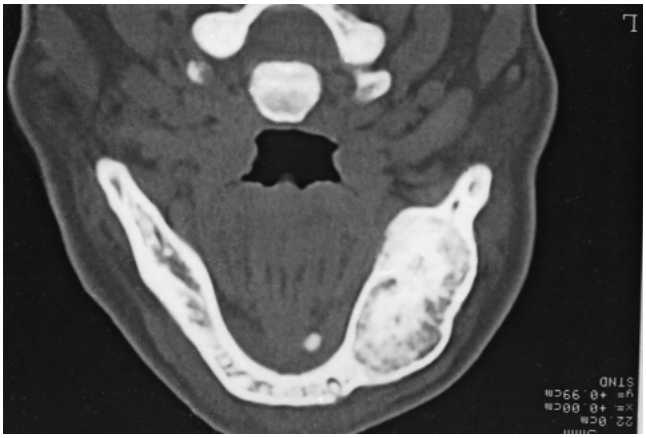

Fig. 1b CT showing a well defined mandibular lesion with thinning and expansion of cortical bone.

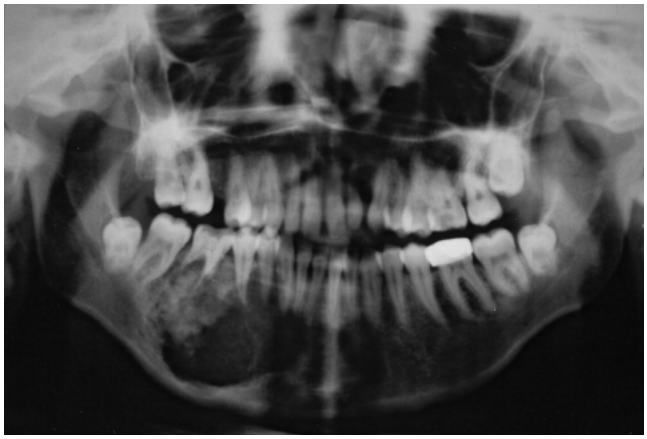

Fig. 2a Panoramic radiograph showing a well circumscribed, non-homogeneous lesion with radiopaque foci and root resorption of the first molar.

or osteoblast-like cells were noticed. No atypical cells or mitoses were observed. The result of the examination suggested a diagnosis of ossifying fibroma (Fig. 2c).

During the operation it became apparent that the lingual cortex of the mandible was perforated and the inferior alveolar nerve was displaced through the lower border of the mandible. While preserving the nerve, a soft yellowish mass containing solid and bone-like components was removed totally with curettage under local anesthesia.

Discussion

Ossifying fibroma is a slow-growing, well circumscribed, painless lesion usually seen in the third and fourth decade of life. Females are predominantly affected (5,6). However, the first of our cases was a male in the sixth decade of life. Our second patient experienced some episodes of pain. The mandible, including the ramus, is the region most commonly affected, and bimaxillary occurrence is extremely rare (7,8).

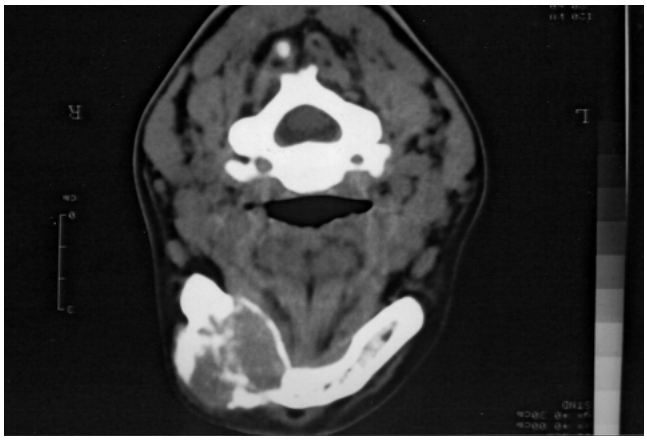

Fig. 2b CT showing an irregular dense lesion and perforation of the bone.

