



## Case report

## Osteosarcoma presenting with malignant pleural effusion in a 55 year old

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## A B S T R A C T

Osteosarcoma is the most common primary malignant neoplasm of the bone with over 60% of the cases occurring in patients 10–20 years old. Osteosarcoma rarely occurs in patients older than 40 years of age, most commonly in bones affected by preexisting conditions such as Paget's disease, prior irradiated bone or osteogenesis imperfecta. Osteosarcoma presenting with pleural metastases is very rare. Herein we describe a case of metastatic osteosarcoma presenting with pleural effusion due to underlying pleural metastases in a 55 year old woman.

## 1. Introduction

Osteosarcoma is the most common primary malignant neoplasm of bone and has the second highest mortality rate among pediatric cancers [1]. It is most commonly diagnosed in patients in the second and third decades of life, with over 60% of the cases occurring in patients between the ages of 10–20 years [1]. Osteosarcomas respond dramatically to treatment however metastatic disease eventually develops in almost 25–30% of the patients with the lung being the most common site of metastases [2].

Osteosarcoma rarely occurs in patients older than 40 years of age, usually in bones affected by preexisting conditions such as Paget's disease, prior irradiated bone or osteogenesis imperfecta [3]. The prognosis in older patients is worse as compared to younger patients. Studies have shown that prognosis worsens with age among adults while being age-independent in children [4]. This has been attributed to the more central location of tumors and decreased ability to tolerate high dose chemotherapy among older adults [5].

Osteosarcoma presenting with pleural metastases is extremely rare. Herein we describe a case of metastatic osteosarcoma presenting with pleural effusion due to pleural metastases in a 55 year old woman.

## 2. Case report

A 55 year old woman presented with persistent shortness of breath and right lower limb weakness.

CT of the chest revealed a right pleural effusion with thickened, focally calcified irregular pleura on the right side and a calcified nodule

in the left lung (Fig. 1). CT of the abdomen revealed a large expansile destructive lytic lesion involving the right iliac bone with a significant soft tissue component (Fig. 2). Areas of matrix calcifications were seen within with the Codman triangle, measuring 13 × 11 × 8 cm with aggressive lamellated and sunburst type periosteal reaction.

A right lateral thoracotomy was performed and the pleural effusion was drained. The pleural mass was biopsied. Microscopic examination revealed a diffuse proliferation of spindle to oval shaped cells showing moderately pleomorphic, hyperchromatic nuclei and frequent mitoses with associated deposition of abundant osteoid. The cells exhibited extreme pleomorphism and nuclear atypia (Fig. 3). The pleural biopsy was highly suggestive of high grade osteosarcoma and in the presence of radiological evidence of osteosarcoma in the right ileum, the diagnosis of metastatic osteosarcoma was made.

## 3. Discussion

Osteosarcoma is an aggressive primary bone tumor arising from primitive bone-forming cells. A number of variants of osteosarcoma have been documented. These include telangiectatic, multifocal, parosteal, and periosteal types in addition to the conventional osteoblastic, chondroblastic, and fibroblastic types [6]. The metastases of osteosarcoma are typically hematogenous and microscopic metastases are usually present at the time of diagnosis. At presentation, 10% of the patients have distant metastases, the most common sites being lung and bone [7]. Isolated pleural metastases in the absence of ipsilateral lung deposits are exceedingly rare [8]. According to the Japan Autopsy Annual Database, 643 patients died of osteosarcoma between 1981 and

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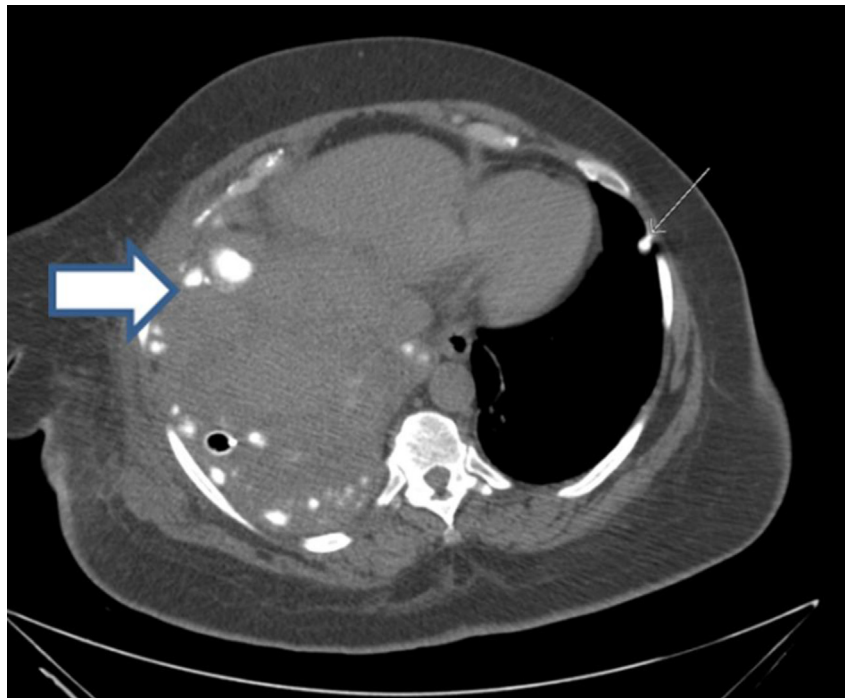


Fig. 1. CT scan of thorax, axial section soft tissue window, showing extensive calcified pleural metastases on the right side (arrow) and a calcified nodule in the left suggesting metastasis (long arrow).



