Case Report

Recurrence of Multicystic Ameloblastoma: Case Report

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Introduction: Ameloblastoma is classified by the WHO as a benign epithelial odontogenic tumor without odontogenic ectomesenchyma with locally invasive behavior and a high recurrence rate. Represents 1% of oral tumors, primarily affecting the posterior mandible. Generally asymptomatic, slow growing, can punch cortical bone, teeth and reabsorb move and cause facial asymmetry. Can be classified according the clinical and radiographic aspects in solid or multicystic ameloblastoma, cystic and peripheral.

Objectives: To report the diagnosis and treatment of multicystic ameloblastoma with marginal resection of the mandible. Patients and methods: Patients 34 years referred to the Department of Stomatology, School of Dentistry of Bauru - USP for evaluation of multiloculated radiolucent lesion involving the body, angle and ramus rights. Clinically, the patient was covered with mucosa of normal color and no facial asymmetry. Marsupialization of the lesion was performed and biopsy diagnosed with Granuloma Pulse. After 4 months, was observed regression of the lesion. One year postoperatively, the patient returned with recurrence of the lesion was performed another biopsy which revealed the histopathological examination that it was ameloblastoma. The patient underwent surgical respective and reconstructive plate fixation for maintenance of the bed. Results: After 18 months of surgery were not observed signs of clinical or radiographic. Conclusions: The interpretation of clinical data and imaging of patients with ameloblastoma is essential for early diagnosis and effective treatment of the injury. Follow up for long periods is an essential part of treatment considering their local aggressiveness and high recurrence rate.

Keywords: Ameloblastoma; Mandible; Diagnosis.

INTRODUCTION

Ameloblastoma is an odontogenic tumor characterized by slow growth, but locally invasive and infiltrative (Scholl et al., 1999; Bataineh 2000; Grempel et al., 2003). It has a destructive potential and the combination of proliferative lesions to malignant transformation have been documented (Gilijamse et al., 2007; Gomes et l., 2006). Its etiology is not completely understood, but it is estimated that originates from the remaining tooth lamina, reduced enamel epithelium, epithelial cell rests of Malassez or the basal cells of the epithelial surface layer (Gomes et l., 2006; Neville et al., 2004). No predilection is shown for gender or race and affects predominantly the mandible (80-85 % of cases) (Silva et al., 2004).

The solid or multicystic ameloblastoma is a subtype that shows a greater propensity to infiltrate the surrounding tissues showing a higher recurrence rate, requiring rapid and precise approach in implementation of treatment (Scholl et al., 1999; Curi et al., 1997). Thus, once the diagnosis is established, the treatment plan is drawn from

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the clinical and radiographic findings, and may also be aided by CT and MRI (Scholl et al., 1999; Som and Bergeron 1991).

By presenting single biological behavior, there is controversy about the best way to treat ameloblastomas. Several therapeutic modalities have been proposed, including curettage, marsupialization, cryosurgery, electrocautery, sclerotherapy and radiotherapy, but classically, solid ameloblastomas are treated, in most cases, by radical surgical excision with 1-2 cm safety margin beyond the limits of radiographic lesion (Scholl et al., 1999; Bataineh 2000; Pogrel and Montes 2009).

Radical techniques bring with them serious inconvenience to the patient, including masticatory dysfunction, alteration of mandibular movements, facial mutilation and deformity (Chem et al., 2005). Treatment should include rehabilitation, restoring the functional, anatomical and aesthetic capacity, as well as assist in their reintegration in society (Zemann et al., 2007).

This study aims to report the case of multicystic ameloblastoma treated with marginal resection of the mandible and discuss relevant issues regarding the diagnosis and treatment of this condition.

CASE REPORT

A 34-year-old patient was referred to the service of Stomatology of Bauru School of Dentistry – USP, for evaluation of multilocular radiolucent lesion involving the body, angle and the right mandibular ramus. Clinically, it was covered with mucosa of normal color and no facial asymmetry. Marsupialization was performed for obtaining material for biopsy and the diagnosis was pulp granuloma. After 4 months, there was a regression of the lesion. In the clinical and radiographic postoperative control one year later, it was observed multicellular radiolucent area in the mandibular angle. New biopsy was performed which by histopathology examination revealed to be an ameloblastoma. The patient underwent a surgical procedure via three-dimensional resection margin including higher cortical and medullary portion of the mandibular body and ascending ramus, preserving the base of the mandible and the posterior region of the ascending ramus. It was performed fixation of 2.4 mm reconstruction plate system overlapping the lower residual cortical bone for maintenance of the bed.

Postoperative controls were conducted every three months and 20 months after the surgery, the patient returns in postoperative control with a radiolucent area of approximately 6 mm in diameter above the area subjected to resection. It was performed excisional biopsy followed by electrocoagulation.

