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CASE REPORT PEER REVIEWED | OPEN ACCESS

# Unusual peritoneal metastases of femoral osteosarcoma

El Houssni Jihane, El Haddad Siham, Dghoughi Basma, Bassel Saber Abdellah, Chat Latifa, Allali Nazik

## ABSTRACT

**Introduction:** Osteosarcoma is the most common malignant bone tumor in children, most often occurring in the lower extremity of the femur, and is known to be particularly aggressive, with a high risk of metastasis. The most frequent secondary localizations are pulmonary and bone metastases; peritoneal metastases are extremely rare and have a poor prognosis.

**Case Report:** We report the case of a 14-year-old child with a history of a treated right femoral osteosarcoma, which presented with peritoneal and pulmonary metastases.

**Conclusion:** Imaging plays a very important role in the management of patients with osteosarcoma, it allows an early detection of metastases through a rigorous clinico-radiological surveillance, metastases usually have the same radiological appearance as the primary tumor and the diagnosis of certainty is essentially based on anatomopathological study.

**Keywords:** Histology, Imaging, Osteosarcoma, Peritoneal metastasis, Pulmonary metastases

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## INTRODUCTION

Osteosarcoma is the most common malignant bone tumor in children, most often occurring in the lower extremity of the femur, and is known to be particularly aggressive, with a high risk of metastasis. The most frequent secondary localizations are pulmonary and bone metastases; peritoneal metastases are extremely rare and have a poor prognosis.

## CASE REPORT

We report a case of a 14-year-old male child with a history of right femoral osteosarcoma treated with neoadjuvant chemotherapy prior to amputation of the right lower limb.

This patient presented with acute abdominal pain in the right hypochondrium, an abdominal ultrasound was performed showing hypoechoic, heterogeneous peritoneal tissue lesions that did not show up any blood flow on color Doppler (Figure 1), associated with a small perihepatic and pelvic peritoneal effusion.

He also underwent a thoracic radiography which revealed a dense and homogeneous opacity, occupying the lower two-thirds of the right thoracic hemi-field, with a clear upper limit and convex toward the parenchyma, pushing the mediastinum toward the contralateral side (Figure 2).

For a better characterization of these lesions, a thoraco-abdomino-pelvic computed tomography (CT) scan was performed, objectifying:

In the abdominal and pelvic level: peritoneal nodules, hypodense, oval containing calcifications, scalloping on the liver, the largest of which measured 89×35 mm and 53×28 mm (Figure 3), associated with a small peritoneal effusion.

And on the thoracic level: a large right lung mass centered on the lower and middle lobes, well limited, with regular contours, hypodense, heterogeneous, containing calcifications, measuring: 170×160×190 mm (AP×T×H), medially: it amputates the middle and lower lobar bronchi, it exerts a mass effect on the esophagus with reduction of its lumen without clear interface, and on the atria with persistence of a fatty separation line, it also laminates the right inferior pulmonary vein and the right middle and inferior lobar arteries which, however, remain permeable, below: it invades the diaphragm and displaces the liver with loss of the separating line in places (Figure 4), posteriorly: it comes into contact with the vertebral bodies from D6 to D12 without bone lysis opposite, as well as with the right costal arches from K6 to K12. There is a multi-lamellar periosteal reaction of the posterior arch of K9 and focal lysis of the middle arch of K10 (Figure 5).

Anatomopathological study of a peritoneal nodule was in favor of pleomorphic anaplastic and osteoblastic spindle-shaped neoplastic cells producing an immature osteoid matrix (Figure 6), the morphological appearance and the immunohistochemical profile are compatible with secondary localization of a high grade osteosarcoma.

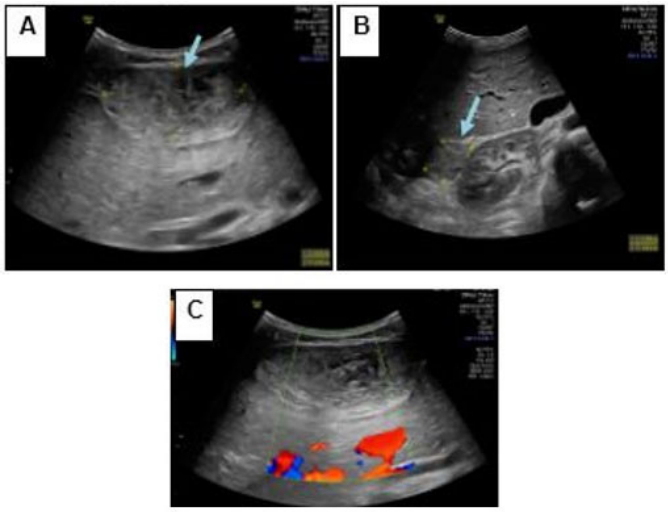


Figure 1: Abdominal ultrasound: peritoneal tissue lesions (A, B), hypoechoic, heterogeneous, not taking color Doppler (C).

