

# Central odontogenic fibroma of simple type: An original observation

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## Abstract

Central odontogenic fibroma is an uncommon, benign, slow-growing intraosseous mesenchymal odontogenic tumour. It presents a diagnostic dilemma to the clinician and the pathologist because its clinical and radiological features resemble other odontogenic and/or non-odontogenic tumours, and the differential diagnosis is based on histological examination. In this report, we describe our experience with a case of a 23-year-old female patient with central odontogenic fibroma of the mandible that was diagnosed as ‘simple type’. Highlighting a subtype that was dropped from the last World Health Organization classification of head and neck tumours is important to accumulate more information about this lesion and to show its different features. Despite its rarity, central odontogenic fibroma should be included in the differential diagnosis of intrabony tumours of the jaws. These findings can better educate oral and maxillofacial surgeons about the unusual nature of this lesion, help establish a correct diagnosis and give the appropriate therapeutic management.

## Keywords

Central odontogenic fibroma, odontogenic tumours, tumour enucleation, differential diagnosis

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## Introduction

Central odontogenic fibroma (COF) is a rare, benign mesenchymal odontogenic tumour occurring within the jaw.<sup>1</sup> It is defined by the World Health Organization (WHO) as a neoplasm made of mature fibrous connective tissue, with variable amounts of inactive-looking odontogenic epithelium, with or without the evidence of calcification.<sup>2</sup> Accounting for about 0.1% of all odontogenic tumours and 6.1% of all central odontogenic tumours,<sup>3</sup> COFs present a diagnostic dilemma to the clinician and the pathologist. The clinical and radiological features of this entity resemble other odontogenic and/or non-odontogenic tumours, and the differential diagnosis is based on histopathological examination.

In 2005, the WHO sub-classified this tumour into two histological types: the simple odontogenic fibroma (epithelium-poor) and the complex odontogenic fibroma, WHO type (epithelium-rich). In the 2017 WHO classification, the consensus group dropped the sub-classification of ‘epithelium-poor or simple type’ of odontogenic fibroma as they decided it was poorly defined and documented.<sup>4</sup> However, recent scientific publications still mention this sub-classification. According to

Seo et al.,<sup>5</sup> investigators should consider that any single COF entity may exhibit the 2 different histological patterns.

In this report, we describe our experience with a case of a young female patient with COF of the mandible that was diagnosed as ‘simple type’. We discuss the clinical, radiological and histological features that enabled the differential diagnosis and the appropriate treatment to manage this tumour.

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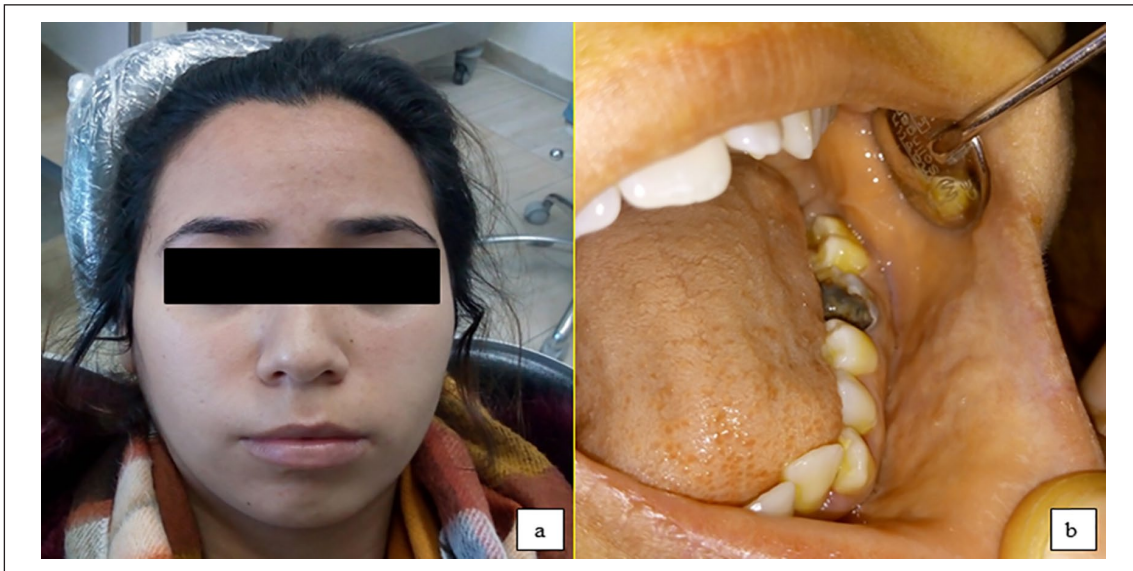
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**Figure 1.** Extraoral and intraoral views of the patient: (a) Face view without particularities (no facial asymmetry). (b) Intraoral view showing insufficient hygiene, absence of swelling and deep decay in tooth 36.

