Lung metastasis of benign giant cell tumor: a case report*

Metástase pulmonar de tumor de células gigantes benigno: relato de caso

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Abstract **Giant cell tumor is the sixth most frequent primary bone neoplasm, affecting long bone metaphysis, most frequently in young adults. On radiological images, this tumor appears as a lytic, well-defined, eccentric lesion. The authors report a case of benign giant cell tumor in a patient who presented with lung metastases five years after undergoing resection of the primary tumor.**
Keywords: Giant cell tumor; Benign; Metastasis.

Resumo **O tumor de células gigantes é a sexta neoplasia óssea primária mais comum. Acomete a metáfise de ossos longos, sendo mais comum em adultos jovens. Na radiologia mostra-se como lesão lítica, excêntrica e de limites definidos. Os autores relatam um caso de tumor de células gigantes benigno em paciente que apresentou metástases pulmonares cinco anos após a retirada do tumor primário.**
Unitermos: Tumor de células gigantes; Benigno; Metástase.

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INTRODUCTION

Giant cell tumors (GCT) are frequent bone neoplasms, corresponding to about 5–10% of all the primary bone tumors, and 15–25% of benign bone tumors[(1)]. Typical sites of these lesions include the distal femur, proximal tibia, distal radius, and, less frequently, bones of hands where studies demonstrate a higher tendency towards metastasis as compared with other sites[(2)]. The majority of patients affected are in the age range between 20 and 40 years, with a female 2:1 predominance. The highest incidence is found in the Chinese population. Typical radiological findings include lytic lesion with insufflation, trabeculation, with no periosteal reaction, and eccentric localization in the metaphysis of long bones. Histological findings include mononucleated stromal cells and multinucleated giant cells. The present study reports a case of benign GCT affecting the metacarpus of a young patient who five year after undergoing surgery developed lung metastases, among which one was giant.

CASE REPORT

A female, 28-year-old patient complaining of effort dyspnea and mild chest pain in the right hemithorax, with a tobacco smoking history and pneumonia being treated for one month in another institution where a chest radiograph demonstrated a mass in the right hemithorax.

The patient was referred to our institution where a chest computed tomography confirmed the presence of an extensive mass, with dense soft tissues, besides small

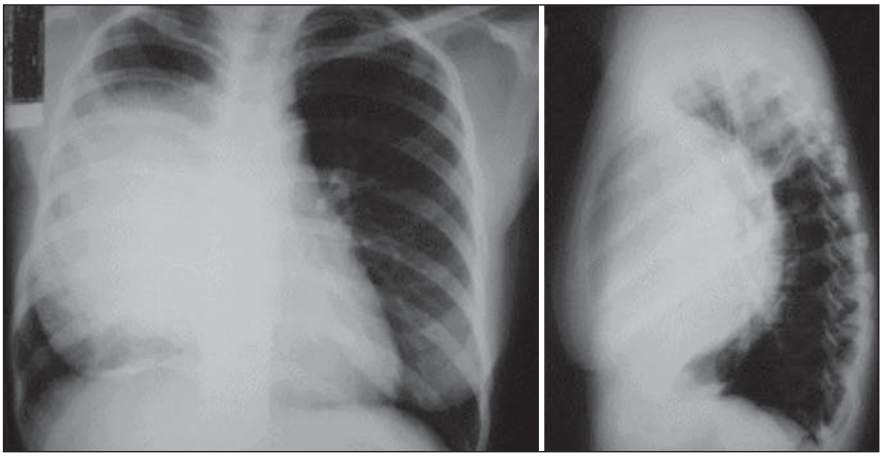

Figure 1. Chest radiography. Posteroanterior and lateral views showing an extensive mass in the right hemithorax, affecting the middle lobe and the anterior segment of the upper right lobe.

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nodules – two in the pulmonary apices and another on the right lung base.

Five years ago, the patient had been submitted to surgery for excision of a tumor localized in the second metacarpal bone of her left hand. Amputation of the metacarpal bone and correspondent chirodactilus was performed. The anatomopathological study demonstrated the presence of a benign GCT, but with extensive adherence of soft tissues.