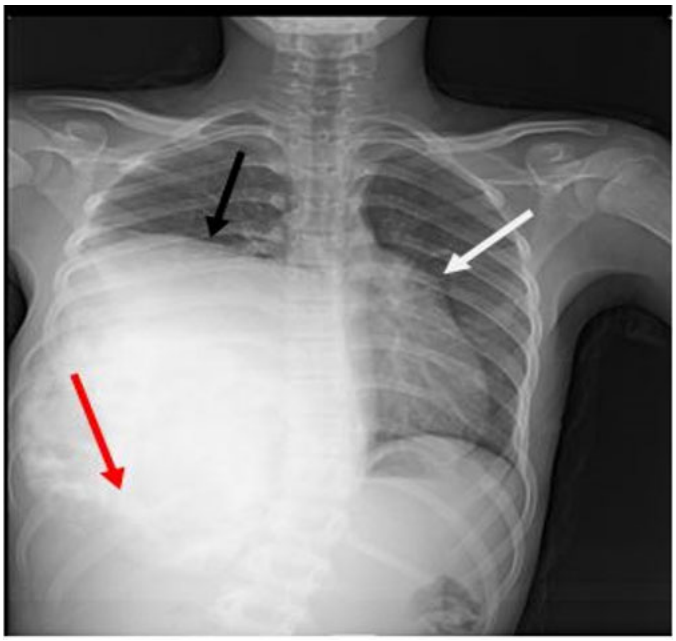

Figure 2: Frontal chest X-ray: Dense and homogeneous opacity, occupying the lower two-thirds of the right thoracic hemi-field, with a clear upper limit convex to the parenchyma (black arrow), pushing the mediastinum towards the contralateral side (white arrow), and erasing the homolateral diaphragmatic cupola (red arrow).

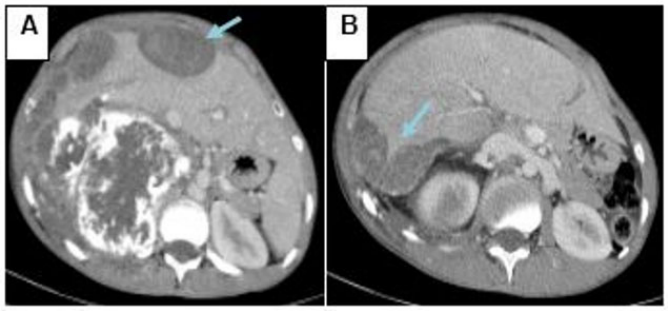


Figure 3: Abdominal CT scan with contrast injection: axial sections (A, B) showing hypodense, oval peritoneal nodules containing calcifications, scalloping on the liver.

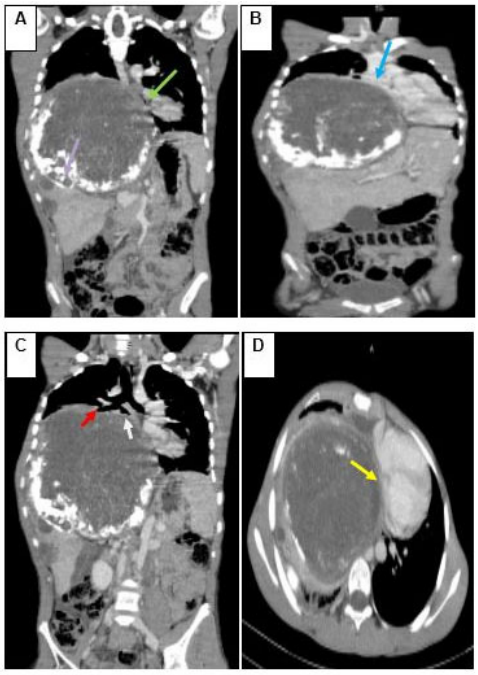


Figure 4: Thoracic-abdominal CT scan with iodinated contrast injection, coronal (A, B, C) and axial (D) sections: Right lung mass located in the lower and middle lobes, well limited, evenly contoured, hypodense, heterogeneous, containing calcifications: (A) It exerts a mass effect on the esophagus with reduction of its lumen without clear interface (green arrow). (B) It laminates the right pulmonary vein (blue arrow).

## DISCUSSION

Osteosarcoma is the most frequent malignant bone tumor in children and adolescents [1, 2], the metaphysis of the long bones is its elective site especially at the distal femur [1].

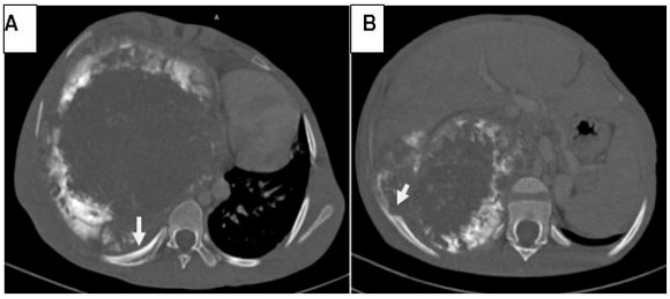


Figure 5: Thoracic CT (bone window): axial sections revealing a plurilamellar periosteal reaction of the posterior arch of K9 (A) and focal lysis of the middle arch of K10 (B).

