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Bone Metastases from a Gastric Stromal Tumor: A Case Report and Literature Review

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Lamiae Amaadour*[1], Hajar Ouahbi[1], Zineb Benbrahim[1], Nezar El Bouardi[2], Youssef Alaoui Lamrani[2], Fatimazahra El Mrabet[1], Samia Arifi[1], Nawfel Mellas[1]

[1]Department of medical oncology, [2]Department of radiology,
Hassan II University Hospital, Fez, Morocco

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ABSTRACT

Gastrointestinal stromal tumors (GISTs) are the most frequently diagnosed mesenchymateuse tumors of the gastrointestinal tract [1]. Liver and peritoneum are the most common metastatic sites, whereas GISTs rarely metastasize to the bone [2]. There are no consensus about the treatment of GISTs' bone metastases. We present here a case of a 61-year-old man with synchronous gastric GIST and bone metastases, and we briefly review the existing data about this rare entity.

BACKGROUND:

GISTs most frequently metastasize to the liver and peritoneum. Bone metastases are uncommon. Their incidence among GIST's different metastatic locations has been estimated to be roughly 5% or less [3]. The purpose of this paper and a brief literature review is to discuss the clinical characteristic, imaging features, and management of this unusual metastatic location of GIST.

CASE REPORT:

A 61 year-old man presented with hematemesis, vomiting and back pain lasting more than three months. Physical examination found a painless gastric mass; neurological examination was normal. A gastric endoscopy revealed an extrinsic growth of bulb whose biopsy was negative. A thoraco-abdomino-pelvic computed tomography (CT) scan showed a 64 X 77mm abdominal tumor lesion above the stomach(Figure 1) and multiple bone lesions of the spine with medullar extension, pelvis(Figure 2), humerus and sternum. Magnetic resonance imaging of the spine confirmed the vertebral bone lesions, and notably showed a L2 – S3intraduralosteolytic mass with medullar compression (Figure 3). Pathologic analysis of CT-scan guided biopsy ofgastric tumor and one bone metastasis revealed a spindle-cell tumor characterised by few mitosis, no necrosis, and an immunochemistry staining strongly positive for CD117 and CD34 in both specimens (Figure 4). Thus the diagnosis of gastric GIST with bone metastases was confirmed. The patient received a radiation therapy at the intradural lesion with a total dose of 30 Gray, then he started treatment with oral Imatinib at a daily dose of 400mg, and zoledronic acid.After three months of treatment, which was perfectly tolerated,the clinical response was good and the patient's pain resolved and CT-scanshowed regression in primary tumor and stabilization of bone lesions. Currently, the patient continues Imatinib, without any symptoms related to the tumor lesions, with a radiological disease stability thirty months on follow-up CT.

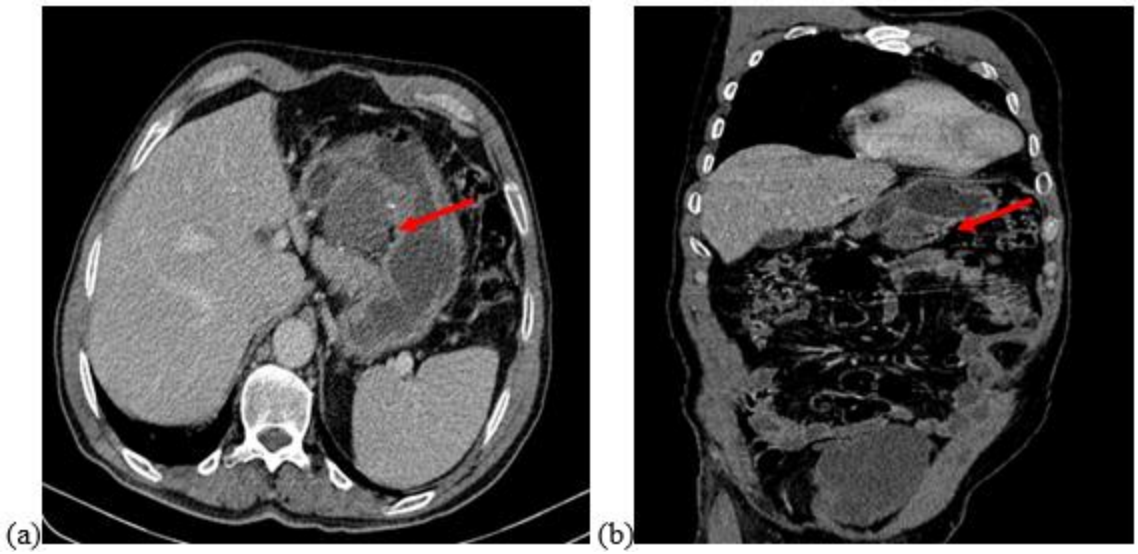

Figure 1: Enhanced CT scan in axial (a) and coronal (b) planes showing the gastric GIST.

