Journal of the Formosan Medical Association (2015) 114, 369–372

Available online at www.sciencedirect.com

SciVerse ScienceDirect

journal homepage: www.jfma-online.com

CASE REPORT

A case of pulmonary metastasis of giant cell tumor of bone presenting as pulmonary arteriovenous malformation

Chang Dong Yeo [a], Sang Young Roh [b], Ok Ran Shin [c], Won Jong Bahk [d], Kwan Hyoung Kim [a], Jin Woo Kim [a,*]

[a] *Department of Internal Medicine, Uijeongbu St. Mary's Hospital, The Catholic University of Korea, College of Medicine, Uijeongbu, Republic of Korea*
[b] *Department of Internal Medicine, Seoul St. Mary's Hospital, The Catholic University of Korea, College of Medicine, Seoul, Republic of Korea*
[c] *Department of Pathology, Uijeongbu St. Mary's Hospital, The Catholic University of Korea, College of Medicine, Uijeongbu, Republic of Korea*
[d] *Department of Orthopaedic Surgery, Uijeongbu St. Mary's Hospital, The Catholic University of Korea, College of Medicine, Uijeongbu, Republic of Korea*

Received 24 November 2011; received in revised form 18 February 2012; accepted 14 March 2012

KEYWORDS
arteriovenous malformation;
giant-cell tumor

Although giant-cell tumor (GCT) of the bone was originally classified as a benign tumor, metastasis has been reported. The radiographic features usually comprise parenchymal solitary or multiple nodules that are round-to-oval nodular opacities of homogeneous density in patients with GCT. However, the patient described in this case presented with a hypervascular mass with feeding vessels and hemothorax, which are common features of pulmonary arteriovenous malformation. To the best of our knowledge, cases of pulmonary metastases presenting as a pulmonary arteriovenous malformation have not been reported. Here, we report a case of giant-cell tumor of the bone that exhibited histologically benign pulmonary metastases and mimicked an arteriovenous malformation.

Introduction

Giant-cell tumor (GCT) represents 5% of bone neoplasm and typically occurs in 20–40-year-old patients.[1] Although these tumors are usually benign and locally destructive,

* Corresponding author. Division of Pulmonology, Department of Internal Medicine, Uijeongbu St. Mary's Hospital, 65-1 Geumo-dong, Uijeongbu-si, Gyunggi-do, Republic of Korea.
E-mail address: medkjw@catholic.ac.kr (J.W. Kim).

0929-6646/$ - see front matter Copyright © 2012, Elsevier Taiwan LLC & Formosan Medical Association. All rights reserved.
doi:10.1016/j.jfma.2012.03.014

metastases have been reported in some patients.[2] Recurrence is most common in the lung, but there have been case reports of metastases at other sites, such as the lymph nodes, bone, skin, and breast.[2,3] Some patients with pulmonary metastases have spontaneous regression or remain asymptomatic for many years. Others have rapidly progressive courses, even though they exhibit histologically benign features. The overall mortality rate in patients with pulmonary metastases is approximately 15%.[1]

In the case presented here, pulmonary metastasis of a giant-cell tumor was proven to be the cause of hemothorax. Moreover, the radiograph showed a hypervascular mass with feeding vessels presenting as a pulmonary arteriovenous malformation, which is a rare manifestation of pulmonary metastasis of giant-cell tumor.

Case report

A 22-year-old woman was admitted to the emergency department presenting with severe dyspnea, chest pain, and right pleural effusion on a chest film. She had been diagnosed as having a GCT of the left distal femur 5 years previously, and had undergone intraregional curettage followed by methylmethacrylate bone cementing to fill the defect. During follow-up on an outpatient basis, any recurrence of the tumor was not occurred at the primary site or evidence of a tumor in the chest. However, 1 month before admission, the patient reported severe pain at the site of the previous excision. Radiography showed local recurrence of the tumor (Fig. 1). She was treated with curettage, cementing, and internal fixation using a Locking Compression Plate and screw.

