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Pulmonary metastasis of malignant ameloblastoma: case report and review

Metástase pulmonar de ameloblastoma malígno: revisão e relato de caso

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ABSTRACT

Despite being a benign tumor of the maxillofacial region, some cases of ameloblastoma can be categorized as malignant ameloblastoma (or metastasizing) when metastases occur. The aim of this study is to report a rare case of lung metastasis from mandibular ameloblastoma, in order to review its risk and analyze the main anatomic sites that can occur with this disease. The case of a 48-year-old woman is described. She presented a metastatic pulmonary ameloblastoma 7 years after the removal of a mandibular ameloblastoma. During routine exams, a tumor in the left lung was observed. It was asymptomatic, near to the mediastinum, measured 7x5.5 cm. Transthoracic needle biopsy revealed ameloblastoma with the same histological characteristics of the primary tumor. After radiotherapy, the patient presented regression of the tumor. The patient has been under follow-up for 5 years and there is no presence of tumor. Ameloblastoma is an aggressive tumor not only in the region of origin, but also in distant regions, mainly in cases of recurrence. Metastases can cause high rates of morbidity, a fact that requires early treatment.

Indexing terms: Ameloblastoma. Metastasis. Radiotherapy.

RESUMO

Apesar de ser um tumor benigno da região maxilofacial, alguns casos de ameloblastoma podem ser categorizados como ameloblastoma maligno (ou metastizante) quando ocorrem metástases. O objetivo deste trabalho é relatar um caso raro de metástase pulmonar de ameloblastoma mandibular, a fim de revisar seu risco e analisar os principais sítios anatômicos que podem ocorrer com esta doença. Descreve-se o caso de uma mulher de 48 anos. Ela apresentou um ameloblastoma pulmonar metastático 7 anos após a remoção de um ameloblastoma mandibular. Durante os exames de rotina foi observado tumor no pulmão esquerdo. Assintomático, próximo ao mediastino, medindo 7x5,5 cm. A biópsia transtorácica com agulha revelou ameloblastoma com as mesmas características histológicas

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do tumor primário. Após radioterapia, o paciente apresentou regressão do tumor. O paciente está em acompanhamento há 5 anos e não há presença de tumor. O ameloblastoma é um tumor agressivo não só na região de origem, mas também em regiões distantes, principalmente nos casos de recorrência. As metástases podem causar altas taxas de morbidade, fato que requer tratamento precoce.

Termos de indexação: *Ameloblastoma; Metástase; Radioterapia.*

INTRODUCTION

The ameloblastoma is the most common odon¬togenic epithelial tumor, which belong to benign neoplasms that present a painless course, and usually occur in the oral and maxillofacial region. It is responsible for about 1% of all cysts and tumors of the jaw, and theoretically originates from the enamel organ epithelium, remnants of the dental blade or the epithelial lining of an odontogenic cyst [1]. The occurrence in the mandible is about 4 times greater than in the maxilla, 70% in the molar region [2]. The slowly growing tumor with a benign histologic appearance shows a local invasiveness and a high incidence (50% to 72%) of local recurrence after therapy [3].

On rare occasions the ameloblastoma can originate metastases even with a histologically benign appearance and is currently classified by WHO as metastatic or malignant ameloblastoma [2]. Most of metastases occur in the lungs and cervical lymph nodes but may also occur in the liver, spine, diaphragm, kidneys, brain, small intestine, spleen, scalp and myocardium [4]. The main metastatic dissemination theories are hematogenic, lymphatic and aspiration or implantation of cells at the time of surgery [5].

The aim of this study is to report a case of pulmonary metastasis of ameloblastoma, which demonstrated radio sensitivity during its radiotherapy treatment.

CASE REPORT

A 48-year-old female patient who underwent segmental resection of the right hemimandible and reconstruction with a rib graft aiming solid ameloblastoma treatment, returned to the maxillofacial surgery outpatient clinic 6 years after surgery. At the physical examination, an extensive tumor was observed in the mandibular symphysis region, compromising the entire buccal floor and superior projection of the tongue (figure 1A), which after incisional biopsy and histopathological analysis have confirmed the recurrence of the ameloblastoma (figure 1B).

The possibility of surgical treatment was discarded due to the extension and involvement of the lesion and the patient was referred to the head and neck surgery service for radiotherapy. In the beginning of the treatment, the tumor seemed to be quite radiosensitive and had an excellent therapeutic response and complete remission of the lesion which was shown by the physical and imaging examination.

