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# A Rare Cause of Bowel Obstruction: Peritoneal Metastases in Osteosarcoma at the Tibia in a Young Female Patient with Brain Metastasis. Case Report

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**Rezumat**

***O cauză rară de ocluzie intestinală: metastazele peritoneale de osteosarcom tibial în cazul unei paciente tinere cu metastaze cerebrale. Prezentare de caz***

Osteosarcoamele sunt cele mai frecvente tumori osoase primare maligne în rândul copiilor și adolescenților. Ca și metastazele cerebrale ale osteosarcoamelor, cele intestinale sunt foarte rar întâlnite. Prezentăm cazul unei paciente în vârstă de 23 de ani, diagnosticată și operată în 2008 de osteosarcom tibial, cu chimioterapie neoadjuvantă și adjuvantă, ce a prezentat metastaze pulmonare operate în 2014. A fost apoi diagnosticată în martie 2015 cu un process expansiv intracranian – metastaza de osteosarcom, pentru care s-a practicat ablație totală a tumorii, în perioada postoperatorie precoce fiind transferată în clinica de Chirurgie Generală pentru durere abdominală, distensie abdominală, vărsături și lipsa tranzitului intestinal pentru materii fecale și gaze. Clinic și imagistic se pune diagnosticul de ocluzie intestinală motiv pentru care se intervine chirurgical, descoperindu-se ocluzie intestinală secundară unei metastaze la nivelul ileonului terminal și metastaze hepatice, confirmate atât anatomopatologic cât și imunohistochimic ca fiind metastaze de osteosarcom. Atât metastazele intestinale cât și cele cerebrale de osteosarcom sunt entități foarte rare iar asocierea acestora, mai ales la pacienții tineri, este excepțională. Cu toate acestea metastazele intestinale trebuie luate în considerare ca și cauză posibilă de ocluzie intestinală la pacienții cu osteosarcom.

**Cuvinte cheie:** osteosarcom, ocluzie intestinală, metastaze, creier

**Abstract**

Osteosarcomas are the most frequent primary malignant bone tumors in children and adolescents. Like brain metastases in osteosarcomas, the bowel metastases are very rare. We present the case of a 23-year-old female patient, diagnosed and operated in 2008 of osteosarcoma at the tibia, for which she had sessions of neoadjuvant and adjuvant chemotherapy, but presented lungs metastases for which she underwent surgery in 2014. Then, in March 2015, she was diagnosed with an intracranial expansive process, an osteosarcoma metastasis, for which a total ablation of the tumor was performed during the early postoperatory period, being transferred to the General Surgery Clinic for abdominal pain, abdominal distention, vomiting, and lack of intestinal transit regarding faeces and intestinal gas. Both clinically and imagistically, the diagnosis was of bowel obstruction. This was the reason for performing surgery, thus discovering a bowel obstruction secondary to a metastasis of the terminal ileum and liver

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metastases that were confirmed as osteosarcoma metastases from an anatomopathological and immunohistochemical point of view. The bowel metastases and the osteosarcoma brain metastases are very rare entities and, their association, most often with young patients, is exceptional. However, bowel metastases must be taken into account as a possible cause of bowel obstruction in patients with osteosarcoma.

**Key words:** osteosarcoma, bowel obstruction, metastasis, brain

## Introduction

Osteosarcomas are the most frequent primary bone tumors in children and adolescents (1). As a result of the improvement of therapeutical management of metastatic osteosarcoma patients due to a multidisciplinary approach of chemotherapy combined with primary tumor resection as well as metastases, the survival rate of patients has improved, but the typical evolution of these tumors has also changed (2). The locations of osteosarcoma metastases are the lungs and the bones. Osteosarcoma brain metastases have an incidence of 1,8-5,6% (3), being usually associated with the presence of lungs metastases in the medical history (4). The cases of bowel metastases, which presented abdominal symptomatology, were rarely reported (5).