## Case report

A 23-year-old female patient with no notable medical and family history was referred to our department after the discovery of a radiolucent image which spans the entire left mandibular ramus. She first consulted her dentist after the sensation of discomfort and slight pain in the left retromolar region. The extraoral examination was without particularities (no facial asymmetry) (Figure 1(a)). The intraoral examination revealed insufficient hygiene, absence of swelling and deep decay in tooth 36 (Figure 1(b)).

The panoramic radiograph showed a well-defined, corticated radiolucent lesion extended over the ramus in relation to the impacted wisdom tooth 38 (Figure 2). The computed tomography (CT) scan revealed that this unilocular osteolytic lesion was with tissue matrix, very slightly enhanced by the contrast agent and contained sparse millimetric calcifications. It was not very blowing, eroding the internal surface of the internal cortical without cortical rupture. This lesion measured 34mm in the coronal plane and 10 × 25mm in the axial plane (Figure 3(a)–(c)). Based on the clinical and radiological features, we made a diagnosis of a calcifying epithelial odontogenic tumour (Pindborg tumour) or ameloblastoma.

The patient was treated under general anaesthesia. After detachment of a full thickness flap, bone milling and resection of the coronoid process to facilitate direct access to the lesion, we performed the total enucleation, curettage and extraction of the impacted wisdom tooth. The immediate postoperative follow-up was straightforward without complications. The lesional tissue was submitted to the pathology department for histopathological evaluation. Macroscopically, the tumour consisted in a whitish-yellow mass of soft consistency related to the impacted tooth (Figure 4(a)). Microscopically, the lesion was essentially made of a tissue of fibrous appearance with cells



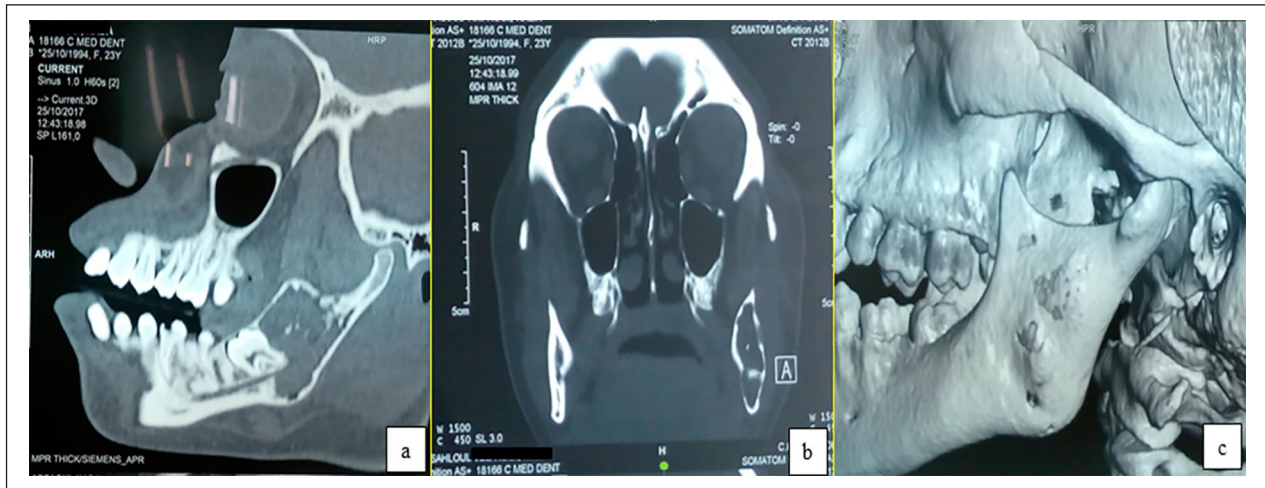
**Figure 2.** Preoperative panoramic radiograph. Panoramic radiograph showing a well-defined, corticated radiolucent lesion extended over the ramus in relation to the impacted wisdom tooth 38 (the arrows indicate the outlines of the lesion).

having an abundant pale cytoplasm, with rounded or ovoid nucleus and fine chromatin. The cell density was moderate with the presence of a few hyaline and oedematous areas. It was associated with calcifications and rare foci where the cells take on an epithelial aspect; they were cubic, with a regular nucleus. There were a few thick-walled vessels and the lesion included clumps of adipocytes at the periphery. No signs of malignancy were seen (Figure 4(b)–(f)). A final diagnosis of COF of simple type was retained.