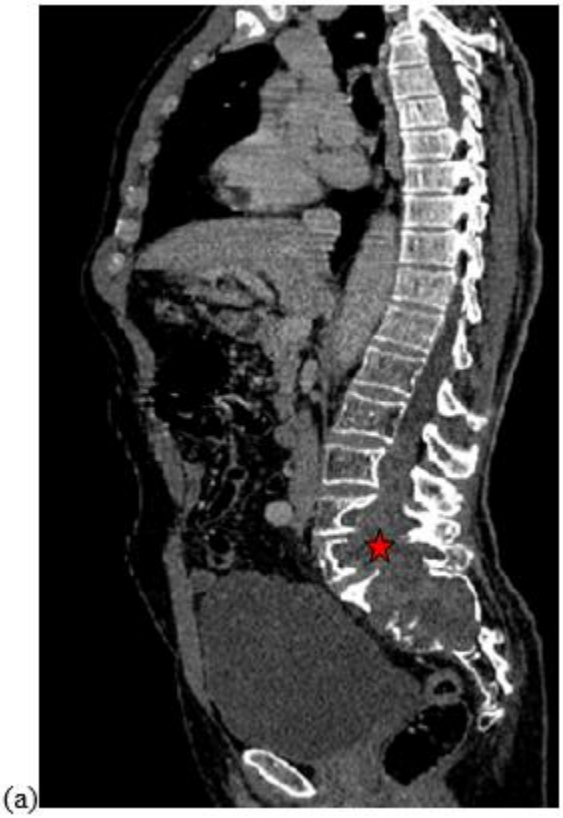

Figure 2: Enhanced Ct scan in a the sagittal (a) and axial planes (b,c) showing the osteolytic L2-S1 bone lesion with intradural extension (arrow) and dense (ilium) bone lesions (star).

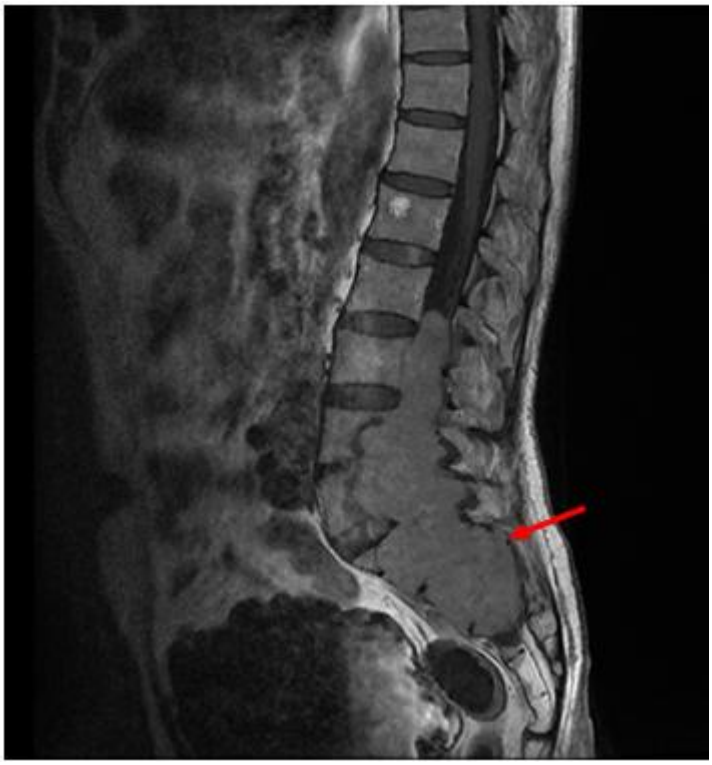

Figure 3: Magnetic resonance imaging of the spine in the sagittal plane showing the large vertebral mass with intradural extension.