## Case report

*We present the case of a 23-year-old female patient who was diagnosed with osteosarcoma of the external left tibia condyle (osteosarcoma of the proximal tibia). She was administered neoadjuvant chemotherapy according to EURAMOS protocol with Doxorubicin 75 mg/m$^2$ and Cisplatin 120 mg/m$^2$, together with the administration of Methotrexate in doses which were progressively increased (from 8 g/ 4 hours to 16 g/m$^2$). In 2009, she was subjected to surgery, a tibia and fibula resection being performed (with free resection margins, intraoperatively) together with a tibia reconstruction with Mutars prosthesis. Adjuvant chemotherapy with Doxorubicin, Cisplatin, Epirubicin, and high doses of methotrexate were administered. The patient had a favorable clinical and paraclinical evolution until 2014, when she developed secondary lung tumors at the level of the left upper lobe and the left lower lobe, for which, in December 2014, an atypical left upper lobe resection, an excision of the nodules at the level of the left lower lobe and mediastinal lymphadenectomy were performed, the anatomopathological result confirming the diagnosis of osteosarcoma metastases. In February 2015, a new series of polychemotherapy sessions was attempted, but, due to a refractory thrombocytopenia, its performance was timed. Moreover, as a result of a soft tissue ultrasound, performed due to the presence of a palpable tumor in the medium third of the right leg, two nodular, hypoechogene images of approximately 18,4/9 mm and 27,4/20,8 mm, respectively were observed. They were characterized by an inhomogeneous texture, with regulated contour and vascular signal present in the medium third of the right leg. Also, two other hypoechogene, inhomogeneous nodular tumors of 30,3/16 mm and 27,5/17,5 mm were observed at the proximal extremity of the right thigh, on the lateral side. They were characterized by a vascular signal, which was more visible at the extremity and which continued to be followed up imagistically.*

*In March 2015, the patient had paresthesias at the level of the right arm and the brain MRI showed a left parietal lobe intracerebral expansive process (Fig. 1).*

*Surgery was performed again on March 26, 2015 with a total ablation of the left parietal lobe intracerebral expansive process. Both the anatomopathological and the immunohistochemistry result was osteosarcoma metastasis, with a favorable neurological and imagistic evolution, but marked by the appearance of a subocclusive syndrome at 4 days post-*

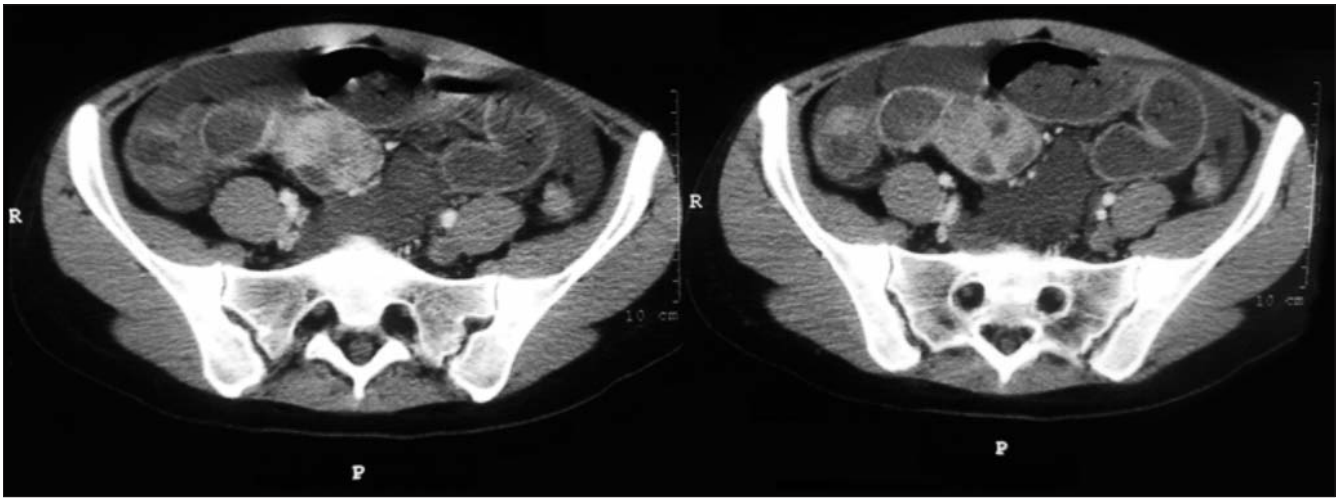


***Figure 1.*** *Preoperatory abdominal CT scan – ileum tumor*


***Figure 2.*** *Preoperatory abdominal CT scan*

*operatory, which manifested with diffuse abdominal pains, vomiting and the lack of a normal intestinal transit as far as gases were concerned. A simple abdominal X-ray was performed, which showed the presence of hydroaeric levels and then an abdominal CT scan, which highlighted tumors at the level of the V and III liver segments, whose sizes were of approximately 5/6 mm and seemed to be secondary tumors (Fig. 2).*

*Also, a high quantity of intraperitoneal fluid effusion and ring shaped sections of small intestine with very thickened walls – mostly at the level of the ileum and the right hip muscles, where two spontaneous iodophors inhomogeneous hypodense tumors, were noticed when administrating contrast substance. Their sizes were of 21/38.7 mm and 26/42,8 mm, respectively (Fig. 3).*

*An emergency surgery was performed, the intraoperatory results showing a mechanical bowel obstruction due to a hard, inhomogeneous tumor of approximately 10/8 cm, located at the level of the last ileum ring and the adjacent mesentery and also 2 liver metastases of 7-8 cm, located at the level of the III and V segments (Fig. 4).*