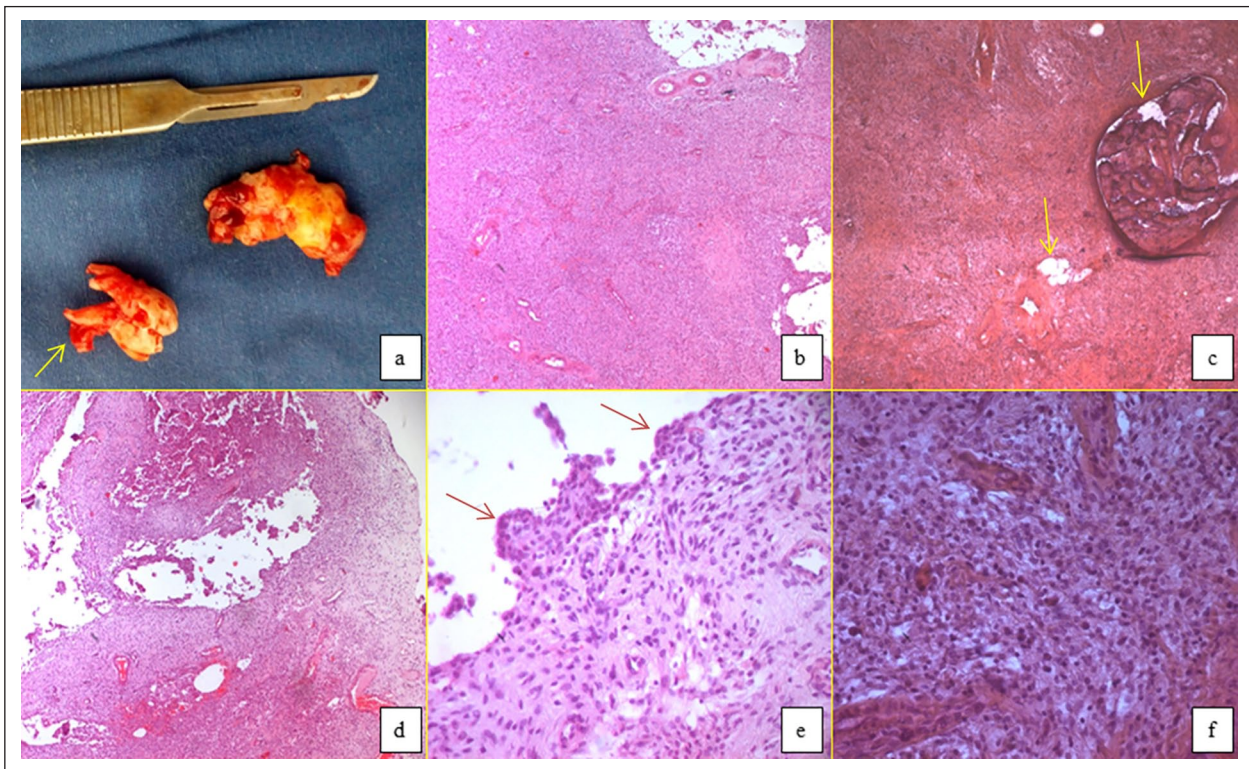
The panoramic radiograph performed 5 months after the surgery showed the beginning of re-ossification of the ramus (Figure 5). There was a full clinical healing without noting inferior alveolar nerve damage or any signs of recurrence. The patient was motivated in oral hygiene and referred to her dentist for the treatment of decayed teeth.

## Discussion

Odontogenic fibroma (OF) is topographically classified into two types according to the WHO: the intraosseous or central



**Figure 3.** Preoperative CT scan: (a), (b) Sagittal and coronal cuts showing a unilocular osteolytic lesion was with tissue matrix containing sparse millimetric calcifications and thinning the cortical bone without rupture. The lesion invades the mandibular canal. (c) 3D reconstruction showing a thinning of the cortical bone.