Anatomopathological study of the pulmonary mass performed by means of transthoracic biopsy confirmed the presence of GCT metastasis showing features identical to those of the previously excised hand tumor. Because of the extensive pulmonary involvement, polychemotherapy was adopted as initial treatment of the lesions. Presently, the patient still remains under treatment. The pulmonary nodules disappeared and a moderate decrease in the volume of the mass can be observed.

DISCUSSION

Two malignant GCT variants are described: one, with a typical seemingly malignant stroma with intermingled giant cells, and another, with malignant transformation into fibrosarcoma, malignant fibrotic histiocytoma or osteosarcoma, particularly following radiotherapy in cases of recurrent GCTs. Additionally, like in the present case, distant metastases from histologically benign lesions occur in 0% to 9% of cases, with a mild prevalence in women[1–3].

Metastases occur preponderantly to the lungs, and rarely to the brain, kidneys, adrenal glands, gastrointestinal tract, other bones and skin. Authors suggest a higher rate of metastases in patients with at least a single episode of local recurrence and in radiologically aggressive tumors extending towards soft tissues. Campanacci et al.[4] have classified this type of tumor into grades I, II and III, according to radiographic findings, grade II and grade III tumors with higher probability of recurrence and metastasis than a grade I tumor. Grade I tumors present well-defined margins and a thin halo of mature bone. Grade II tumors also present well-defined margins, however, the sclerotic halo is absent. Grade III lesions present ill-defined margins suggesting aggressiveness. Meanwhile, the histo-

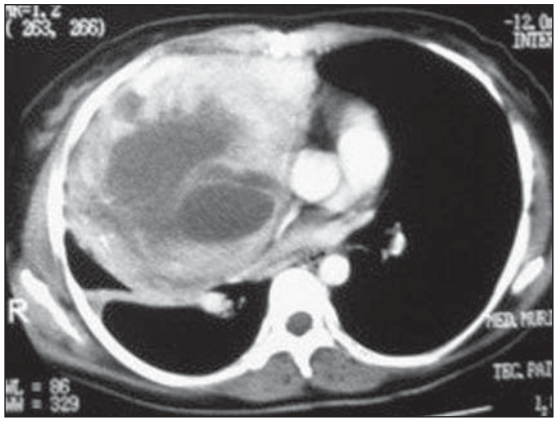

Figure 2. Computed tomography. Axial view, mediastinal window, demonstrating a mass compressing the superior vena cava, with dense soft tissues and heterogeneous contrast enhancement.

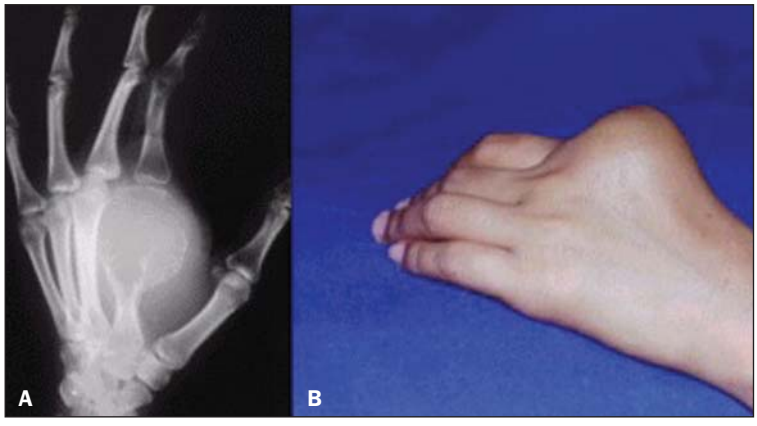

Figure 3. A: Hand radiography demonstrating lytic lesion with insufflation affecting the distal half of the second metacarpal bone. **B:** Photo of the hand showing the expansive nature of the lesion.

