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Intra-abdominal telangiectatic osteosarcoma: a case report

M. P. Kgagudi[1*], N. Mahlatsi[2] and M. Jingo[1]

Abstract

Background Telangiectatic osteosarcoma is rare and it rarely affects flat bones, especially the bones of the pelvis. It is uncommon for telangiectatic osteosarcoma to be considered as a differential diagnosis when assessing a large intrabdominal mass.

Case report We present our case of a 33-year-old African female who presented with a sizeable telangiectatic osteosarcoma of the left iliac bone. She reported a 3-year duration of a painless, slow-growing mass arising from the left flank. At examination, a large bony hard mass extending from the left ilium to the umbilicus was noted, almost mimicking an intra-abdominal pregnancy. All laboratory tests were within normal limits and an unconventional surgical approach was used for a one-stage excision of the tumor without complications. The definitive histopathological diagnosis postexcision was that of a telangiectatic osteosarcoma only on the second review of the histological specimen.

Conclusions Pelvic telangiectatic osteosarcoma is rare, and the ilium is the commonly affected pelvic bone. These tumors can be sizeable at presentation with intra-abdominal or pelvic extension with a high chance of misdiagnosis. Fortunately surrounding soft tissue involvement seems to be a rare and late finding when present.

Keywords Telangiectatic osteosarcoma, Pelvic mass, Intra-abdominal mass

Introduction

Osteosarcoma (OS) is the most common primary malignant bone tumor with multiple histological variants [1, 2]. Telangiectatic osteosarcoma (TOS), a rare subtype of OS, is histologically characterized by dilated blood-filled cavities with sarcomatous features [1–3]. Fortunately, TOS accounts for only 4% of all OS cases [1–3]. The pelvis is rarely affected by TOS with the literature reporting that only 3.1% of cases of TOS occur in the pelvis [3, 4]. We present our case of iliac TOS as a rare case presentation.

*Correspondence:
M. P. Kgagudi
dr.pkgagudi@gmail.com
[1] Division of Orthopaedic Surgery, School of Clinical Medicine, Faculty of Health Sciences, University of the Witwatersrand, Johannesburg, South Africa
[2] Chris Hani Baragwanath Academic Hospital, Johannesburg, South Africa

Case report

Ms. P. L. was a 33-year-old African female who presented to our tumor clinic after she was referred by her local treating hospital. Her main complaint was that of a slow-growing mass around the left groin for a period of 3 years. She reported recent pain and discomfort but no changes in her bowel habits. She had an unremarkable background medical history. On examination, she was clinically stable with a large, firm, irregular mass spanning from the left iliac bone to just above the umbilicus (almost mimicking a gravid uterus of about 20 weeks). Systemic and distal neurological examinations were both normal. Pelvic radiography in Fig. 1, showed a massive osteolytic expansile lesion eroding the upper outer half of the left ilium with intralesional calcifications. On T2 magnetic resonance imaging (MRI), images of the lesion showed a

