Metachronous, multicentric giant cell tumors in Fibula and Rib

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Abstract

The authors report on a rare case of metachronous, multicentric giant cell tumor of bone in a young woman. The first lesion occurred in the right fibular head at 26 years of age, which was surgically treated. Twenty-seven months later, the second lesion developed in the right rib, and the patient underwent an excision of the tumor. Histologically, both tumors showed typical appearance of conventional giant cell tumor.
Giant cell tumor of bone accounts for approximately 5% of all primary bone tumors [3]. The tumor is locally aggressive and has the high local recurrence rate. In addition, giant cell tumor can metastasize to distant organs including lung. Most giant cell tumors are unifocal and multicentric giant cell tumor is a rare variant of the condition. Multicentric giant cell tumors occur synchronously or metachronously [1-9]. Fewer than 30 cases of metachronous, multicentric skeletal involvement by the tumor have only been reported in the English-language literature, to our knowledge [4,5,7]. The authors report on an additional case in which multicentric giant cell tumors metachronously developed in the fibula and the rib in a young woman.

Case Report

A 26-year-old woman presented with a 7-month history of a gradually increasing painful mass in the lateral aspect of her right knee. The patient had no history of trauma to the knee prior to presentation. Physical examination revealed a mass in the posterolateral aspect of the right knee. The mass, measuring 8x6 cm in size was well-defined, rough-surfaced, hard and tender. The range of motion of the knee was slightly restricted due to the mass. No joint effusion or instability was noted. The patient had no systemic illness.

Radiographs of the right knee showed a large osteolytic lesion in the fibular head, with cortical expansion and thinning (Fig 1). There were no foci of calcification or ossification within the lesion. Contrast-enhanced CT scans showed cortical thinning and partial breakthrough. The intramedullary lesion contained cystic and solid areas. The solid areas were significantly enhanced by contrast media (Fig 2). On magnetic resonance (MR) imaging, the lesion showed homogeneous low signal intensity on T1-weighted images. T2-weighted images showed that the lesion contained areas with mildly greater signal intensity than muscle and areas with markedly high signal intensity (Fig 3). $^{99m}$Technetium scintigraphy showed...
increased isotope uptake in the right fibular head. Laboratory data showed no abnormalities.

Fig. 1: AP radiograph of R knee with large osteolytic lesion in the fibular head

Fig. 2: Contrast-enhanced CT scan shows a lesion with cortical expansion and thinning.

Fig. 3: T2-weighted MR image shows solid and cystic areas in the fibular head

The patient subsequently underwent a total excision of the lesion. Histological specimens obtained at surgery consisted of solid tumor tissues and multiple, secondary aneurysmal bone cysts. The solid tumor tissues were composed of benign spindle-shaped stromal cells, numerous osteoclast-type multinucleated giant cells, and a few inflammatory cells. Pathologic diagnosis was grade I giant cell tumor of bone. The postoperative course was
uneventful. There was no local tumor recurrence in the right knee at a follow-up examination 24 months postoperatively.

At 27 months after surgery, however, the patient was incidentally found to have a right rib mass on a plain chest radiograph at another hospital, and referred to our institution again. Physical and radiographic examination revealed no tumor recurrence in the right knee. A mass, measuring 2x3cm, was palpated on the right 9th rib. The mass was hard, smooth-surfaced, ill-defined and nontender. Radiographs of the right ribs showed an osteolytic lesion in the 9th rib, with cortical expansion. CT scan showed an osteolytic lesion in the rib (Fig 4).

![Fig. 4: CT scan shows an osteolytic lesion in the right 9th rib.](image)

MR imaging demonstrated a lesion in the right 9th rib, with iso-signal intensity to muscle on T1-weighted and T2-weighted images. ⁹⁹ᵐ₆₇Technetium scintigraphy showed increased isotope uptake in the right 9th rib. The patient underwent an excisional biopsy of the rib lesion. Histological features of the specimens were similar to those of the right fibular head lesion, and giant cell tumor of bone was diagnosed (Fig 5). At a follow-up examination eight months after the second operation, there was no evidence of tumor recurrence in the right knee or in the rib, and the patient was asymptomatic.
Fig. 5: Histologic specimens obtained from the rib consist of numerous multinucleated giant cells and benign spindle-shaped stromal cells. Specimens obtained from the right fibula show similar findings. (hematoxylin and eosin, original magnification, x200).

Discussion

Multicentric giant cell tumor of bone is rare and represents less than 1% of all giant cell tumor cases [3]. Several investigators reported no difference between multicentric and unicentric giant cell tumor with regard to age or sex [1]. Multicentric giant cell tumor has a tendency to involve the small bones in the hands and feet [9]. Multicentric lesions are more likely to affect the metaphysis and the diaphysis of long bones in contrast to the conventional metaepiphyseal location [2]. The condition has occasionally been reported in skeletally immature patients and, in such cases, the condition has more aggressive course, with frequent pathologic fractures [4,6]. Histologically, there were no differences between unifocal and multicentric giant cell tumors [2,3,5,9].
Multicentric giant cell tumor of bone should be histologically and biochemically differentiated from other multiple lesions including brown tumor of hyperparathyroidism, multiple myeloma, metastases, eosinophilic granuloma, and Paget’s disease. In addition, direct extension or tumor seeding of a primary lesion should be ruled out. In our patient, the first and the second lesions were not contiguous. The metachronous occurrence of the lesions was supported by the scintigraphic findings; the scintigraphic study performed at the initial presentation detected only the knee lesion.

In rare cases of the condition, patients with more than 10 giant cell tumor lesions have been reported [2]. Park et al. [7] described a multicentric giant cell tumor case in which two lesions metachronously occurred at different sites in a single bone.

The possible pathogenesis of multicentric giant cell tumor includes distant metastasis and multiple independent foci of disease. Peiner et al. [8] suggested a metastatic spread along vascular pathways.

Previous studies reported that the interval original treatment and diagnosis of a second lesion ranged from 4 months to 16 years [9]. Therefore, long term follow-up is recommended in patients with giant cell tumor of bone. The present case suggested that postoperative, scheduled bone scan is beneficial in detecting asymptomatic lesions in patients who underwent an excision of a solitary giant cell tumor.

References


