Abstract

Ameloblastomas are benign odontogenic tumors that exhibit a local malign behavior. Case report: A recurrence of an ameloblastoma occurring after 33 years of hemimandibulectomy in a 69-year-old woman is reported. The tumor arose as a painless mass. Clinical exam showed a lesion placed in the right third molar region. The right mandible was absent from the bicuspid area to the condyle in radiography and computed tomography. After excision, microscopic features diagnosed an ameloblastoma. The patient is under close follow-up and shows no recurrence or metastasis. Conclusion: Despite the radical initial treatment, the tumor showed an uncommon behavior and recurred.

Keywords: Ameloblastoma. Odontogenic tumor, squamous. Recurrence. Mandibular neoplasms. Surgery.

Introduction

Ameloblastoma is known to be the most significant odontogenic tumor.1 It was first described by Broca in 1868 and named by Churchill.2 The tumor develops from odontogenic epithelium and tooth development cell remnants1,3 and is known to be a benign tumor with a high tendency to recur after inadequate treatment.1,3-6

We describe an unusual recurrence of an ameloblastoma after 33 years of hemimandibulectomy.

Case Report

A 69-year-old woman was referred to the Oral and Maxillofacial Surgery Service at Santa Casa de Misericórdia Hospital, Juiz de Fora, Minas Gerais, Brazil having as her main complaint a right intra-oral swelling with mild pain. The patient’s history showed she underwent a right partial hemimandibulectomy without any reconstruction 33 years earlier. Since then, no other treatment was performed. The lesion was diagnosed, according to the patient, as an ameloblastoma. No exam regarding the first surgery was presented.

Physical examination showed a lack of the right mandible and a round and large swelling in the third molar region. The swelling was tender and painless to
The overlying mucosa was clinically intact. An incisional biopsy was performed showing histological features of ameloblastoma. Conventional radiography was done, including panoramic (Figure 1) and mandibular posteroanterior projections. Computed tomography (CT) was performed in axial and coronal views (Figure 2 and Figure 3). Findings showed a lack of the right mandible from the second premolar to the condyle. CT showed a large round soft tissue mass with no osseous borders. The patient showed no interest in mandibular reconstruction, but only in tumor removal. Under general anesthesia the patient was submitted to dissection and excision of the lesion (Figure 4). Surgical exploration showed a partial encapsulated tumor. The wounds were closed with absorbable sutures. Healing was uneventful and the patient was discharged home after 5 days.

The specimen was immersed in 10% formalin. Macroscopically the mass was about 4,0 x 4,0 cm (Figure 5). Histological examination showed an ameloblastoma. There were three microscopically patterns recognized as follicular (Figure 6), plexiform and granular cell types. A trace of bone matrix, in only one microscopic field, was observed.

After a year the patient is still under clinical and radiographic follow-up and shows no recurrence or metastasis (Figure 7).
Solid and multicystic ameloblastomas exhibit no gender predilection. The age range is 30–60 years, and in 80% of the cases the tumor occurs in the posterior mandibular area. The lesion spreads by infiltration and may resorb cortical plates and reach soft tissues. Histologically, solid and multicystic ameloblastomas can be divided in two forms commonly diagnosed: the follicular and the plexiform. Other patterns, less common, are basal cell, granular cell and acanthomatous.\(^3\)

In a recent study of some selected odontogenic tumors, the immunoexpression of calretinin, a calcium-binding protein, was detected only in ameloblastomas. It was postulated that this protein could regulate apoptosis and its expression might have a role in the transition of odontogenic remnants in ameloblastomas. In addition, it was hypothesized that ameloblastomas behavior could be related to this protein as it was not present in other odontogenic neoplasms.\(^7\)

Ameloblastomas are expected to recur after curettage in 50% to 90%.\(^1,4,6\) Other modalities of surgery, as marginal resection and radical treatment with the inclusion of adjacent soft tissues, account for 5% to 15% of recidivation.\(^1,6\) The treatment of choice, mainly when the lesion reaches the cortical plates and soft tissues, is wide resection. Bone grafts can repair the defect. Recurrence after radical surgery and reconstruction is very rare but can develop from the stumps, soft tissues and intraoperative contamination.\(^5\)

Al-Bayaty et al.\(^4\) believed that soft tissue recidivation of ameloblastomas was rarer than in bone grafts. They described a case of a temporal mass in a 32-year-old woman. She underwent a right hemimandibulectomy with costocondral reconstruction in 1992. After 6 years of the first surgery a recurrence was found causing facial deformity. The lesion was surgically exposed and dissected from the temporalis muscle revealing microscopic features of an ameloblastoma. The authors believed that the recrudescence of this neoplasm was due to tumor cell implantation in the site.

A similar case was documented by To et al.\(^6\) A 45-year-old woman developed a tender temporal mass 30 years after the first treatment. Her medical history revealed local enucleations and curettage due to recurrences of an ameloblastoma. When she was 35 old, the patient was submitted to a right hemimandibulectomy. The site was reconstructed with a rib graft. 10 years after mandibular resection a right tender temporal mass developed. By a hemicoronal flap, the neoplasm was removed and diagnosed as an ameloblastoma. The hypothesis was that an infratemporal tumor seeding, during intraoral surgery, could explain the temporal recidivation.

Although rarely expected, soft tissues recurrence is a possibility because of tumor implantation. Our might explain this case as a recurrence by seeding. The neoplasm might have recurred in the same site of the first involvement. However, we can not prove this due to a lack of data or exams from the first surgery. Another
fact that remains to be elucidated is the trace of bone trabecula in one microscopic field. It is then difficult to say whether the bone was remnant of the first surgery or developed after it.

The terms metastasizing ameloblastoma and ameloblastoma with atypia are subjected to doubts as concerns the cases they describe. The first disease does not differ from jaw ameloblastoma but its metastasis may appear in other sites such as the lungs. The second one can be defined as an ameloblastoma with malignant cytological features. In this case the tumor is named ameloblastic carcinoma. As regards genetics, Kumamoto et al. stated that aberrations in the p53 pathway (p14ARF-MDM2-p53 cascade), related to cell cycle regulatory system, is correlated to neoplastic transformation. Ameloblastomas and metastasizing ameloblastomas showed very similar nuclear expression of gene p53. However, ameloblastic carcinomas present a marked increased p53 reactivity suggesting malignant transformation of odontogenic epithelium.

Ciment presented a case report of a metastatic ameloblastoma after 29 years of a hemimandibulectomy of a 55-year-old-man. The patient was asymptomatic. Lung lesions were found after chest exams for cardiac function. He suspected of granulomas despite the lack of symptoms. A diagnostic thoracotomy identified a metastatic malignant ameloblastoma. According to the author, the lung is the most common site of metastases (80%), followed by regional lymph nodes, pleura and other organs.

As 2% to 5% of ameloblastomas do metastasize, we carefully submitted our patient to clinical exams and perform maxillofacial and chest radiographies. Until now, there are no changes in the patient’s medical status.

Conclusion

The surgical approach to ameloblastomas must include a close follow-up of the patient because of possible recidivation at any time. We believe the case presented is sui generis. Despite the radical initial treatment the tumor showed an unusual behavior and recurred.

References