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Case Report

Ameloblastoma: A Case Report

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Abstract

Ameloblastoma is a neoplasm arising from the epithelium involved with the formation of teeth. They are usually benign, locally aggressive and recurrent, however, metastases are rare. The treatment is not clearly defined and the main therapeutic tool is surgical intervention. Radiotherapy and chemotherapy have not been shown encouraging results. As there are few cases reported in the medical literature and the treatment is challenging, we reported a case of a 53-year-old woman with recurrent ameloblastoma, with local and distant recurrence, that remains alive over 25 years from diagnosis.

Keywords: Ameloblastoma. Recurrence. Chemotherapy. Radiotherapy. Radiation therapy. Surgery.

Introduction

Ameloblastoma is a tumor originated from the epithelium involved with the formation of teeth. It has aggressive behavior and recurrent course, but it is rarely metastatic.[1] Ameloblastoma represents 1% of all tumors and cysts that involve maxillomandibular area[1-6] and about 10% of odontogenic tumors.[7]

The term ameloblastoma was suggested by Ivy and Churchill in 1934[8] based on this odontogenic epithelial etiology.[9] However, the first report of this neoplasm was done by Broca, in 1868.[8] About 80% of cases are found in jaw, and less frequently on maxilla.[10]

Ameloblastoma can occur in any age, mainly on third and fourth decades, in both genders, although some authors report a predominance in female group.[1,11-12]. According to race, there are some reports showing higher incidence in black people.[12-15]

Csiba et al (1970)[16] tried to show the relation between ameloblastoma and some risk factors as: non-specific irritating factors (dental extractions, caries, trauma, infections or sore), dietary deficit and viral pathogenesis.

The diagnosis is done in routine odontological exam, because this disease usually is asymptomatic. Patients may present with pain, paresthesia, bacterial infection and fractures due to loosening of teeth tissue[11]. Differential diagnostic include reparative giant cells granuloma, odontogenic mixoma, multilobular follicular cyst and reverse papiloma.[17-18]

Ameloblastoma can be classified in three types, considering clinical and radiographic features: solid or multicystic (86% of cases), unicystic (13% of cases) and peripherical (1% of cases).[19]

There are a lot of histological subtypes of ameloblastoma: plexyform, follicular, unicystic, basal cells, granulous cells, clear cells, acantomatous, vascular and

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desmoplastic.[12,20] The most common histological patterns are plexiform and follicular. There are no data that prove the relation of the subtypes with clinical course.

Surgical treatment involves enucleation, curettage, partial resection as well as hemimandibulectomy.[21-22] Recurrence after curettage and enucleation occurs in 55% to 90% of the cases.[12,23] Other options of treatment are cryosurgery, eletrocautherization, esclerotherapy and radiotherapy.[3]

Radiotherapy is indicated in few situations due to radio resistance of this malignancy. It can be considered for unresectable tumors and in post operative setting when the surgical margins are positive.[24-26] Neoadjuvant radiotherapy has been suggested in an attempt to shrink the tumor and allow surgery later, when the initial surgery can cause facial esthetical deformity and mastication difficulties.

Chemotherapy is not effective[27] and should be reserved for palliative circumstance when the radiotherapy and the surgery are not feasible [27-29].

Surveillance of these patients is essential because of high rates of recurrence. Some cases of bone,[30] pulmonary and cervical nodal metastasis have been described in literature.[27,31-32]

Case Report

A 53 year-old female patient presented at Dentistry Department of our hospital with history of repeated gingivobuccal sore for 2 years. On initial evaluation a biopsy of buccal mucosa was performed and showed blastoma.

After two years of the diagnosis, the patient related dysphagia and ulcerated nodule in the left retromolar trigone. Patient was submitted to left hemimandibulectomy (the neoplasm involved both bone and soft tissue). Anatomophatologic analysis revealed ameloblastoma with positive margins.

Another recurrence developed three years after the initial surgery. The physical examination revealed a lesion in left temporal area. Again she was undergone to resection of ameloblatoma. She kept going on clinical follow up when a second recurrence in left buccinators muscles was seen and another resection was done.

During the following years other five recurrences were observed, in infra-temporal, mandibular, cervical and parapharyngeal spaces and also treated with resection.

After six years of the former recurrence the patient showed the eighth recurrence on temporo-mandibular joint (she received pre-operative radiotherapy, followed by surgical resection). She had the ninth recurrence in the same year, in cervical soft tissue. Surgery and post operative radiotherapy with electrons were performed.

Last year an expansive lesion was identified. It was infiltrative in buccinator, parapharyngeal and submandibular sites, with 4.7cm on the largest diameter. At that time, a chest computed tomography (CT) showed multiple lung nodules (Figure 1). Biopsy of lung guided by CT showed basaloyd neoplastic cells (Figure 2).

Patient was undergone to pelveglossectomy and surgical reconstruction with myocutaneous flap. She was evaluated by thoracic surgeon and the plan was active watching of the lung nodules, since the patient has neither shortness of breath nor cough and the role of chemotherapy is doubtful.