**Figure 4.** Macroscopic view and histological examination of the lesion leading to the diagnosis of COF of simple type: (a) The tumour was a whitish-yellow mass of soft consistency related to the impacted tooth 38 (Macroscopic view). (b), (d) Tissue of fibrous appearance containing fibroblasts and rare amounts of epithelial cells (HE  $\times 40$ ,  $\times 100$ , respectively). The cell density was moderate with the presence of a few hyaline and oedematous areas. There was no atypia or mitotic activity. (c) Presence of intralésional calcifications (Yellow arrows) (HE  $\times 40$ ). (e) Rare foci where the cells take on an epithelial aspect (Red arrows) (HE  $\times 200$ ). They were cubic with a regular nucleus. (f) Fibrosis: the fibroblasts have an abundant pale cytoplasm with rounded or ovoid nucleus and fine chromatin (HE  $\times 200$ ). There were a few thick-walled vessels.

type which was our case and the extraosseous or peripheral type.<sup>6</sup> The latter primarily involves the mandibular gingiva, usually from the anterior region to the premolar area.<sup>7</sup> It is a

tumour that can appear at any age (mean=40 years) and display a slight predilection for females similarly to the reported case.<sup>8</sup> The aetiology of COF remains unclear, and no obvious



**Figure 5.** Panoramic radiograph (5 months follow-up). Beginning of re-ossification of the ramus. No signs of recurrence.

causative environmental factors have been identified. Diniz et al.<sup>9</sup> speculated that some odontogenic tumours, including COF, probably arise because of developmental defects in normally quiescent genomes, which then trigger oncogenic mutation pathways without progressing to the stage of malignant transformation.

COFs occur equally in the two jawbones. The most common locations are the anterior region of the maxilla (71%) and posterior region of the mandible (73%).<sup>10</sup> In our case, we retrieved a posterior mandibular location and the tumour was associated with the inclusion of the wisdom tooth 38. Clinically, COFs are usually asymptomatic and present slow-growing expansible swellings of the jaws. Rarely, they can be aggressive and provoke dental displacements and radicular resorptions.<sup>7</sup> For our patient, she consulted for a discomfort and a slight pain. There was no apparent swelling.

Radiographically, conventional radiographs and CT images of this tumour present a uni- or multilocular radiolucent areas with well-defined borders associated with an impacted tooth.<sup>11</sup> A mixed radiolucent and radiopaque lesion was found in only 10.7% of the cases in the mandible.<sup>8</sup> However, these features are not specific to COF. Hara et al.<sup>6</sup> reported that dynamic magnetic resonance imaging (MRI) could be useful for diagnosing COF because the time-signal intensity curves (TICs) pattern of OF are different from those of other odontogenic tumours. COF is reported to show several signs like cortical bone perforation, tooth displacement, cortical thinning, bone expansion and rare areas of radiopacity.<sup>12</sup> In our case, we observed a unilocular lesion with well-defined margins containing sparse calcifications and associated with the impacted wisdom tooth.

The clinical and radiographic differential diagnosis includes cysts of odontogenic origin, calcifying epithelial odontogenic tumour (Pindborg tumour), adenomatoid odontogenic tumour, ameloblastic fibroma and ameloblastoma. Due to the rarity of COF, it was excluded in our differential diagnosis list and we expected Pindborg tumour or ameloblastoma in first intention. It is important to note that histopathological examination is the only definitive way to obtain the appropriate diagnosis.

Histologically, the epithelium-poor type is characterized by a fibromyxoid stroma with scattered odontogenic epithelial islands and cords. The epithelium-rich type, which is

more frequently observed, consists mainly of cellular, fibrous connective tissue stroma where islands of odontogenic epithelium are an integral component.<sup>8,12</sup> Occasional foci of calcifications that consist of osteoid, dentinoid or cementum-like materials may occur.<sup>13</sup> Immunohistochemical staining or ultrastructure analysis cannot distinguish between the 2 types of COF.<sup>5</sup> As for our case, small nests of odontogenic epithelium that appeared entirely inactive were present in minimal amounts. There was a preponderance of fibrosis with some sparse calcifications. According to the histological findings, a final diagnosis of COF of simple type was retained. Despite this subtype is poorly documented in the literature, this is another case that confirms its existence.

Whether it is of simple or complex subtype, treatment of COF is conservative surgery. The best choice involves enucleation, since the lesion is easily removed, showing little bone adhesion and no tendency to undergo malignant transformation.<sup>12</sup> The prognosis of the lesion is usually good with very few cases of recurrence reported in the literature.<sup>7</sup> The cause of recurrence is thought to be incomplete removal of the lesion rather than the type of COF.

## Conclusion

This case report described a mandibular COF of simple type that occurred in a young female patient. Highlighting a subtype that was dropped from the last WHO classification of head and neck tumours is important to accumulate more information about this lesion and to show its different features. Despite its rarity, it should be included in the differential diagnosis of intrabony tumours of the jaws. These findings can better educate oral and maxillofacial surgeons about the unusual nature of this lesion, help establish a correct diagnosis and give the appropriate therapeutic management.

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## Declaration of conflicting interests

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## Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.

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## Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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