*A right ileal hemicolectomy with latero lateral ileotransverse anastomosis was performed together with an atypical liver resection for the secondary tumors located at the level of*

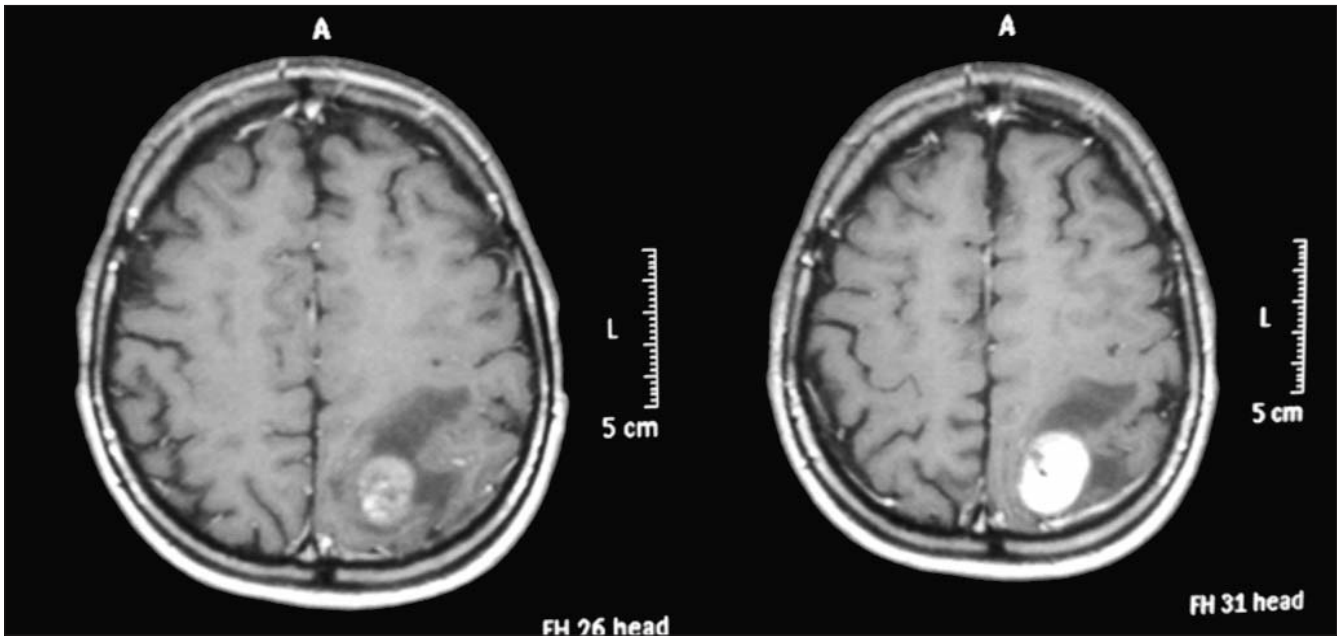


***Figure 3.*** *Postoperative brain MRI – secondary brain determination*

***Figure 4.*** *Secondary metastatic ileum osteosarcoma – intraoperatory aspect*

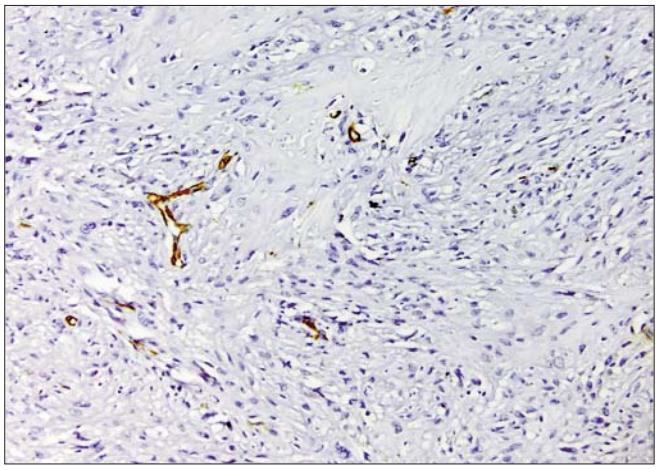

***Figure 5.*** *Positive MSA liver metastasis - immunohistochemical aspect – 200x*

*the V and III segments. Postoperatory, the patient had a clearly favorable uncomplicated evolution. Both the anatomopathological and the immunohistochemical examination revealed the diagnosis of osteosarcoma metastases at the level of the liver and ileum. The liver metastasis was the only one to form an osteoid, this also being a diagnosis element for osteosarcoma. The rest of the metastases were diagnosed by morphological (Fig. 5) and immunohistochemical similarities, except for MSA (Muscle Specific Actin) (Fig. 6), which tested positive in rare tumor cells, the rest of the markers testing negative.*