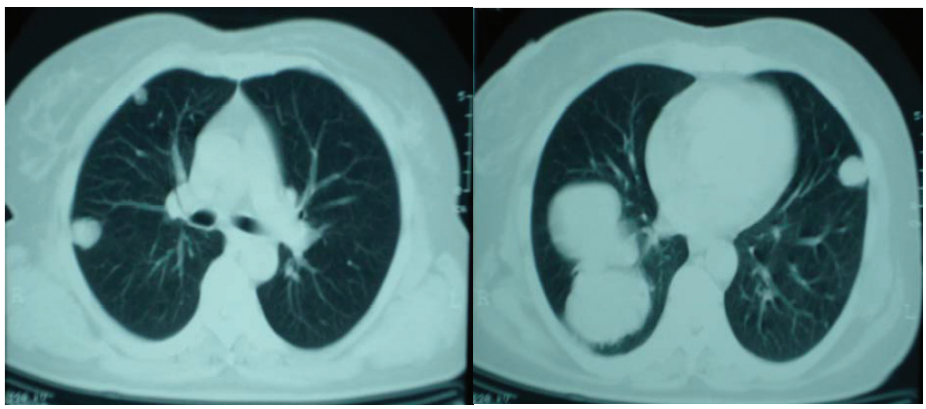

Figure 1 - A and B Chest computed tomography showing pulmonary metastasis

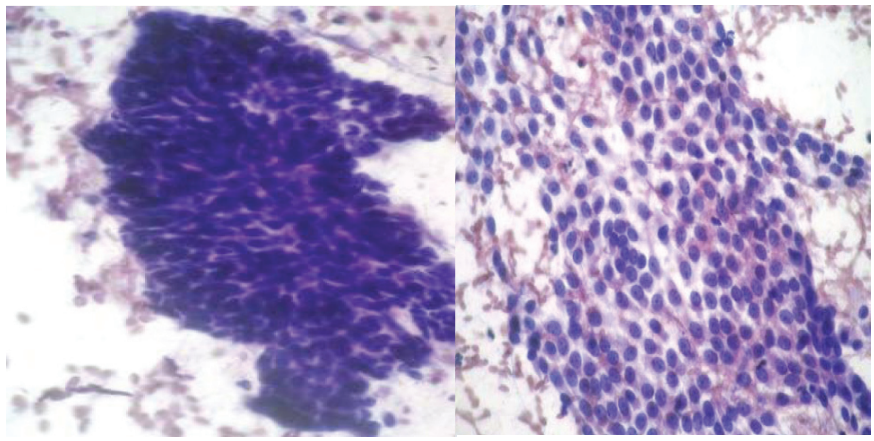

Figure 2 - A and B- Chest computed tomography showing pulmonary metastasis

During the evolution of ameloblastoma, the patient showed a *borderline* ovarian adenocarcinoma clinical stage III C, treated with surgery and adjuvant chemotherapy with six cycles of carboplatin and paclitaxel.

Discussion

Literature data show that although ameloblastoma is usually a benign odontogenic tumor, representing only 1% of all tumors and cysts of maxilla and mandible .[1-6] It is highly recurrent and rarely metastatic.[1]

The majority of patients with ameloblastoma are asymptomatic and the symptoms appear with the tumoral expansion. In the case reported here, the patient showed initially clinical history of previous chronic gengivitis, that, as described by Csiba, is a risk factor of ameloblastoma.[16]

In our patient, the neoplasm, had a locally advanced stage in the beginning and she was treated with wide surgical resection (hemimandibulectomy), as we can see in the literature data, where the backbone of treatment is the surgery and chemotherapy.

Locorregional recurrence occurs up to 90%[12,23] if the tumor is not completely resected. Even after surgical treatment, our patient showed locorregional recurrence nine times. However, although this disease is radioresistant, our patient undertook radiation twice, as neoadjuvant and postoperative setting. Other unusual fact is that our patient developed distant metastases (lung) after several local recurrences.

Now the patient has metastatic lung nodes and the medical oncology team chose close follow up until symptomatic stage or clearly progressive disease before starting the chemotherapy,that showed no benefit in previous series. There are some protocol with cisplatin and fluoraucil.

Grünwald et al.[33] related a case of lung and pleura metastatic ameloblastoma that was treated with palliative chemotherapy (carboplatin and paclitaxel) and the assessment did not show any evidence of disease.

During the course of ameloblastoma, patient developed a second primary neoplasm of ovary. She was treated with 6 cycles of carboplatin and paclitaxel, and even so, showed local relapse, making sure the non-effectiveness of chemotherapy in ameloblatoma.[32]

Conclusion

Ameloblastoma is a rare neoplasm notorious for having slowly growth and frequent relapses and the main therapeutic modality is surgery. Recurrences must have rescued by surgical resection as ever as possible. Radiotherapy is a good option when the surgery is not feasible, in order to become the tumor resectable or in the postoperative time if the margins are positive.

Chemotherapy has no role in the initial management of ameloblastoma. If the tumor area was irradiated previously and the surgery cannot be done, it can be considered, as well as in symptomatic distant metastasis.

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