

# Metastatic osteogenic sarcoma or multifocal osteosarcoma: A case report

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## Abstract

Multifocal osteosarcoma is a rare condition in which multiple osteogenic sarcoma at various regions are present at diagnosis. These account for 1-3 percent of all osteogenic sarcoma. A thirteen-year-old female presented with pain and swelling over her right thigh for three months with no history of trauma. The diagnosis of osteosarcoma was suspected through radiology which incidentally also showed the other femur to have involvement on magnetic resonance imaging scan. A suspicion of either metastatic osteogenic sarcoma or multifocal osteosarcoma was made, following which computed tomography of the chest was done to look for pulmonary metastasis and diagnosis of metastatic osteogenic sarcoma was confirmed by biopsy from the right thigh.

**Key words:** Biopsy; Osteosarcoma; Tumour.

## INTRODUCTION

Multifocal osteosarcoma is the simultaneous occurrence of multiple skeletal osteosarcomas. It is a rare condition which accounts for about 1-3% of all osteogenic sarcoma<sup>1</sup> and has a poor prognosis.<sup>2</sup> The lung is the most common site for metastasis for osteogenic sarcoma.<sup>3</sup> We report a case of biopsy proven metastatic osteosarcoma with pulmonary metastases. This case is

of interest because there was high suspicion that it was a case of multifocal osteosarcoma which after finding of pulmonary metastasis turned out to be metastatic osteogenic sarcoma.

## CASE REPORT

A thirteen-year-old female presented with complaints of pain and swelling over her right thigh for three months with no history of trauma. She had generalised weakness, loss of weight and appetite for three months. There was no family history of any hereditary disease or malignancy. On examination, she was anaemic and had diffuse ill-defined swelling over the right thigh. Her initial blood examinations showed low haemoglobin, raised alkaline phosphatase and Lactate Dehydrogenase (LDH).

X-ray of the right hip with femur showed sclerotic lesion extending from the femur neck up to the middle of diaphysis of femur with appearance of Codman's triangle (Figures 1 and 2). Moreover, findings from the chest X-ray were unremarkable. For further evaluation Computed Tomography (CT) chest was done which showed pulmonary metastasis.

Extensive signal abnormality was noted in metaphyseal-diaphyseal region of right femur including head and neck region extending across the right hip joint involving acetabulum, ischium, quadrilateral plate and iliac bone

### Access this article online

**Website:** [www.jkmc.com.np](http://www.jkmc.com.np)

**DOI:** <https://doi.org/10.3126/jkmc.v10i3.41244>

### HOW TO CITE

Dahal S, Rajak A, Rajak A. Metastatic osteogenic sarcoma or multifocal osteosarcoma: A case report. J Kathmandu Med Coll. 2021;10(3):161-5.

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ISSN: 2019-1785 (Print), 2091-1793 (Online)



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with mild right hip joint effusion. The lesion also extended to involve the surrounding muscle in upper and mid-thigh viz. vastus intermedius, medialis, and quadriceps with haemorrhagic layering of contents within it. The neurovascular bundles were partially encased in medial thigh, however, the posterior neurovascular bundle was spared. The process was also noted across both knee joints and left hip to involve left femur, tibia and fibula bilaterally (Figures 3, 4 and 5).

Multiple small nodules are seen in right lower lobe of lung that show predominant subpleural distribution. These nodules are suspicious for metastasis (Figure 6).

Bone scan revealed multiple foci of moderate to intense uptake in cervical, thoracic, lumbar vertebrae, ribs, humerus, both sacroiliac joints and both femoral bones (Figure 7). The uptake in right femur is likely to be the site of primary tumour and uptake in rest of skeleton could be due to osseous involvement.

Tissue biopsy from the right hip region revealed immature, irregular woven bone and osteoid; there is infiltration of pleomorphic tumour cells with moderate amount of cytoplasm and hyperchromatic nuclei suggestive of osteogenic sarcoma (Figure 8).



