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 cellularity. It represents 0.1-5% of all odontogenic tumors, affecting mostly females with a M:F ratio of 1:2.8. Age range from 11 to 66-year-old with a mean of 40. Mandible is more frequently affected mainly in the posterior region with 65%. In the maxilla, it occurs usually in the anterior region. Clinically, it represents a slow-growing painless swelling often with cortical expansion. Radiographically, it appears as a well-defined unilocular radiolucency with sclerotic borders. Scallopings margins are detected in larger lesions. A varied amount of calcification may be also identified. Tooth displacement and association are reported. Root resorption are described in maxillary lesions. Curettage and enucleation are the treatments of choice. Recurrence is very rare. 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 displacement. 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.

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%. Intesting, 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

Table 1 - Clinicopathological data of recurrent or root resorption COF

<table>
<thead>
<tr>
<th>Age/Gender</th>
<th>Site</th>
<th>Signal and Symptoms</th>
<th>Radiographic finding</th>
<th>Tooth resorption</th>
<th>Initial treatment</th>
<th>Recurrence (years)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Present case</td>
<td>29/F</td>
<td>Maxilla, anterior</td>
<td>Asymptomatic</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage, tooth maintenance</td>
</tr>
<tr>
<td>Hirchi et al., 2012</td>
<td>38/F</td>
<td>Mandible, posterior</td>
<td>Swelling, pain</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage, tooth removal</td>
</tr>
<tr>
<td>11/F</td>
<td>Maxilla, posterior</td>
<td>Swelling, pain</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage, tooth removal</td>
<td>No</td>
</tr>
<tr>
<td>17/M</td>
<td>Mandible, posterior</td>
<td>Swelling</td>
<td>Unilocular</td>
<td>Yes</td>
<td>Enucleation/curettage, tooth removal</td>
<td>No</td>
</tr>
<tr>
<td>Melo et al., 2010</td>
<td>16/M</td>
<td>Maxilla, anterior</td>
<td>Swelling</td>
<td>Unilocular</td>
<td>No</td>
<td>Enucleation with maintained tooth</td>
</tr>
<tr>
<td>Cercadillo et al., 2006</td>
<td>51/F</td>
<td>Mandible, posterior</td>
<td>Swelling, teeth displacement</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage, tooth removal</td>
</tr>
<tr>
<td>Ciconnetti et al., 2006</td>
<td>17/F</td>
<td>Maxilla, posterior</td>
<td>Swelling, teeth displacement</td>
<td>Unilocular</td>
<td>Yes</td>
<td>Enucleation/curettage, tooth removal</td>
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<td>Daniels et al., 2004</td>
<td>30/F</td>
<td>Mandible, posterior</td>
<td>Swelling, tooth mobility</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation, tooth removal</td>
</tr>
<tr>
<td>Kinney et al., 1993</td>
<td>66/F</td>
<td>Mandible, posterior</td>
<td>Swelling</td>
<td>Multilocular</td>
<td>No</td>
<td>Enucleation, tooth removal</td>
</tr>
<tr>
<td>Handlers et al., 1991</td>
<td>41/F</td>
<td>Maxilla, anterior</td>
<td>Swelling</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage</td>
</tr>
<tr>
<td>46/F</td>
<td>Maxilla, anterior</td>
<td>Tooth mobility</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage</td>
<td>No</td>
</tr>
<tr>
<td>66/F</td>
<td>Mandible, posterior</td>
<td>Swelling</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage</td>
<td>No</td>
</tr>
<tr>
<td>39/F</td>
<td>Maxilla, posterior</td>
<td>Asymptomatic</td>
<td>Multilocular</td>
<td>Yes</td>
<td>Enucleation/curettage</td>
<td>No</td>
</tr>
<tr>
<td>Jones et al., 1989</td>
<td>51/F</td>
<td>Mandibular, anterior</td>
<td>Swelling, paresthesia</td>
<td>Multilocular</td>
<td>No</td>
<td>Curettage</td>
</tr>
<tr>
<td>Svirsky et al., 1986</td>
<td>45/F</td>
<td>Mandible, posterior</td>
<td>Asymptomatic</td>
<td>Unilocular</td>
<td>No</td>
<td>Curettage</td>
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<tr>
<td>Janssen et al., 1985</td>
<td>44/M</td>
<td>Maxilla, anterior</td>
<td>Tooth mobility</td>
<td>Unilocular</td>
<td>Yes</td>
<td>Enucleation, tooth removal</td>
</tr>
<tr>
<td>Heimdal et al., 1980</td>
<td>20/F</td>
<td>Mandible, posterior</td>
<td>Swelling</td>
<td>Unilocular</td>
<td>No</td>
<td>Enucleation, tooth removal</td>
</tr>
</tbody>
</table>
non-capsulated characteristic limiting tumor removal during surgical procedures.

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 Heath 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 uniloculated 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 cases. 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.

Prognosis is favorable, although treatment should include removal of associated tooth to prevent recurrence.

REFERENCES


RESUMO


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

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