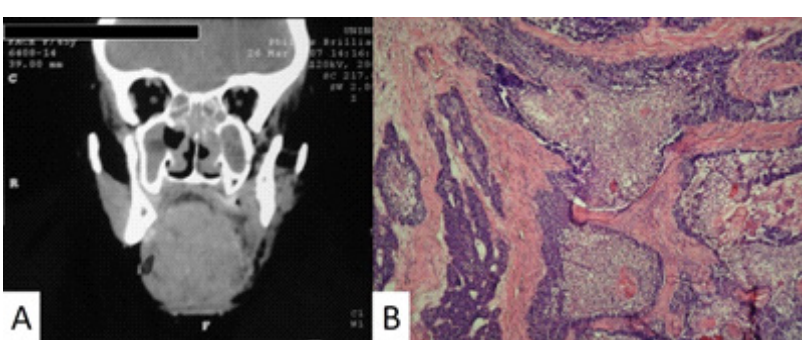

Figure 1 – A) Computed tomography demonstrating extension of mandibular lesion. B) Mandibular surgical biopsy sample showing characteristics of benign ameloblastoma (hematoxylin-eosin, x 300).

One year after radiotherapy treatment, during routine exams, a tumor in the left lung was observed, asymptomatic, near the mediastinum, measuring 7x5.5 cm (figures 2A and 2B). Transthoracic needle biopsy revealed ameloblastoma with the same histological characteristics of the primary tumor (figure 2C). Thus, she was referred to the thoracic surgery team, which decided not to perform a surgery, because they considered the impossibility of guaranteeing adequate surgical margins, in addition to the high risk of the procedure. Therefore, it was decided to follow the same strategy of radiotherapy, considering the radio sensitivity of the tumor, with a total dose of 50Gy in the mediastinum region (Opposite Pair) and 10Gy in tumor boost (Angulated) during 30 sessions, with regression to 1cm.

The patient has been under follow-up for 5 years and there is no presence of tumor (figure 3).

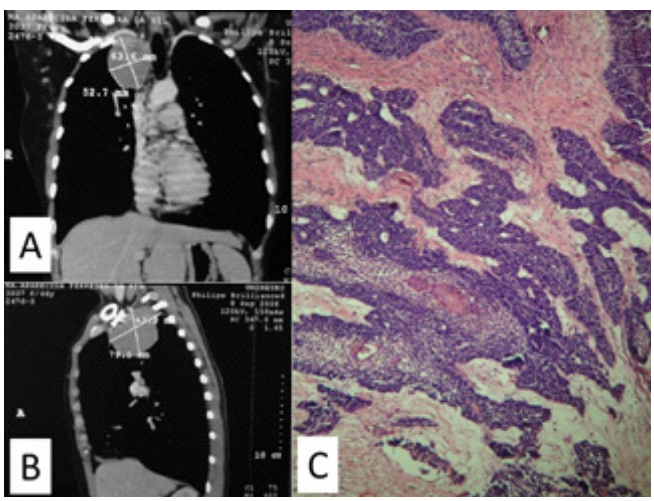

Figure 2 – A and B) Computed tomography shows metastatic nodules in left lung.

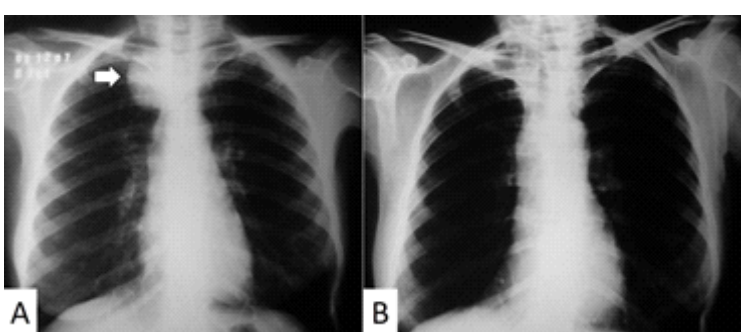

Figure 3 – A) Chest preoperative X-ray view showing lung nodules (white arrow). B) Chest postoperative X-ray demonstrating success in treatment.

DISCUSSION

Ameloblastoma is one of the most common benign odontogenic neoplasms of the jaws. They affect mainly the posterior region of the mandible and are locally aggressive, with a high rate of recurrence [6]. Some authors [7,8] tend to regard it as a potentially malignant tumor, but metastasis is rare. However, a histologically benign-appearing ameloblastoma can metastasize to local lymph nodes or other distant organs, such as the brain, lung and skin [7]. The World Health Organization (WHO) categorized malignant ameloblastoma into two types: metastasizing ameloblastoma

and ameloblastic carcinoma. Metastasizing ameloblastoma histologically resembles the benign ameloblastoma but demonstrates metastatic spread to distant regions. Ameloblastic carcinoma exhibits malignant histological features and can be further divided into two subtypes: primary and secondary. Primary ameloblastic carcinoma arises without previous injury, while secondary ameloblastic carcinomas are a result of malignant transformation of a previously diagnosed benign ameloblastoma [9].