DISCUSSION

The ameloblastoma is classified by the WHO as a benign epithelial odontogenic tumor without ectomesenchyma with locally invasive behavior and a high recurrence rate. It represents 11-18 % of all odontogenic tumors primarily affecting the posterior mandible (Neville et al., 2004; Gardner 1996). Generally asymptomatic, slow growing, it can perforate the cortical bone of teeth, resorb and cause facial asymmetry (Scholl et al., 1999).

Ameloblastomas are classified into unicystic, multicystic (solid), peripheral and malignant subtypes, being the conventional solid or multicystic (86 % of cases), unicystic (13 % of cases) and peripheral or extra-osseous (about 1 % of cases) (Neville et al., 2004). Such distinction is important, as it helps in the treatment choice.

Conventional ameloblastoma tends to infiltrate in the bone trabeculae of cancellous bone in the periphery of the lesion, prior to bone resorption being visible radiographically. Therefore the true tumor margin often extends beyond the apparent clinical or radiographic margin (Bataineh 2000).

The best form of treatment has been controversial for several years. Radiotherapy is not indicated because the lesion is radioresistant. There is also an indication in the literature of electrocautery, cryosurgery and injection of sclerosing agents as alternative treatments (Som and Bergeron 1991).

A high potential for recurrence (60-80 %) after simple treatment with enucleation or curettage is frequently observed, justifying the choice of professionals for more aggressive treatments (Pogrel and Montes 2009). An adequate alternative for the treatment of small lesions of multicystic ameloblastoma is the marginal resection which is widely used, but due to the recurrence rate of approximately 15 %, it is recommended that the safety margin should be at least 1 cm beyond the radiographic limits (Neville et al., 2004).

Evaluating several studies in order to compare the recurrence rate of treatments with complete and marginal resection, no significant difference was observed between the approaches (Table 1).

The advantages of less extensive surgical treatments such as marginal resection of the mandible are significant, improving the patients prognosis. Facial deformity, masticatory dysfunction, abnormal jaw movement and the need for further reconstructive surgeries are consequences often associated with treatments with complete resection and can often be minimized with the use of marginal resection (Pogrel and Montes 2009; Nakamura et al., 2002). However, in cases of extensive lesions affecting great part of the maxillary cortical bone or mandibular lesions by anatomic considerations suggests a more
Table 1. Comparison of recurrence rate between the treatments with partial and total resection.

<table>
<thead>
<tr>
<th>Authors</th>
<th>Partial resection</th>
<th>Recurrence (%)</th>
<th>Total resection</th>
<th>Recurrence (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hatada et al, 2001</td>
<td>X</td>
<td>68</td>
<td>X</td>
<td>18</td>
</tr>
<tr>
<td>Batineh, 2000</td>
<td>X</td>
<td>23</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Takahashi, Miyachi, Sato, 1998</td>
<td>X</td>
<td>2</td>
<td>+</td>
<td>+</td>
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<td>Chidzonga, 1996</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Olaitan, Adeola, Adekeye, 1993</td>
<td>X</td>
<td>61</td>
<td>0,6</td>
<td>X</td>
</tr>
<tr>
<td>Nakamura, 2002</td>
<td>+</td>
<td>+</td>
<td>+</td>
<td>+</td>
</tr>
<tr>
<td>Hong et al, 2007</td>
<td>X</td>
<td>43</td>
<td>11,6</td>
<td>X</td>
</tr>
<tr>
<td>Beceli et al, 2002</td>
<td>X</td>
<td>27</td>
<td>X</td>
<td>15</td>
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</table>

Figure 1. Initial panoramic radiograph.

Figure 2. Radiograph showing the recurrence of the lesion.
Figure 3. Panoramic radiograph aftermarsupialization.


Figure 5. Postoperative radiograph (1 year).
aggressive approach with segmental resection (without maintaining bone continuity) including the periosteum and overlying soft tissues (Ferretti et al., 2000; Rosenstein et al., 2001; Müller and Stootweg 1985).

The interpretation of clinical and imaging data of patients with ameloblastoma is essential for early diagnosis and effective treatment of injury. The therapeutic decision must target the elimination of pathology considering the morbidity of treatment method and life quality in the rehabilitation of patients.

The long-term postoperative follow-up with imaging is essential because, although more than 50% of recurrences happen within the first five postoperative years, the injury has the potential to develop late recurrences (Bataineh 2000; Som and Bergeron 1991). Due to its slow growth, the reappearance of the lesion may occur after many years or even decades from the first surgery and when this happens, the lesions tend to exhibit a greater potential for invasion and bone destruction than the original injury (Ferretti et al., 2000).

CONCLUSIONS

Treatment by marginal resection with a safety margin is a viable alternative in cases of multicystic ameloblastoma in the mandible, to avoid mutilation by a more radical treatment. In the case described, the conservative approach proved effective so far, but a long-term follow-up is an essential part of treatment considering their local aggressiveness and high recurrence rate.

REFERENCES


