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Case report

Central odontogenic fibroma of the mandible: A case report with diagnostic considerations

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HIGHLIGHTS

- Attention to any gingival enlargement: it may be a clinical COF manifestation.
- A wrong diagnosis may determine a serious delay in patient proper treatment.
- It is highly recommended a periodic clinical and radiographic examination.
- A careful cyto-histological examination of fibrous lesions of the jaw is necessary.
- A proper differential diagnosis will allow the correct patient management.

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ABSTRACT

Introduction: Odontogenic fibroma (OF), a rare odontogenic tumor of mesodermal origin, has been thought to originate from either dental follicle, periodontal ligament, or dental papilla [1]. Different studies reported high variability in the incidence rate as being between 3 and 23% of all odontogenic tumors [2,3]. OF manifests a dual character at the histopathological examination showing odontogenic epithelial structures mimicking those observed in biopsy of ameloblastoma and, in addition, peculiar fragments of cellular stroma. The clinical and radiological features of OF are similar to other odontogenic and/or non-odontogenic tumours and the differential diagnosis may first occur at fine-needle aspiration biopsy.

Presentation of case: In the case reported, a young patient showed a localized gingival enlargement involving radiologically the superior margin of the right angle of the mandible and associated with an un-erupted tooth. The morphological characteristics together with clinical and radiologic findings confirmed the tumor to be a central odontogenic fibroma (COF) with secondary gingival involvement.

Discussion and conclusion: Benign odontogenic tumors may be distinguished from other odontogenic/non-odontogenic neoplasias and from malignant tumours through a cytologic differential diagnosis as treatment differs accordingly.

1. Introduction

Odontogenic fibroma (OF) is a rare tumor of odontogenic origin, with variable percentages of incidence [2,3], regarded by the World Health Organization (WHO) as a benign odontogenic neoplasm derived from mesenchymal odontogenic tissue [4,5].

The lesion occurs most commonly in the mandible although several cases have been reported in the maxilla [6]. It has been

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reported in patients ranging in age from 11 to 80 years with a mean age of 34 years. Daley et al. reported in a literature review a slight female predominance [7]. The lesion grows slowly, in an asymptomatic manner. On radiographs OF can appear as a single well-defined radiolucency, or as a multilocular lesion frequently associated with unerupted or displaced teeth.

OF is characterized by varying amount of inactive-looking odontogenic epithelium embedded in a mature relatively dense collagenous fibrous stroma. Most pathologists believe that the absence of odontogenic epithelium does not preclude a diagnosis of odontogenic fibroma [1,2,8].

Wesley et al. had the merit to clarify the nature of the tumor, differentiating it from other pathological entities as odontogenic myxoma, hyperplastic dental follicle, and other fibro-osseous lesions [1].

Gardner has identified three histological separate variants of the OF: the hyperplastic dental follicle or fibrous hyperplasia, the epithelium-poor type (simple type) and the epithelium-rich type (complex or WHO-type) [8].

The hyperplastic dental follicle is composed of fibrous connective tissue with scattered focal nests of odontogenic epithelium, smaller than the epithelial islands found in ameloblastomas or ameloblastic fibromas and without stellate reticulum and signs of cellular polarization.

The simple type is minimally cellular with delicate collagen fibers, a considerable amount of fibromyxoid matrix and only scattered and small remnants of inactive-looking odontogenic epithelium. The WHO type is more complex and consists of cellular mature fibrous connective tissue in which sparse or often conspicuous strands of odontogenic epithelium are found, together with calcified tissue either in the form of dysplastic dentin, or cementum-like material.

Topographically, two variants can be distinguished: extraosseous or peripheral type (POF) and an intraosseous or central type (COF) [4,5]. Table 1 summarizes the different classifications of OF [5,8–10].

Herein we presented the clinical, radiological and morphologic features of a particular case of central odontogenic fibroma (COF) with secondary gingival involvement, with the aim to reinforce the concept of a necessary multidisciplinary diagnostic approach for all gingival growths, also those ones seemingly as innocuous lesions.

2. Presentation of case

A 12-years-old boy presented with a slow-growing, asymptomatic gingival enlargement distal to the mandibular right first molar. His medical, surgical, family, and social histories were unremarkable. Oral examination showed a soft swelling at the lower right second molar region. The overlying mucosa was normal. There was no cervical lymph-node involvement.

A panorex (Fig. 1A) showed a large uni-locular, radiolucent lesion localized at the superior margin of the right angle of the mandible, limited by a sclerotic border and in relation to an impacted second molar. A subsequent CT scan confirmed an heterogeneous mass eroding the superior margin of the right mandibular angle with a significant enlargement of the overlying mucosa. The patient was referred for a fine-needle aspiration (FNA) biopsy. This procedure was performed via several passes with a 22-gauge needle through transmucosal sampling of the mass. Both bloody and viscous tan material was obtained. Citological examination showed cohesive sheets of epithelial cells and stromal component consisting in of cellular fibroblasts with blurred borders. The diagnosis of possible benign odontogenic tumor was hypothesized, and, consequently, conservative enucleation of the lesion, extraction of the impacted molar and curettage of the surrounding tissues were performed under general anaesthesia. During the surgery, the lesion appears as an expansion of the cortical plate of the mandible, it was easily removed, not showing any adherence to bone, the inferior alveolar nerve was preserved and the patient retained normal sensation. In this case, no guided bone regeneration has been performed.

The surgical specimen was fixed in 10% neutral buffered formalin and sent for histological examination. Grossly, it measured $3 \times 4 \times 3$ cm, and showed smooth whitish appearance with circumscribed outline (Fig. 1B).

Microscopically, tissue sections exhibited nests and irregular branched strands of inactive epithelial cells in a densely fibrous, moderately cellular, and only focally myxoid connective background (Fig. 1D, F). The epithelial tumor component was characterized by serpentine strands and islands of odontogenic epithelium, without palisading aspects and stellate reticulum formation, surrounded by fibrous stroma with fascicular configuration. Tumor stroma consisted of plump oval to spindle cells in prevalently mature collagen tissue highlighted by Van Gieson staining (Fig. 1E). Only focal trabeculae of calcified material or bone have been detected.

The microscopic characteristic together with clinical and radiologic findings confirmed the tumor to be central odontogenic fibroma (COF) with secondary gingival involvement during its expansion.

Healing and postoperative course were uneventful, the clinical examination showed a normal alignment of the teeth and the patient was discharged for further follow-up. One year after surgery, there were no signs of recurrence and both bone and soft tissue healing was satisfactory (Fig. 1C).

3. Discussion

Odontogenic fibroma is a rarely reported lesion of mesenchyme and odontogenic ecto-mesenchymal origin exhibiting a slow clinical growth. The World Health Organization (WHO) defined it as a benign odontogenic neoplasm of fibroblastic origin characterized by relative mature collagenous fibrous tissue with or without varying amounts of embedded inactive-looking odontogenic

Table 1
Different classifications of OF.

Histological classifications				
Gardner, 1980 [8]	Hyperplastic dental follicle	Simple type fibrous neoplasm with collagenous fibrous connective tissue containing inactive-looking odontogenic epithelium	Complex or WHO-type lesion with dysplastic dentine or tissue like cementum, fibrous tissue with myxoid area and sparse or often conspicuous inactive looking odontogenic epithelium	
Lukinmaa PL et al., 1990 [9]; Langlais et al., 1995 [10]		Simple type	Complex or WHO type	Granular cell type
WHO, 2005 [5]		Epithelium-poor type	Epithelium-rich type	
Topographical classification				
Intra-osseous or central				Extra-osseous or peripheral