Fig. 2. CT scan axial section in bone window showing lesion involving the right iliac crest with soft tissue component (arrow).

2002 in Japan, of whom, only 78 (12.1%) patients had pleural metastases [9].

The most commonly reported CT impression is of single or multiple pleural based nodules with concomitant lung parenchymal metastases.

Diffuse pleural thickening with a mass lesion, as seen in our case is a rarely reported incidence. Also there are no conclusive published data regarding the occurrence of pleural effusion in patients with osteosarcoma.

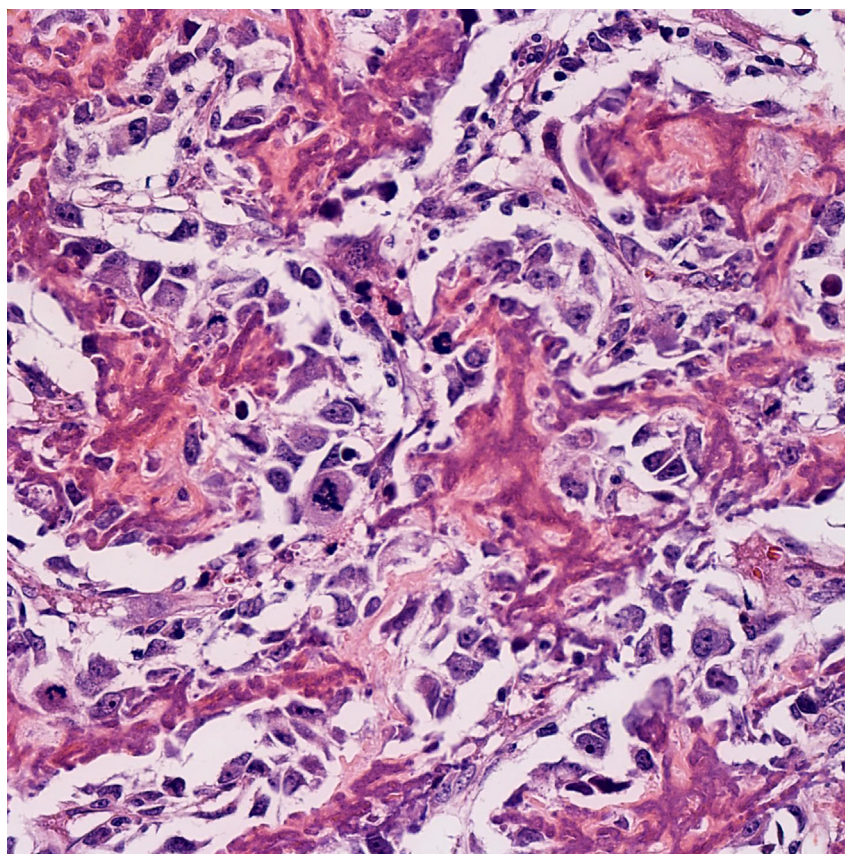


Fig. 3. 400X view showing pleomorphic cells with mitoses and osteoid formation.

Probable routes of pleural metastases in patients with osteosarcoma may be through direct contact of pleura with the lung metastases or hematogenous spread [10]. It is not possible to track the definite route of pleural metastases. The presence of a pleural based mass, without any radiologically evident ipsilateral lung nodules favors hematogenous spread of the tumor in our patient, however microscopic metastases in the ipsilateral lung cannot be ruled out. Also the lack of pleural metastases on the opposite side despite the presence of metastatic lung nodules speaks against a local spread.

Overall management and prognosis in cases of metastatic osteosarcoma is determined by the number, site and size of the metastases [1]. Complete surgical resection is a prerequisite for cure and improves the survival of the patient. Osteosarcoma presenting with metastatic parietal pleural lesions, either in isolation or in association with a lung lesion, should be completely resected with free pleural margins [11]. Patients with unresectable disease may benefit from radiotherapy [12].

#### 4. Conclusion

Osteosarcoma presenting with pleural effusion due to pleural metastases is extremely rare. Surgical resection of the localized pleural tumor improves patient outcome in osteosarcoma with pleural metastases. In patients with undiagnosed pleural effusion and radiological suspicion of osteosarcoma, thoracoscopy is recommended to identify and biopsy pleural metastases for establishing the definitive diagnosis.

#### Declarations

The work described has been carried out in accordance with the code of ethics of the world medical association (Declaration of Helsinki). The authors have no conflicts of interest to disclose.

#### Declarations of interest

None.

#### Conflicts of interest-

We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome.

#### Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.rmcr.2018.10.017>.

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