**Figure 1:** X-ray of the right hip with femur



**Figure 3:** MRI of bilateral hip showing sagittal view



**Figure 2:** X-ray of bilateral hip with pelvis



Figure 4: Axial section of MRI of bilateral hip and thigh

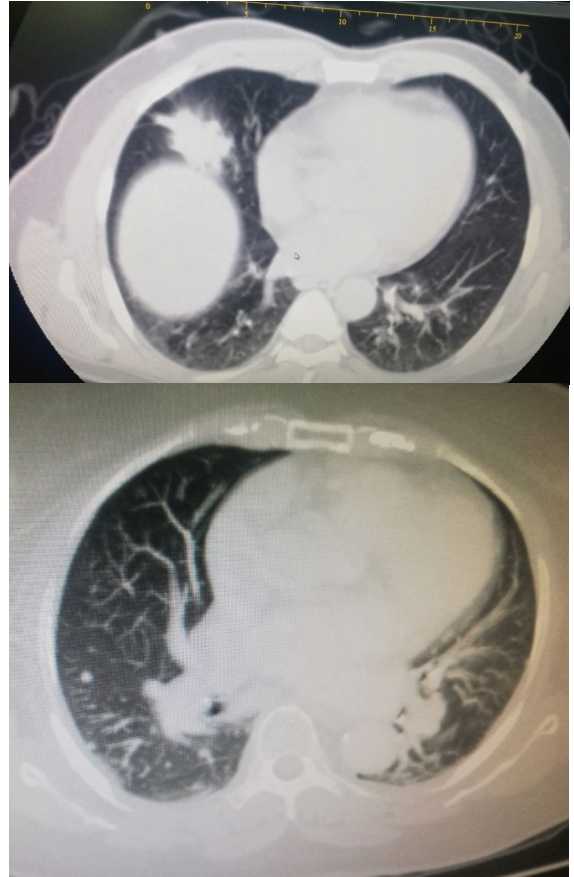


Figure 6: CT scan of the chest



Figure 5: Coronal section of MRI of bilateral leg

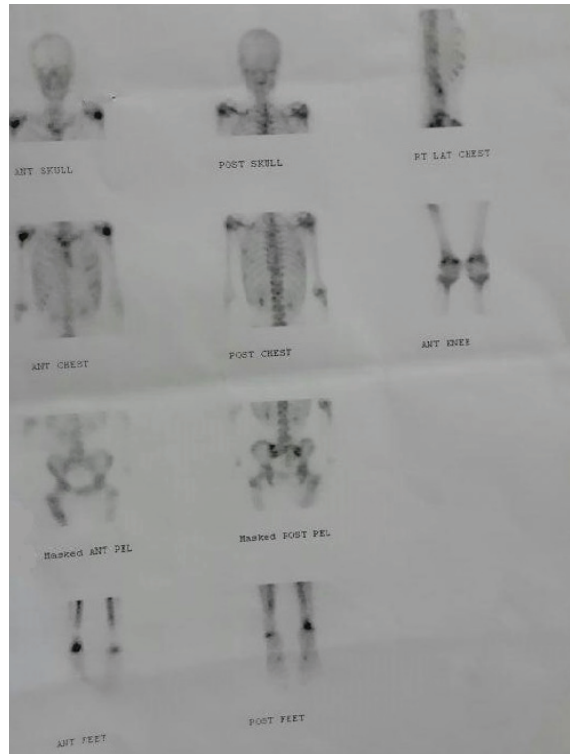
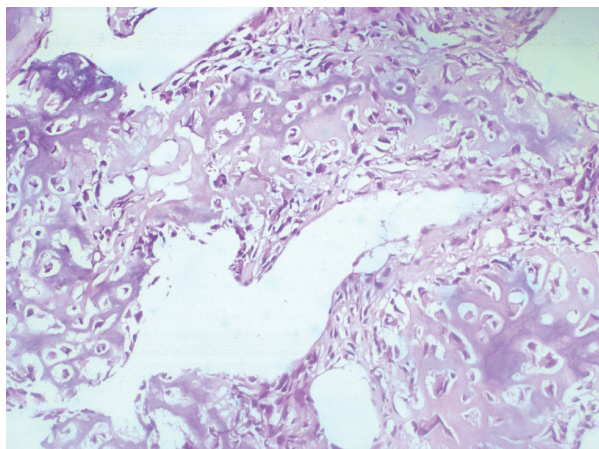