The primary site of ameloblastoma occurs more often in the mandible than the maxilla, and is usually subclassified into solid or multicystic. Metastases are most common in the lung (occurring in 70% of cases), followed by lymph nodes (28%) and bones (12%) [2].

The diagnosis of metastatic ameloblastoma is usually made using a combination of patient history, physical examination, imaging findings and typical histopathological characteristics in a representative biopsy sample. In this case, symptomatology of metastasis was absent and was visualized on chest radiography, confirming what is described in the literature in which some cases are asymptomatic and observed only during routine exams.

Although they are usually visualized in conventional imaging tests (radiography and CT scans), some vertebral thoracic tumors can be difficult to find with these techniques.

To obtain a correct diagnosis of malignant or metastatic ameloblastoma, the primary lesion and the metastatic lesion should have the same histological characteristics as benign ameloblastoma. Thus, metastatic ameloblastoma with significant cellular atypia are considered ameloblastic carcinomas [2].

In cases of pulmonary metastasis, the biopsy can be performed by CT-guided percutaneous pulmonary puncture. This is an important method in the diagnosis of these diseases [8]. This technique was successfully used in the definitive diagnosis of the current reported clinical case.

The time that the metastasis takes place is variable and can happen at the same time of diagnosis of the primary tumor or several years after the treatment of the tumor. In the case described in this study, metastasis was observed 1 year after recurrence and 6 years after treatment of the primary lesion.

The treatment of most cases was perfomed by surgical excision of all pathological tissue. The recurrence of ameloblastoma is not likely to happen after bone ressection. However, Valkadinov et al. [4] presented a case of a 43-year-old woman with a large mass involving the neck and left mandible. The mass had formed over years and had been neglected. The woman was diagnosed with a primary ameloblastoma of the mandible. Surgical resection was performed, followed by adjuvant radiotherapy. Ten years later, she returned, and the findings were consistent with metastases of the previously identified ameloblastoma in the lungs. The patient was referred to further chemotherapy. The current case represents the classical course of a rare disease, which in this instance involved the common presentation of malignant ameloblastoma. In the present case, there was also a local recurrence of the lesion, which happened 6 years after resection of the mandibular lesion and the treatment was performed through radiotherapy with excellent local therapeutic results. This demonstrated good radiosensitivity of the lesion.

Some authors recommended this combination of surgery and radiotherapy in cases of extensive lesions and metastases. Yang et al. [1] treated bilateral metastatic lung ameloblastoma with palliative chemotherapy and 50 Gy radiotherapy with near complete remission of the lesion. The same dose was used in the current case; however, it was chosen by the addition of 10Gy in boost tumor.

In the present case, the same radiotherapy strategy was chosen after diagnosis of pulmonary metastasis due to observation of the impossibility of guaranteeing adequate surgical margins, and high surgical risks. This treatment plan was guided by the good response observed in the radiotherapy treatment used at the time of local tumor recurrence.

After reviewing the literature, Lin et al. [10] were able to observe that multiple surgeries and recurrences are risk factors for ameloblastoma metastasis, although ameloblastoma without recurrence may also develop tumor at distant regions. The authors suggest that the follow-up of treated patients of this tumor should be done regularly and for life with the objective of early diagnosis of possible metastases, especially in patients who had recurrence of the lesion.

Other authors [11] reported eleven patients of malignant ameloblastoma involving a single cervical lymph node metastasis and one new case treated by neck dissection and simple excision. Among the studied patients, eight had treatment with neck dissection. Two of them presented multiple organ metastases such as liver and lung seven months and 13 years after neck dissection respectively. Four patients underwent a simple node excision and there was no report of a regional neck recurrence and distant metastasis during follow-up of 1 to 7 years.

Death is not so uncommon in cases of malignant ameloblastoma. Seward et al. [12] reported a case of death in a patient with ameloblastoma metastasized to the lung 11 years after resection of the primary tumor on the face. According to the authors, death occurred as a result of the spinal and hepatic spread of the tumor, and thrombosis of the renal veins. Overall, 70% of patients have a 5-year survival rate, but this depends on the region of metastasis and surgical accessibility [2].

CONCLUSION

Based on what was hereby reported, it was concluded that ameloblastomas are aggressive tumors not only at the region of origin, but also in distant regions, mainly in cases of recurrence. Metastases can cause high rates of morbidity, a fact that requires a detailed planning and perfect execution by the surgical team

Collaborators

EFC Nogueira, manuscript development; text review. IS Catunda, case management; text correction. SCAS Carneiro, case management; text correction. PEFR Carvalho, manuscript development; text review. BCE Vasconcelos, case management; text correction.

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