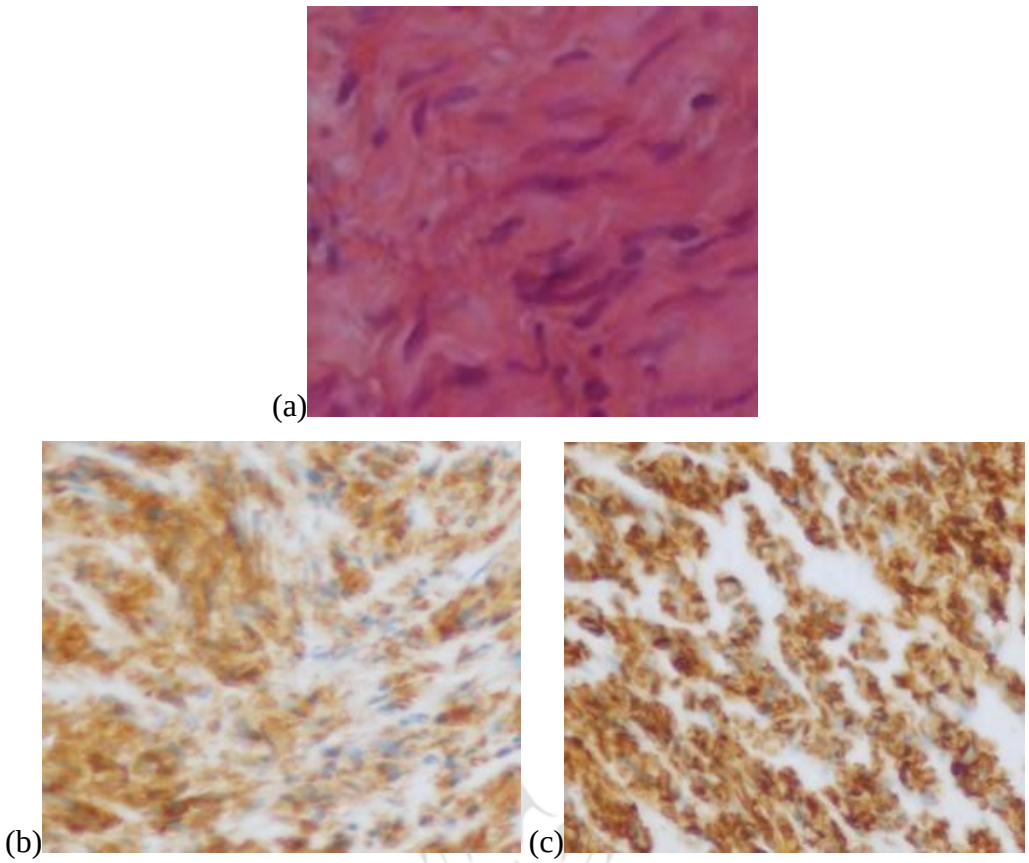

Figure 4 : Gastric GIST biopsy : (a) Spindle cells with few mitosis. (HE X400). Positive immuno staining with CD117 (b) and CD34 (c) (HEX400).

DISCUSSION:

GISTs have an uncertain clinical behavior ranging from benign to frankly malignant, making the outcome totally unpredictable. In localized cases, GISTs are categorized into low-, intermediate, or high-risk group, depending on the tumor size, mitotic index and anatomical location of the primary tumor [4]. The most common metastatic locations of GISTs are the liver and peritoneum. Bone metastases are extremely rare [16], and their incidence among different metastatic locations has been estimated to be approximately 5% or less [9,16]. However, the specific characteristics of patients with bone metastasis have not yet been identified.The location of the stomach, like in our case, was the most frequent location of the primary GIST, followed by the ileo-jejunum, the rectum, and the duodenum [5]. Bone metastases of oesophageal and mesenteric GIST were report in one case each [6,7]. Contrary to our patient, most of the reported cases described a high number of mitoses of over 5/50 high-power fields [5]. All cases showed

kit positive immunochemistry stain [5]. Bone metastases can be diagnosed rarely at disease presentation, and more frequently during the follow-up of treated primary GISTs within a median delay of 48 months [range: 4–120 months] [5,8]. Our case is one of the rare observations suggesting that the diagnoses of primary tumor and bone metastases can be synchronous [5].

Spine and pelvis were the most frequent sites of bone metastases reported in case series [5,8,9]. Bone metastases mostly occurred concomitantly with or after other metastatic sites. They can be rarely the only metastatic manifestation [9]. The bone metastases from GIST are classically lytic and well-defined lesions [3]. In the best of our knowledge, our case is the first to showa dense bone lesion. The available literature does not provide consistent data on the treatment of bone metastases in GISTs. The use of Imatinib is the standard of care of advanced GISTs, enabling the long-term survival of patients [10]. It has shown response in 50% patients, and in approximately 75–85%, patients have at least stable disease. Imatinib wasproven also effective in the treatment of bone metastases of GISTs [10]. A median survival of 17 months [range: 3–40] was reported in a series of 13 patients with GIST metastases to bone [9]. Thirty months of response were recorded in our paper. Local treatment of bone metastases can be offered to selected patients.Surgery, sometimes associated with graft or prosthesis, can be realized for patients with functional impairment or oligo metastatic disease for different skeletal localizations. Some papers suggest long-term disease-free survival benefit in patients who underwent complex surgical bone resections with negative margins [11,12]. Palliative radiotherapy was associated with a significant improvement of bone pain [5]. The concomitant use of imatinib during radiotherapy is well tolerated [15]. Interestingly, in our case, radiotherapy allowed a satisfying analgesic effect in the vertebral mass. Bisphosphonates seem to be also effective for the management of GIST's bone metastases [8]. Only 6 cases in the literature, including ours, report their use [8,13,14]. 1/6 case of jaw osteonecrosis was described after imatinib and zoledronicacid concomitant use [8].

CONCLUSION

Bone metastases originating from GISTs are rare. Their frequency is increasing because of the improvement of patients outcome in the tyrosine-kinase inhibitors era. Our work emphasis on careful evaluation of any suspicious bone lesions especially in the absence of other metastatic

sites. Imatinib, palliative radiotherapy and bisphosphonates are effective therapeutic options that improve symptoms and prolong survival.

Disclosure:

The authors declare no conflict of interest.

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