

Giant cell tumor of ilium: An unusual location

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ABSTRACT

Giant-cell tumor (GCT) is a histologically benign, locally aggressive tumor that typically affects the ends of long bones, most commonly involving distal femur, proximal tibia, distal radius, and proximal humerus in the order of frequency. The involvement of flat bones of pelvis is extremely rare. We herein describe the unusual case of histologically benign GCT at ilium distinguished by marked destructive features on X-ray, CT, MRI, and histological examination necessitating hemi-pelvectomy in a 15-year-old female. Characteristics features of the giant cell tumor and its treatment options are reviewed.

Keywords: Giant cell tumor, ilium, osteoclastic tumor

INTRODUCTION

Giant-cell tumor (GCT) of bone is an uncommon neoplasm of uncertain origin that accounts for about 5% of all bone tumors.^[1] Statistically, 80% of GCTs have a benign course, with a local rate of recurrence of 20% to 50%. About 10% undergo malignant transformation at recurrence and 1% to 4% give pulmonary metastases even in cases of benign histology.^[2] It typically affects the ends of long bones, most commonly involving distal femur, proximal tibia, distal radius, and proximal humerus in the order of frequency. Involvement of the flat bones of pelvis is extremely rare. In particular, GCT of the ilium and ischium represent less than 0.05% of all GCT.^[3]

We herein describe the unusual case of histologically benign GCT at ilium distinguished by marked destructive features, necessitating hemipelvectomy. We also review the characteristic features of this unusual tumor.

CASE REPORT

A 15-year-old female presented with complaint of pain and swelling in the left buttock region and difficulty in

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sitting since last 8 months. She had a history of trauma at the same site 1 year back. There was no history of fever or loss of appetite. Palpation revealed tenderness over the left iliac region. Inguinal lymph nodes were palpable but not significantly enlarged. The systemic examination of the patient was unremarkable. An X-ray pelvis was done that revealed an expansile lytic lesion of soft tissue intensity measuring 6.5 cm × 6 cm × 5.5 cm involving the left iliac blades. Computed tomography (CT) scan and magnetic resonance imaging (MRI) confirmed the findings of x-ray as T2 weighted MRI imaging revealed hyperintense areas in the same lesion [Figures 1 and 2]. Her routine laboratory investigations were within the normal limits. The serum calcium, serum phosphorus, and alkaline phosphatase were normal. An x-ray of the chest was unremarkable. Needle biopsy was performed that suggested the diagnosis of the giant cell tumor.

Based on these findings, internal hemipelvectomy was performed. Tumor mass was isolated from surrounding structures. Postoperative course of the patient was uneventful.

On gross examination, hemipelvectomy specimen measuring 16 cm \times 12 cm \times 9 cm, external surface was congested, dark red colored. Cut surface showed tumor measuring 7 cm \times 5.5 cm \times 4.5 cm. The tumor mass was soft friable, with solid, cystic and hemorrhagic areas. Microscopically, the tumor was composed of large number of uniformly distributed osteoclastic giant cells separated by mononuclear stromal cells with round to oval nuclei. At places blood filled spaces lined by fibrous tissues and

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giant cells were identified. Vascular invasion was present and there was no evidence of any sarcomatous change in mononuclear stromal component. Resected surgical margins were free from tumor infiltration. Resected lymph nodes revealed changes of reactive lymphadenitis. The final diagnosis based on clinical, radiological, and pathological findings made was giant cell tumor of the left iliac bone [Figure 3].

