Giant ameloblastic fibroma

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ABSTRACT. Background A 10 year-old male was referred to our department for a delay in the eruption of his lower canine, premolars and molars on the right side. The panoramic radiograph showed a multilocular radiolucent lesion approximately 3.5 x 7.5 cm in diameter including the right canine, first and second premolar and second mandibular molar. The lesion was clinically diagnosed as dentigerous cyst. It was enucleated via curettage of the bone bed and diagnosed as ameloblastic fibroma at the histopathological examination. After twenty-one months, radiographs showed that the surgical defect had filled with new bone.

Key words: Ameloblastic fibroma; Paediatric fibroma; Giant cells fibroma.

Introduction

Ameloblastic fibroma (AF) is one of the rare malformations classified as a subtype of benign mixed odontologic tumor by the World Health Organization WHO [Nouri et al., 2007; Mohapatra et al., 2000], and it is composed of both epithelial and mesenchymal elements [Mohapatra et al., 2000]. It is a relatively rare tumor and its worldwide prevalence is reported between 0.3% and 6.5% of all odontogenic tumours [Garcia-Pola Vallejo et al., 2001; Taiwo Adebayo et al., 2005; Buchner et al., 2006; Jing et al., 2007], whereas one study demonstrated its prevalence as 1.5% of all odontogenic tumors diagnosed in Istanbul [Olgac et al., 2006]. AF is commonly found in the younger age groups, between 15 and 25 years of age and males are more affected than females with the ratio of 1.26:1 [Mcguinness et al., 2001; Dallera et al., 1996; Chen et al., 2007]. Most of the AFs occur in the mandible and especially in the posterior region of the dental arch, with the molar area predominant over the premolar region [Mohapatra et al., 2000]. The tumour grows slowly and painlessly while it expands the jaws [Mcguinness et al., 2001, Dallera et al., 1996]. Radiographically, it appears as a unilocular area of radiolucency with a smooth outline [Mcguinness et al., 2001]. It can vary in size 1 to 8.5 cm, but only a few cases of AF bigger then 4 cm in size have been reported [Mohapatra et al., 2000].

The differential diagnosis should be made with other odontogenic tumors and cysts. It is important to differentiate AF from ameloblastoma; it does not exhibit a locally invasive growth pattern and the stellate cells are much less abundant [Mohapatra et al., 2000]. The lesion is called ‘peripheral AF’ when the classic microscopic features of central ameloblastic fibroma are present but no bony involvement [Abughazaleh et al., 2008]. As therapy, simple excision of AF is adequate in most of the cases [Mcguinness et al., 2001]. The 5-year and 10-year recurrence rates are 41.6% and 69.2%, respectively [Chen et al., 2007]. AFs rarely progress into malignant lesions like ameloblastic fibrosarcoma or ameloblastic carcinosarcoma [Kobayashi et al., 2005; DeLair et al., 2007]. Related case reports are shown in table 1 [Mohapatra et al., 2000; Dallera et al., 1996; Kobayashi et al., 2005; DeLair et al., 2007; Abughazaleh et al., 2008; Kusama et al., 1998; Naidoo et al., 1998; Usubitüren et al., 2002; Martin-Granizo-Lopez et al., 2003; Chindia et al., 2005]. As seen in the table only a few cases of AF were seen in the maxilla [Abughazaleh et al., 2008; Chindia et al., 2005].

We present a case of giant AF in a 10 years old male boy treated by conservative procedures including enucleation, curettage and simple excision.

Case report

A 10-year-old male was referred to our department for a delay in the eruption of his lower canine, premolars and molars on the right side. There was no history of systemic disease, trauma or pain. The extraoral examination revealed a normal face structure without swelling or expansion. Cervical lymphadenopathy was not detected. The intraoral clinical examination revealed an expansion of the right posterior part of the mandible. There was a light pink tissue around the lesion area (Fig. 1). His oral hygiene was good and the occlusion was not affected by the
lesion. The panoramic radiograph showed a radiolucent lesion, approximately 3.5 x 7.5 cm in diameter, including right canine, first and second premolar and second molar of the mandible (Fig. 2). These findings suggested a dentigerous cyst and the lesion was clinically diagnosed as dentigerous cyst. Enucleation and curettage of the lesion was planned and the patient and his family members were informed about the procedure. The lesion was enucleated through a full mucoperiosteal triangular flap under local anesthesia using 1% lidocaine. Soft and cold food was recommended on the day of surgery to protect the area from oedema. Antibiotics and analgesics were prescribed. Follow-up examinations were performed on the second, fifth and ninth day after the operation. The patient reported a mild pain in the early postoperative period. Healing was uneventful. No recurrence was observed over the follow-up period of 21 months.

The tumor measured 3.0 x 3.7 x 7.5 cm in its largest dimension, exhibiting a round, well circumscribed grayish myxoid mass surrounded by a thin transparent capsule. The density of the lesion was gelatinous. The microscopic examination revealed odontogenic epithelial and connective tissue components. The epithelium was composed of ameloblastic-like cells surrounding stellate reticulum resembling cells. These epithelial cells were clearly circumscribed by a basal membrane and formed islands; strands and leaf-like proliferations in cellular fibrous stroma (Fig. 3). The border was not infiltrative and the neoplasm was focally rimmed by a well vascularized, loose connective tissue. A final diagnosis of ameloblastic fibroma was made.