On admission, blood pressure values were 100/60 mmHg; the pulse rate was 100/min and regular. Decreased vesicular breath sounds were heard over the right chest with dullness to percussion. Hemoglobin was 8.4 g/dl with normal red cell indexes, and the platelet count was normal. Three-dimensional (3-D) helical computed tomography (CT) revealed a well-defined oval and hypervascular mass in the right lower lobe with feeding vessels, highly suggestive of an arteriovenous malformation (Fig. 2). Paracentesis of the hemothorax showed true blood and a right thoracotomy was therefore performed. Thoracotomy revealed large blood clots and a notable mass having feeding vessels in the right lower lobe, up to 2 cm in dimension. She underwent wedge resection and the mass was removed successfully. The histological sections disclosed classic GCT metastasis with benign natures (Fig. 3). She recovered without sequelae and did not receive any adjuvant therapy. At present, the patient is well and there is no apparent recurrence at the primary tumor site or chest.

Discussion

GCT usually occurs in young adults and presents as an epiphyseal osteolytic bone lesion. Although this type of lesion was originally classified as a benign tumor, its potential for metastasis has been reported variously in 1—9% of patients.[2,3] A large study of 470 cases of GCT reported an incidence of metastasis of 5.1%.[3] The lung was the most common site of metastasis (21 of 24 patients) and

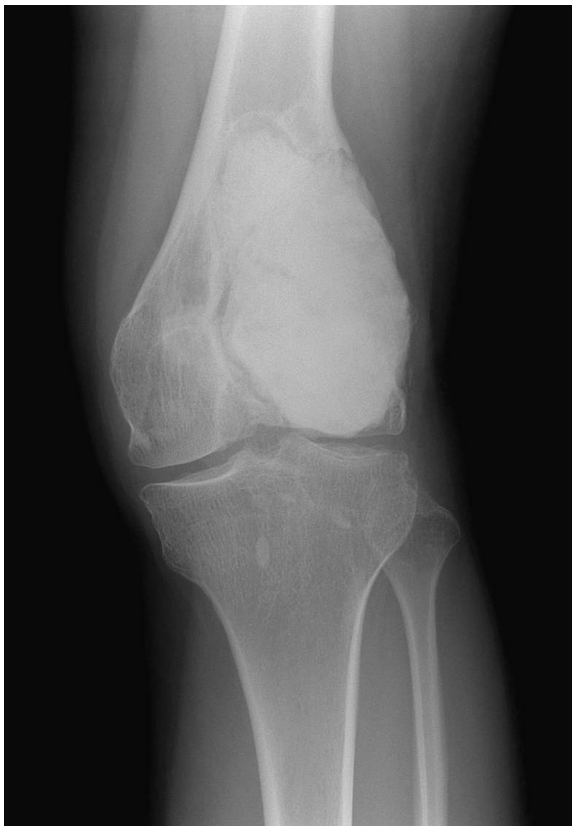

Figure 1 Radiograph of the distal femur shows a circumferential lucency around the bone cement.

extrapulmonary metastasis was rare. Patients' age and sex are not risk factors for metastatic disease and there is no predilection for the right or left side. The reported interval between diagnosis of the primary tumor and detection of metastasis ranges from 0 to 10 years (average, 3.5 years).[3] Although a case of endobronchial metastasis has been reported, metastatic lesions usually present as parenchymal solitary or multiple nodules, that are round-to-oval nodular opacities of homogeneous density.[4—6]

Although lung metastasis may spontaneously evolve into necrosis or ossification, the treatment of lung metastases is usually surgical resection.[6] If removal of the lesion is unfeasible, chemotherapy including adriamycin, dacarbazine, vincristine, cytoxan, actinomycin, or bleomycin can control evolution of the disease.[7] Radiotherapy is contraindicated because of the risk of induced malignancy. The mortality of pulmonary metastatic GCT is variable and uncertain, depending on the length of the follow-up. Cases observed for 8 or 9 years exhibit a mortality of 14—23%, suggesting the prognosis of a GCT with pulmonary metastasis is quite favorable.[8]

Pulmonary arteriovenous malformation is a rare pulmonary vascular anomaly. Although most patients are asymptomatic, this malformation can cause dyspnea, hemoptysis, hemothorax, and neurological complication because of paradoxical embolism.[9] The typical chest radiographic feature of pulmonary arteriovenous malformation is a round or oval mass of uniform density with prominent feeding vessels that is frequently lobulated and ranges from