

A RECURRENT CASE OF CENTRAL ODONTOGENIC FIBROMA IN MAXILLA SHOWING ROOT RESORPTION: A CASE REPORT

FIBROMA ODONTOGÊNICO CENTRAL: RELATO DE CASO RECORRENTE E LOCALMENTE AGRESSIVO NA MAXILA

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ABSTRACT

Objective: The aim of this report is present an aggressive recurrent case of central odontogenic fibroma (COF) with tooth resorption in anterior maxilla, and discuss both events based on literature. **Case description:** A 29-year-old woman was referred for examination of a non-swelling intraosseous lesion detected by routine radiographic exams for orthodontic planning. Panoramic exam revealed a well-defined multiloculated radiolucency in the right anterior maxilla. Periapical radiography highlighted dental resorption of canine and first premolar. After the incisional biopsy the COF diagnosis was confirmed.

Tumor was removed by enucleation. Recurrence was detected three years later and the lesion was removed together with involved teeth. No indication of recurrence has been observed in the past eight years. **Conclusion:** COF is a benign tumor and can be aggressive. Recurrence and root resorption simultaneously are rare features reported in literature. The treatment must include tooth removal and curettage. Prognosis is good and follow-up is important.

KEYWORDS: Odontogenic Tumors; Fibroma; Recurrence; Root Resorption.

INTRODUCTION

Central odontogenic fibroma (COF) is a rare benign odontogenic tumor, derived from ectomesenchyme with variable cellularity¹. It represents 0.1-5% of all odontogenic tumors², affecting mostly females with a M:F ratio of 1:2.8. Age range from 11 to 66-year-old with a mean of 40¹. Mandible is more frequently affected mainly in the posterior region with 65%. In the maxilla, it occurs usually in the anterior region².

Clinically, it represents a slow-growing painless swelling often with cortical expansion². Radiographically, it appears as a well-defined unilocular radiolucency with sclerotic borders. Scalloping margins are detected in larger lesions¹. A varied amount of calcification may be also identified. Tooth displacement and association are reported^{3,4}. Root resorption are described in maxillary lesions. Curettage and enucleation are the treatments of choice. Recurrence is very rare⁴. Microscopically, COF is characterized by fibrous stroma with varying amounts of odontogenic epithelium cords. A higher amount of odontogenic islands are reported in recurrent tumors. Here, a local aggressive COF in the maxilla is presented with tooth resorption and recurrence is reported. Review of recurrent cases and root resorption are discussed.

CASE DESCRIPTION

A 29-year-old woman was referred for examination of a non-swelling intraosseous lesion that was detected by routine radiographic exams for orthodontic planning. No clinical symptoms

were registered. Panoramic radiography revealed radiolucent area in right anterior maxilla measuring 2 x 2 cm. Periapical radiography suggested a multilocular lesion with scalloping and well-defined borders, extending from the canine to first premolar. Dental resorption was identified (Figure 1A and B). Fine-needle aspirative biopsy was negative and incisional biopsy was performed. Microscopically, the lesion showed a few epithelial odontogenic rests in fibrous tissue (Figure 2A). Before surgery, endodontic treatment of canine and first premolar was executed due to negative pulp test vitality. Enucleation was the treatment of choice and related teeth were preserved. During surgical procedure, bone perforation of palatal cortical was observed. Recurrence was detected three years later and clinically appeared as an asymptomatic lesion and no evidence of swelling (Figure 3B). Panoramic radiograph revealed multilocular image recurrence in the same region extending to second premolar (Figure 3A). Enucleation and curettage was performed under general anesthesia and involved teeth were removed. Microscopic examination showed fibrous connective tissue of varying density with high cellularity (Figure 2B). No indication of recurrence has been observed in the past eight years of follow-up (Figure 4).

DISCUSSION

COF is a slow-growing, progressive, painless swelling often associated with cortical expansion and tooth displacement¹. Only sixteen cases of COF showing root resorption or recurrence have

been reported, summarized in Table 1. A local aggressive behavior is identified in only 11 cases, characterized by tooth resorption. These patients are usually younger with age ranging from 11 to 66-year-old - mean 36.3. Female is more affected with a M:F ratio of 1:4.5. Maxilla is slightly more involved in 54.5% and posterior region represents 66.6%. Radiographically, multiloculated radiolucency is registered in 72.7%. Treatment of choice includes enucleation and curettage with tooth removal. Differential clinical diagnosis of COF should include non-endodontic lesions in periapical regions, such as traumatic bone cyst, central giant cell tumor, odontogenic, non-odontogenic benign and malignant tumors^{11,12}.