DISCUSSION

Giant cell tumor is described as neoplasm of undifferentiated mesenchymal stromal cells with the presence of abundant, multinucleated giant cells. They are considered benign but may present with aggressive, potentially malignant clinical course. They can recur in a high percentage of cases, become sarcomatous yet produce metastasis even without apparent malignant morphology. In 1912, Bloodgood named the tumor as a benign, "giant cell tumor." The tumor typically involves the epiphyseometaphyseal region of long bones and is usually expansile and eccentric in position.^[4] The ilium is very unusual site for giant cell tumor.^[3] Balke et al., in their series of 20 cases of giant cell tumor of the pelvic bone over a period of 20 years found nine cases involving the ilium.^[5] In the series of seven cases of GCT of innominate bones done by Kattapuram et al., only one case involving the ilio-sacral region was identified. Both of these studies highlighted the rarity of the lesion. The locally aggressive nature of the lesion warrants proper assessment and planning for management. GCT of the pelvic bone mostly occurs in third or fourth decade of life with a clear female predilection.^[6] There are very few series that document GCT of bone in the immature skeleton and the reported incidence in the literature varies from 1.8% to 10.6%.[7] But in our case patient presented at her teenage. Most patients present with symptoms of pain. The average size of the tumor in this location is large that is quite comparable with the size of tumor in this case.^[8]

The diagnosis of a giant cell tumor of the pelvis may be delayed and challenging. In this case, the duration of symptoms was 8 months. The delay is related to several factors. GCT exhibits purely lytic destruction of bone. The area of lysis may not be recognized on plain radiographs during the early stage of the disease or lytic areas may be confused with the appearance of gas in the bowel. Any shielding of the ovaries during diagnostic radiographic procedures may hide the lesion. Deep location of the tumors may also be the factor. The clinical presentation of these lesions may be confused with that of low back pain, muscle strain or strain of sacro-iliac joint.^[9] A high index of suspicion is required to diagnose a young female patient



Figure 1: X-ray pelvis shows lytic lesion in the iliac blades with irregular margins



Figure 2: T2 weighted MRI imaging shows hyperintense areas in the same lesion



Figure 3: Microphotograph shows large number of uniformly distributed osteoclastic giant cells separated by mononuclear stromal cells with round to oval nuclei (H and E, 100×)

presenting with pain and involvement of sacro-coccygeal area keeping in mind the possibility of GCT.

The arteriogram, computed tomography, and magnetic resonance imaging (MRI) are extremely helpful in delineating the lesion and planning the mode of treatment. MRI is the best imaging modality to study GCT and its soft tissue extension because of superior contrast resolution. MRI is better in determining extra-osseous extent of tumor.^[10]

Giant cells in histopathology are also commonly seen in other lesions, the so called variants of giant cell tumors. These lesions include nonossifying fibroma, unicameral bone cyst, localized osteitis fibrosa, aneurysmal bone cyst, chondroblastoma, and the "brown tumor of hyperparathyroidism. One of the main microscopic differences between true GCT and these so-called variants resides in the spatial relationship between giant cells and stromal cells. The giant cells tend to be regular and uniformly distributed in GCT, whereas in lesions that simulate it, foci containing numerous, clumped giant cells alternate with large areas completely lacking this component. Microscopic grading of GCT is not of great value except for the obviously sarcomatous (Grade III) lesion.^[4]

Curettage with or without resection is treatment of choice of GCT involving the pelvic bones. In advanced cases of aggressive lesions with marked cortical destruction and extensive soft tissue involvement, wide resection even up to the extent of amputation may be necessary for complete cure. The radical surgery may be able to definitely prevent local recurrence and distant metastasis.^[6]

It seems astonishing to the pathologists that a benign GCT is capable of recurrence and metastasis. Many authors have done painstaking studies to assess the aggressiveness, recurrence, and metastasis of GCT of bones that are histologically benign. Their studies had revealed that importance should be given to all gross, histological, radiological and clinical features rather than only histopathological appearance to assess the metastatic potential of the tumor.^[11]

CONCLUSION

Giant cell tumor accounts for 4–5% of all bone tumors. It usually occurs after the closure of growth plates. Ilium and ischium constitute less than 0.05% of all the sites and slightly more common in females. Although histologically benign, it may show local recurrence and metastasis. Diagnosis in a case involving pelvic bones is difficult to make on radiology alone and requires the histopathologic confirmation.

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