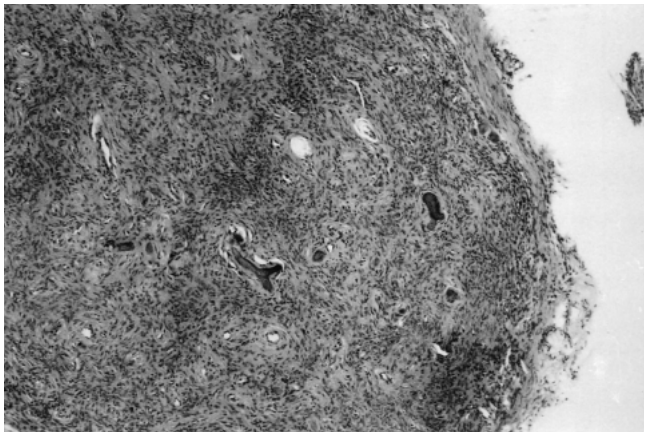

Fig. 2c Photomicrograph revealing a tumor made up of spindle fibroblastic cells and scattered osteoid trabeculae. (HE ×100)

There are few cases of multiple occurrence of ossifying fibroma. Takeda and Fujioka (9) reported multiple lesions in three jaw quadrants and Hauser et al. (10) presented a case of massive, bilateral ossifying fibromas in the maxillary sinuses that caused facial deformity and orbital compression. Subsequently, Hwang et al. (11) described a case of ossifying fibromas in all quadrants discovered at different times. Recently, Bertolini et al. (2) reported an additional case in the maxilla and mandible. Regezzi and Sciubba (3) commented that multiple ossifying fibromas are usually sporadic but there is a familial inclination in some cases. They also stated that chromosomal translocations have been identified in a few instances. To our knowledge there is only a single reported case of familial occurrence in which ossifying fibromas involved three jaw quadrants, and the patient's mother had left mandibular and maxillary lesions of ossifying fibroma (7). In our present cases, two generations of a family exhibited lesions with similar histological and clinical properties. Yih et al. (7) suggested the possibility of a hereditary element. In our cases, these kinds of lesions had never previously been seen in the family. Unfortunately, genetic analysis could not be performed because of the patients' poor cooperation.

It is known that ossifying fibroma is composed of fibrous proliferation containing several kinds of mineralized material. Such deposits consist of woven or lamellar bone or cementum-like spheroids (1). Lesions larger than 8 cm in diameter, such as that in Case 1, are termed giant ossifying fibromas (5).

In the craniofacial region the lesion enlarges slowly and symmetrically resulting in bone expansion and facial deformity, but some lesions are asymptomatic and are only discovered during routine radiographic examination (6,8). Centrifugal growth will commonly cause bowing of the inferior border of the mandible but cortical perforation is rare (1). In Case 2, perforation was noticed on CT and was confirmed at the time of surgery. CT is a valuable tool for detecting soft tissue involvement. Radiographic appearances vary considerably from case to case and depend on the stage of lesion maturity (5,12), ranging from immature, radiolucent and cyst-like with scattered radiopaque foci to mature dense sclerotic lesions (5).

The lesion may occasionally cause displacement of the mandibular canal (2). In Case 2, displacement of the mandibular canal through the lower border of the mandible was noticed on CT and confirmed during surgery. Resorption and/or divergence of roots can occur with the continuous growth of the lesion, as in Case 2 where the

root of the mandibular first molar was resorbed (1).

Uncomplicated cases can be treated by simple enucleation of the lesion with curettage alone. Because the lesions are well circumscribed, they are removed easily from the surrounding tissue. On the other hand, larger lesions will require more radical surgical resection (1,8). It is also recognized that if there is no facial deformity and no evidence of concomitant osteomyelitis, a 'wait and see' approach is an appropriate treatment choice (13).

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