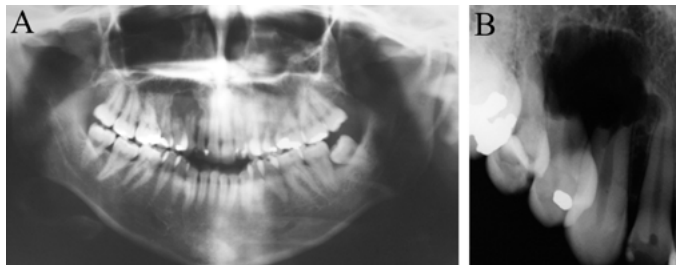


Figure 1 - Initial radiographs of COF. A, Radiolucency detected in the anterior maxilla with scalloping, well-defined borders. B, Root resorption involving canine and first premolar.

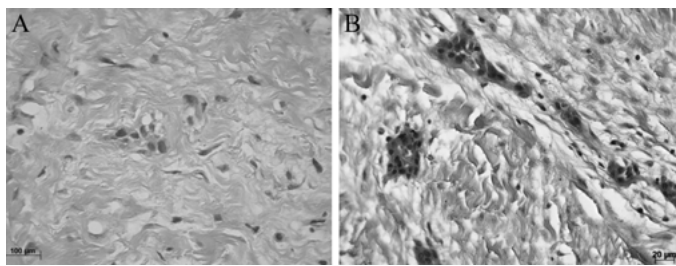


Figure 2 - Typical microscopic features of COF. A, Few epithelial odontogenic rests in a fibrous tissue. B, Note increase of odontogenic islands in the recurrent tumor.

Table 1 - Clinicopathological data of recurrent or root resorption COF

	Age/ Gender	Site	Signal and Symptoms	Radiografic finding	Tooth resorption	Initial treatment	Recurrence (years)
Present case	29/F	Maxilla, anterior	Asymptomatic	Multilocular	Yes	Enucleation/curettage, tooth maintenance	3
Hrichi <i>et al.</i> , 2012	38/F	Mandible, posterior	Swelling, pain	Multilocular	Yes	Enucleation/curettage, tooth removal	No
	11/F	Maxilla, posterior	Swelling, pain	Multilocular	Yes	Enucleation/curettage, tooth removal	No
	17/M	Mandible, posterior	Swelling	Unilocular	Yes	Enucleation/curettage, tooth removal	No
Melo <i>et al.</i> , 2010	16/M	Maxilla, anterior	Swelling	Unilocular	No	Enucleation with maintained tooth	5
Cercadillo <i>et al.</i> , 2006	51/F	Mandible, posterior	Swelling, teeth displacement	Multilocular	Yes	Enucleation/curettage, tooth removal	No
Ciconetti <i>et al.</i> , 2006	17/F	Maxilla, posterior	Swelling, teeth displacement	Unilocular	Yes	Enucleation/curettage, tooth removal	No
Daniels <i>et al.</i> , 2004	30/F	Mandible, posterior	Swelling, tooth mobility	Multilocular	Yes	Enucleation, tooth removal	No
Kinney <i>et al.</i> , 1993	66/F	Mandible, posterior	Swelling	Multilocular	No	Enucleation, tooth removal	1
Handlers <i>et al.</i> , 1991	41/F	Maxilla, posterior	Swelling	Multilocular	Yes	Enucleation/curettage	No
	46/F	Maxilla, anterior	Tooth mobility	Multilocular	Yes	Enucleation/curettage	No
	66/F	Mandible, posterior	Swelling	Multilocular	Yes	Enucleation/curettage	No
	39/F	Maxilla, posterior	Asymptomatic	Multilocular	Yes	Enucleation/curettage	No
Jones <i>et al.</i> , 1989	51/F	Mandibular, anterior	Swelling, paresthesia	Multilocular	No	Curettage	1
Svirsky <i>et al.</i> , 1986	45/F	Mandible, posterior	Asymptomatic	Unilocular	No	Curettage	2
Janssen <i>et al.</i> , 1985	44/M	Maxilla, anterior	Tooth mobility	Unilocular	Yes	Enucleation, tooth removal	No
Heimdal <i>et al.</i> , 1980	20/F	Mandible, posterior	Swelling	Unilocular	No	Enucleation, tooth removal	9