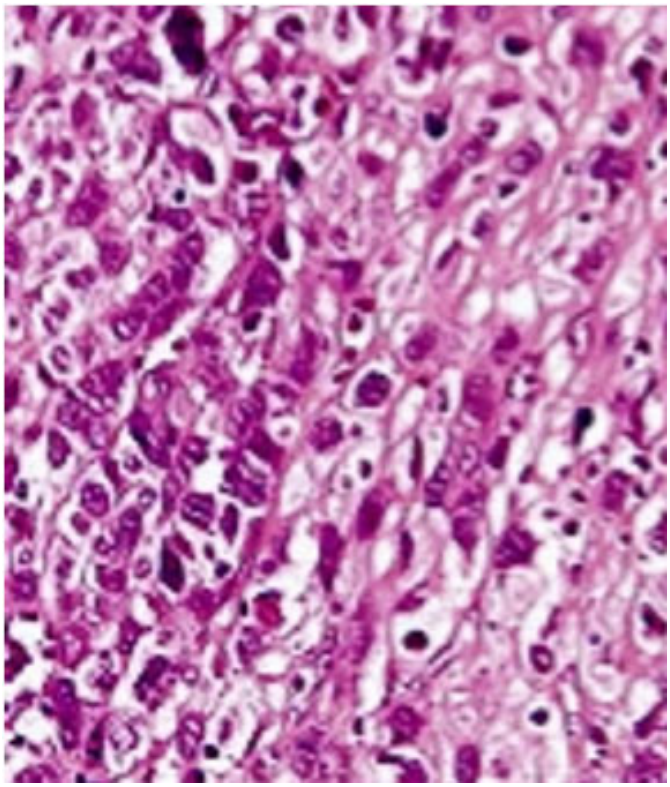

Figure 6: Pleomorphic spindloid and osteoblastic neoplastic cells with osteoid matrix.

Osteosarcoma is an aggressive tumor [3] and is characterized by a metastatic pattern dominated by pulmonary metastases (98%) followed by bone metastases (37%) [1, 3], abdominal metastases are very rare [4], secondary peritoneal localizations account for <5% of osteosarcoma metastases [5, 6], and result from the migration of tumor emboli via the bloodstream [7]. Wester et al. suggested that new chemotherapy regimens favor the occurrence of extrapulmonary metastases [1, 8]. Our patient received chemotherapy courses before the occurrence of peritoneal metastases.

Postchemotherapy surveillance is very important, a careful clinical examination and a chest CT scan, which is the examination of choice for early detection of lung metastases, should be performed on a regular basis [7, 9] monthly during the first year after chemotherapy, at three-month intervals during the second year, and every six months for two to five years, and then once a year [7]. Other paraclinical examinations are ordered according to clinical signs [7].

Currently, the National Comprehensive Cancer Network recommends that positron emission tomography (PET) and/or bone scans be considered for restaging and post-treatment surveillance.

The diagnosis of abdominal metastases is made in the presence of abdominal signs, which can be established acutely by abdominal ultrasound and/or CT [3, 7]. The abdomen should be investigated especially in case of osteosarcoma recurrence [7].

In our case, the patient presented with acute abdominal pain in the right hypochondrium. Metastases usually have the same radiological appearance as the primary tumor [5].

Abdominal metastases usually manifest as a solid tissue mass without or with calcifications [5, 7] rarely as a cystic lesion [7].

Any abdominal mass in a patient with a history of osteosarcoma should suspect abdominal metastasis even if it is cystic and without calcification [7].

In our patient, abdominal ultrasound and CT scan showed peritoneal tissue lesions with calcifications.

Lung metastases appear as multiple nodules of variable size, well limited, predominantly in the lower lobes, often sparing the apexes [10], which may or may not contain calcifications [10]. They may be the site of an excavation, especially after chemotherapy [10].

Our patient presents with a single, unexcavated lung mass in the lower and middle lobes, sparing the upper lobe, containing calcifications.

Pneumothorax is a frequent complication in patients with lung metastasis, it is due to the presence of a broncho-pleural fistula secondary to tumor necrosis [10], other radiological manifestations of lung metastasis have been reported such as micronodules, calcified adenopathies, and tumor thrombi [10], these manifestations have not been reported in our case.

Metastatic osteosarcoma has a very poor prognosis, with approximately 20–30% of long-term survivors [11]. Abdominal metastases represent a poor prognostic indicator, with a median survival after diagnosis of approximately 2–3 years [3].

## CONCLUSION

Osteosarcoma is an aggressive malignant tumor characterized by a metastatic pattern dominated by pulmonary metastases. Secondary extrapulmonary localizations, especially peritoneal metastases, are exceptional and have a poor prognosis. Monitoring of patients with osteosarcoma is essential. Imaging is effective for early detection of metastases.

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### Author Contributions

El Houssni Jihane – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

El Haddad Siham – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Dghoughi Basma – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Bassel Saber Abdellah – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Chat Latifa – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Allali Nazik – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

### Guarantor of Submission

The corresponding author is the guarantor of submission.

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None.

### Consent Statement

Written informed consent was obtained from the patient for publication of this article.

### Conflict of Interest

Authors declare no conflict of interest.

## Data Availability

All relevant data are within the paper and its Supporting Information files.

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