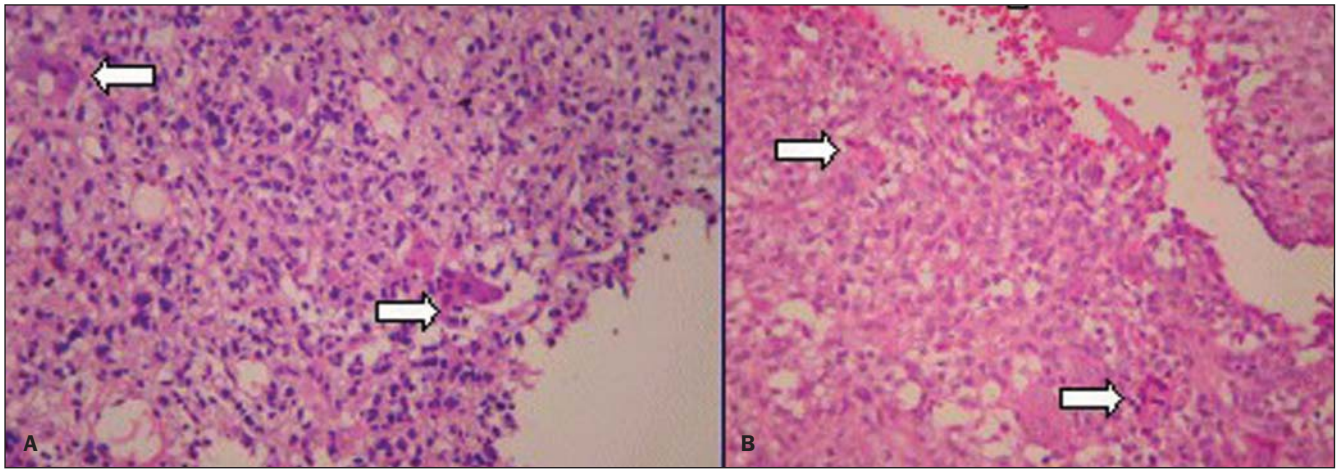

Figure 4. Hematoxilin-eosin transthoracic (**A**) and percutaneous biopsies of the finger (**B**) showing a benign stroma with intermingled giant multinuclear cells (arrows).

pathological classification of GCT of bone is not indicative of prognosis[5].

Immunohistochemical studies have been developed with α-SMA, MIC-2, p53, cyclin D1, tc-erb-B2 and Ki-67 antibodies in primary, recurrent and metastatic tumors. Authors have demonstrated a higher ratio of Ki-67-positive cells in more aggressive tumors[5]. In the present case, the immunohistochemical analysis was negative for p53 expression in the primary tumor, indicating a low risk for metastatic disease. The lung metastasis resulted positive for CD-68, and negative for S 100 protein, confirming the presence of GCT metastasis.

Regarding chronology, in about 75% of benign GCTs with lung metastasis, the time interval between the primary tumor diagnosis and metastasis detection is < 3 years. There are cases with simultaneous primary lesion/metastasis detection. In the present case, the time interval was longer than expected (five years). Metastases tend to present a slow growth, in some circumstances, they stop growing and, rarely a spontaneous regression occurs[3].

Tubbs et al.[6] have described the radiological findings of lung metastasis from benign GCT in 13 cases, such as round or oval nodular opacities, homogeneous density, sizes ranging between 0.5 cm and 8.0, well-defined margins, and a narrow transition zone between the lesions and the adjacent pulmonary parenchyma. The greatest majority of these lesions were peripherically located (85%) and in the pulmonary base (62%). In this study, CT was superior to chest radiography for detecting a higher number of nodules and presence of calcifications within these lesions. In the present study, the lesion presented with well-defined margins, but with huge dimensions (18.0 cm × 13.0 cm × 12.0 cm), heterogeneous density, localized in the middle and right upper lobes. The smaller nodules presented with the same features of those described by Tubbs et al.[6].

CONCLUSION

Although rare, CGT metastases must always be considered, requiring periodical follow-up after the primary tumor resection, considering that the symptoms show up late in the disease process, when these lesions may no longer be curable by resection.

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