Figure 7: Bone scan



**Figure 8: Histopathological section of the biopsy lesion taken from the thigh**

## DISCUSSION

Multifocal osteosarcoma is considered a rare distinct form of disease that is characterised by presence of multiple sclerotic foci bilaterally, mostly at the metaphysis of the long bones with or without a dominant lesion.<sup>2</sup> There is still a controversy whether the lesions are multicentric in origin or unicentric in origin with multiple intraosseous metastases. Multicentric origin is favoured by the similarity in size, radiological features, histology, absence of dominant lesion and lack of pulmonary metastasis. Presence of dominant lesion and pulmonary metastasis favours unicentric origin with metastasis. Increased level of serum alkaline phosphatase levels and lactate dehydrogenase levels are useful in following the clinical evolution of multifocal osteosarcoma and are considered to have worst prognosis.<sup>4,5</sup>

However, presence or absence of pulmonary lesions cannot be used as a single differentiating factor between multifocal osteosarcomas and unicentric metastatic osteosarcomas. This is because rapid appearance of secondary skeletal lesions without pulmonary metastases occur via the Batson's vertebral venous plexus or lymphatics or intraosseous embolisation through marrow sinusoids.<sup>6</sup>

Attempts have been made to classify multifocal osteosarcoma but due to variation in aetiology, disagreement regarding the accuracy of any classification system exists. The most commonly used classification system is of Amstutz<sup>7</sup> who based it on the age of the

patient; distribution and symmetry of the lesions; and presence or absence of pulmonary metastases. However, this classification is considered to be capricious and is no longer used.<sup>8</sup> Presently, Resnick classification is widely accepted which has described the lesions under three headings.<sup>9</sup>

Even though the entity of multifocal osteosarcoma is debatable, it is agreed that multifocal osteosarcoma is either synchronous or metachronous. Synchronous represents a rare distinctive form of osteosarcomatosis, common in young children, with symmetrical metaphyseal involvement with osteosclerotic lesions. Irrespective of age or presence or absence of metastasis, a dominant lesion with subsequent metastasis can be grouped as metachronous. The case presented above may have been a metastatic osteosarcoma or a case of metachronous type.

Due to the combination of surgery and adjuvant or neoadjuvant chemotherapy, the five-year overall rate of survival for patients with non-metastatic osteosarcoma of an extremity has improved dramatically from less than 15% in the 1970's<sup>10,11</sup> to more than 65%<sup>12,13</sup> over the last twenty years. Patients with osteosarcoma of the extremity who present with lung metastasis have a poorer prognosis than those without metastases. With the combination of chemotherapy and complete surgical removal of primary and secondary lesions, the survival of patients with metastases has improved from less than 5% to more than 20%.<sup>14,15</sup> However, when complete excision of all of the secondary lesions is impossible, the five-year survival rate becomes extremely low and almost all of these patients die within three years from the time of diagnosis<sup>10</sup>.

The limitation of this study was that the patient was lost shortly after biopsy. Further treatment could have given more information about such type of disease.

## CONCLUSION

It is still controversial whether the lesions of multiple osteosarcoma are multicentric in origin or unicentric in origin with multiple intraosseous metastases. Early detection and knowledge of this variable should be kept in mind while managing such cases.

**Conflict of interest:** None

**Source(s) of support:** None

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