**Discussion**

Ameloblastic fibroma and related lesions are defined as ‘neoplasms composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles the dental papilla, and with varying degrees of inductive change and dental hard tissue formation’ [Takeda, 1999]. It is believed that ameloblastic fibroma may be a true mixed tumor, in which the epithelial and the ectomesenchymal elements are neoplastic [Takeda, 1999].

The long-term follow-up of AF is very important due to its high recurrence rate and its rare but potential risk of carcinogenesis [DeLair et al., 2007]. The treatment of AF is rather controversial with conservative surgical enucleation and curettage being preferred [Chindia et al., 2005].

It is the opinion of the present authors that the high recurrence rate of the AF is a remarkable feature of this lesion compared with other benign odontogenic tumors, although it is generally believed that most of the recurrences occur due to the incomplete resections of the odontogenic tumors. The present case was followed-up for almost two year and there was no clinical or radiographic evidence of recurrence 21 months after surgery.

The present case of AF shows characteristics similar
<table>
<thead>
<tr>
<th>Author / Year</th>
<th>Age-Gender</th>
<th>Location</th>
<th>Treatment</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>P. Dallera et al. (1996)</td>
<td>4 female and 2 male patients aged 7-47 (average 18,5)</td>
<td>5-in the molar regions of the mandible</td>
<td>5- enucleation + curettage 1- segmental resection</td>
<td>Between 4-35 years No recurrence</td>
</tr>
<tr>
<td>K. Kusama et al. (1998)</td>
<td>40 years old female</td>
<td>Right premolar area of the mandible</td>
<td>Lesion and part of the alveolar bone were surgically removed</td>
<td>No evidence of recurrence 4 years 6 months after surgery</td>
</tr>
<tr>
<td>L. Chris et al. (1998)</td>
<td>15 months old child</td>
<td>Left molar area of the mandible</td>
<td>Enucleation</td>
<td>After a year the family moved to another region, no long term follow-up</td>
</tr>
<tr>
<td>P.K. Mohapatra et al. (2000)</td>
<td>15 years old female</td>
<td>Symphysys of the mandible</td>
<td>Both canines are extracted and the lesion is surgically removed with an envelope type of incision</td>
<td>No recurrence 3 years after surgery</td>
</tr>
<tr>
<td>A. Usubütün et al. (2002)</td>
<td>17 years old male</td>
<td>Right premolar area of the mandible</td>
<td>Enucleation and extraction of the associated unerupted tooth</td>
<td>No recurrence 18 months after surgery</td>
</tr>
<tr>
<td>Martin-Granizo-Lopez et al. (2003)</td>
<td>9 years old male 8 years old male</td>
<td>Left molar area of the mandible Right molar area of the mandible</td>
<td>Resection of the lesion Resection of the lesion Right molar area of the mandible</td>
<td>No recurrence 12 years after surgery No recurrence 1 year after surgery</td>
</tr>
<tr>
<td>M.L. Chinda et al. (2005)</td>
<td>4 male and 3 female patients (10-22 years)</td>
<td>6-in the molar region of the mandible One in the maxillary molar area</td>
<td>Enucleation and curettage of the lesions</td>
<td>No recurrence from several months to 8 years</td>
</tr>
<tr>
<td>K. Kobayashi et al. (2005)</td>
<td>26 years old male</td>
<td>Left posterior region of the mandible</td>
<td>Surgical curettage of ameloblastic fibroma Afterward a left mandibulectomy RT and CT</td>
<td>2 years later an ameloblastic fibrosarcoma was seen, 5 years after initial operation the patient died</td>
</tr>
<tr>
<td>D. DeLair et al. (2007)</td>
<td>19 years old female</td>
<td>Left posterior region of the mandible</td>
<td>Left hemimandibulectomy because AF changed to ameloblastic carcinosarcoma</td>
<td>No recurrence 2 years after surgery</td>
</tr>
<tr>
<td>K. Abughazaleh et al. (2008)</td>
<td>3-year-old girl</td>
<td>Right anterior region of the maxilla</td>
<td>Excision and curettage</td>
<td>No recurrence 10 months after surgery</td>
</tr>
</tbody>
</table>


To other cases reported in the past, with regard to age distribution, gender, size, location, symptoms, radiographical and pathological features, treatment and follow-up information.

An interesting point is that the patient and his family did not seek medical advice for a long time, notwithstanding the swollen region in the mouth, until they noticed a delay in the eruption of his inferior canine, premolars and molars on the right side when compared with the left side of the mandible. The reasons were that the patient had felt no pain and his family accepted the swelling as a normal course of
eruption of the teeth until they noticed that eruption of
the teeth in the left side was faster.

Conclusions
AF could frequently be misdiagnosed as a
dentigerous cyst due to their similar radiographical and
clinical appearance. The same happened in our case
because of the association of the tumor with unerupted
 teeth, radiologically, and features such as like age
distribution, gender, symptoms, size and location,
clinically. Due to these similarities, the importance of
the histopathological investigation of every lesion
should never be neglected.

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