*The positive testing of one of these markers could have led the diagnosis towards another direction specific for cellular diagnosis. The GFAP-negative in brain metastasis excludes the primitively brain origin of the tumor (for example the glioblastoma) (Fig. 7).*

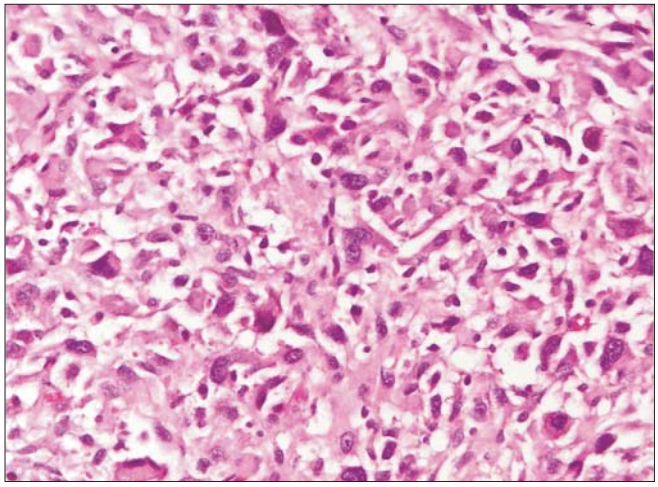

***Figure 6.*** *HE liver metastasis – 400x*

## Discussions

The small-bowel obstructions can be caused by biliary calculi, ties, volvuli, invagination, or tumors. The tumor pathology of the small-bowel often raises diagnosis problems, both by an uncharacteristic clinical presentation and by the rarity of these tumors at this level. The primary neoplasia of the small intestine represents 1-5% of all the neoplasias of the digestive tract, the ileum being the location in which neoplasias often develop, metastatic tumors being rarely met (6). The osteosarcoma metastases at the level of the ileum are rarely met, being described until present in approximately 10 cases, mainly in adults (7).

Regarding the brain metastases, it was estimated that approximately 3% is represented by sarcoma metastases and between 1 and 8% of the patients with sarcoma can develop brain metastases (8,9). Most often, osteosarcoma brain metastases appear after lungs metastases, although there have been cases of brain metastases without the existence of any lungs metastases. As most of the lesions are usually single, the surgical excision by chemotherapy and palliative radiotherapy has

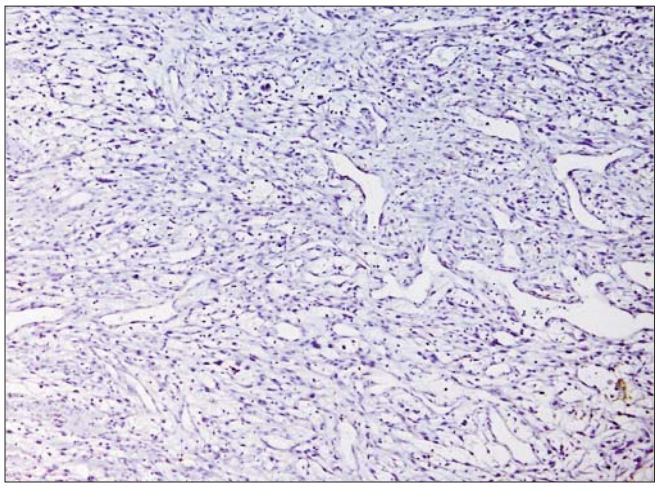

***Figure 7.*** *GFAP-negative in brain metastasis – immunohistochemical aspect - 100x*

proven to be a standard treatment (10).

In the case presented, the diagnosis of bowel obstruction was delayed, taking into account that the patient was in an early postoperatory stage, when a bowel paresis after total anesthesia was normal, only 2 having passed from the resection of the brain tumor at the moment the digestive symptoms have started. The digestive symptoms have first manifested as a subocclusive syndrome, the whole picture being defined in approximately 4 days postoperatory. Moreover, considering that the bowel metastases are very rare in young patients with osteosarcoma, the mechanical cause of bowel obstruction has only been taken into account after performing the abdominal CT scan.

Unfortunately, no matter the therapeutic treatments applied, the prognosis is very low, taking into account the association of lung, brain, and bowel metastases (7).

Although some therapies do not aim this category of patients anymore, surgery is necessary to solve the problem of occlusive syndrome and to try to control the metastatic disease, mostly in case of unique secondary tumor (11).

## Conclusions

Both the metastatic osteosarcoma of the bowel and of the brain are very rare, their association, especially in young patients, like in the case presented, being almost exceptional, this leading to the delay in establishing the correct diagnosis. That is why the metastatic osteosarcoma of the bowel has to be taken into account as a possible cause of mechanical obstruction, most often in patients who present secondary lung tumors in medical history.

***Conflict of interest:*** None declared.

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