Recurrence was detected in only five cases summarized on Table 1. In these cases, age varies from 16 to 66-year-old with a mean age of 39.6. Female is more affected with a M:F ratio of 1:4. Posterior mandible is involved in 80% of cases and tumors usually appear as unilocular in 60%. Interestingly, tooth resorption is not reported in recurrent cases. Curettage is the treatment of choice in these reported cases and it may be associated with enucleation and tooth removal. Recurrences are registered after 1 to 9 years follow-up. Microscopically, recurrent lesion may show increase of cords and islands of odontogenic epithelium^{6,7}. Some authors associate recurrences with the



Figure 3 - Recurrent lesion detected after three years. A, Note a radiolucency at the same prior site. B, Advanced root resorption seen in the right canine.



Figure 4 - Panoramic radiograph. Eight years later.

Table 2 - Clinical features in recurrent, tooth resorption (present case included), and classical COF

	Recurrent COF	Tooth resorption-associated COF	Classical COF (WHO)
	n (%)	n (%)	n (%)
Total	6 (35.2)	12 (70.5)	-
Mean age (years)	37.8	35.7	40
Gender	F, 5 (83,3)	F, 10 (83,3)	F (64.2)
Site	Posterior mandible (80)	Maxilla (54.5), posterior (66.6)	Posterior mandible (84.6)
Signal and symptoms	Swelling (66.6)	Swelling (66.6), painless (83.4)	Swelling, painless
Radiographic findings	Multilocular (50)	Multilocular (75)	Unilocular (50)

non-capsulated characteristic limiting tumor removal during surgical procedures⁸.

The present case represents the first case of COF showing both tooth resorption and recurrence. Among all cases showing on table 1, this patient had no symptoms and the tumor was localized in anterior maxilla. Enucleation was the treatment of choice and recurrence was registered three years later. Table 2 summarizes clinical features in recurrent, tooth resorption, including the present case, and classical COF.

World of Health Organization defines COF as a rare neoplasm characterized by varying amounts of odontogenic epithelium embedded in a mature, fibrous stroma. Patients are mostly older with mean age of 40 and female predilection. Clinically, classical COFs appeared as a slow-growing, progressive and painless swelling, mostly in the mandible, with occasionally tooth displacement. Contrarily, pain was reported in two cases with tooth resorption, and paresthesia in one recurrent case. Radiographically, half of classical lesions are usually unilocular radiolucency with well-defined often sclerotic borders. However, multilocular radiolucency with scalloping margins was predominant only tooth resorption associated COF.

In the present case, the involved teeth were initially maintained, which could explain its recurrence and suggests the periodontal ligament as the origin of the lesion. Some tumors are associated with the crown of an unerupted tooth and suggests it was derived from the dental follicle and from the periodontal ligament. The treatment was extraction of the associated teeth and curettage of the lesion. No recurrence was observed in these cases⁹. Furthermore, the proximity to nerves hinders its removal, leaving tumor cells behind. Microscopically, the recurrent tumor showed a higher amount of odontogenic islands in a dense fibrous tissue as reported by other authors^{7,6,10}. Prognosis is favorable, although treatment should include removal of associated tooth to prevent recurrence.

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RESUMO

Objetivo: O objetivo deste relato é apresentar um caso clínico de fibroma odontogênico central (FOC) recorrente e com reabsorção dental em região anterior de maxila, e discutir estes dois eventos baseados na literatura. **Descrição do Caso:** Paciente do sexo feminino, 29 anos, foi encaminhada para avaliação de lesão intraóssea não-expansiva, detectada por exames radiográficos solicitados para planejamento ortodôntico. Exames

radiográficos mostraram uma área radiolúcida multiloculada bem delimitada com reabsorção radicular de canino e primeiro pré-molar direito. Após biópsia incisiva e exame histopatológico, a lesão foi diagnosticada como fibroma odontogênico central. O tumor foi enucleado e três anos depois, detectou-se recorrência. A lesão recorrente foi removida junto aos dentes adjacentes. Nenhuma recorrência foi observada nos últimos oito

anos de acompanhamento. Conclusão: FOC é um tumor benigno que pode apresentar comportamento agressivo. Recorrência e reabsorção radicular simultâneas são características raramente relatadas na literatura. O tratamento deve incluir exodontia e

curetagem do sítio cirúrgico. O acompanhamento é importante, embora seja uma lesão de prognóstico favorável.

PALAVRAS-CHAVE: Tumores odontogênicos; Fibroma; Recorrência; Reabsorção radicular.

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