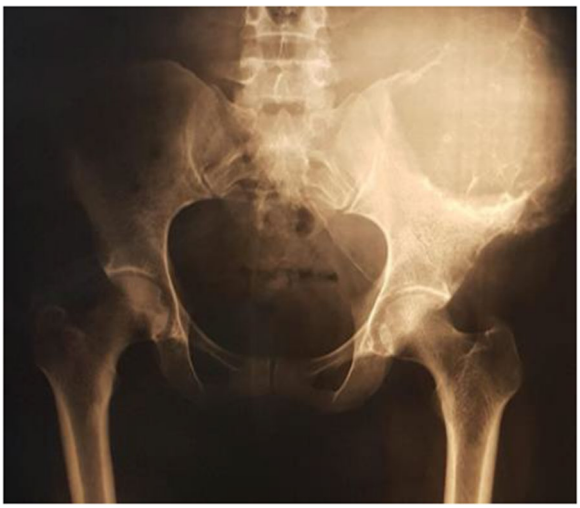

Fig. 1 Anteroposterior view of the pelvis

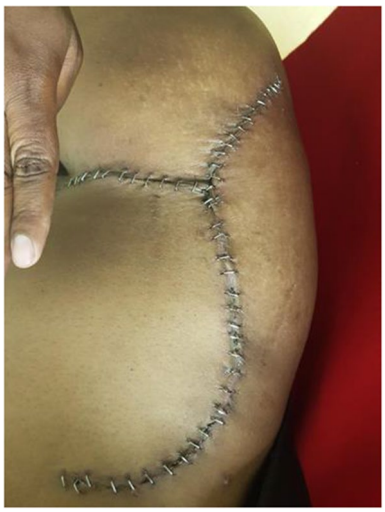

Fig. 3 Postexcision surgical scar

large heterogeneous intra-abdominal lesion displacing the large intestine with obvious fluid–fluid levels (see Fig. 2).

Her laboratory and metastatic workup were normal. She underwent surgical excision of the tumor with a stellate incision based on the anterosuperior iliac spine to facilitate anterior and posterior access for resection (Fig. 3). The mass, which measured 260 mm × 180 mm × 80 mm and weighed 1000 g, was excised in totality, thus leaving behind a raw cancellous iliac bone. Hemostasis and stability of the pelvis were assessed and noted at wound closure with no need for embolization or reconstruction, respectively. The final histological diagnosis was that of telangiectatic osteosarcoma of the ileum. At the final follow-up, the wounds had healed and the patient had no new complaints.

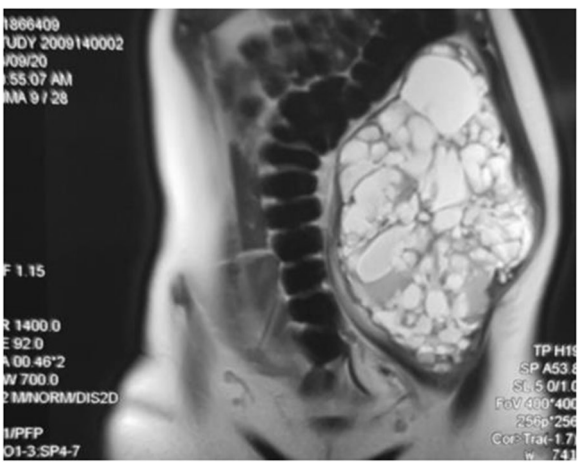

Fig. 2 MRI of the lesion

Discussion

Telangiectatic osteosarcoma is a condition that has a predilection for long bones and it rarely affects the bones of the pelvis [1, 3–5]. About 3.1% of all cases of TOS are found in the pelvis and sacral region [3–5]. TOS of the pelvis should be considered in individuals between the ages of 30 to 40 years [2]. Our patient was 33-year-old and female at diagnosis, although commonly this tumor has a slight male preponderance [2]. Pelvic TOS presents similarly to TOS in the extremities [4]. Most patients report a painful mass of gradual slow growth over time [4, 5]. Our case of discussion denied a history of a painful mass and only reported a recent sensation of discomfort, hence the large mass at presentation.

Radiologically, the distinction between telangiectatic osteosarcoma versus giant cell tumor (GCT) versus aneurysmal bone cyst (ABC) is challenging but feasible in cases of extremity TOS [1]. The eccentric, expansile appearance of an ABC and the classic abutment of epiphysis in GCT could help the course of diagnosis [1]. However those features of distinction are difficult to interpret in radiographs of the flat bones of the pelvis. MRI images of all three conditions also further confuse the situation as they can all depict large cysts with fluid–fluid levels [2, 3]. Our case showed a large expansile lesion with multiple large fluid–fluid levels on T1 MRI images. Histologically the presence of malignant cells along the cysts separating septae with osteoid formation are highly suggestive of TOS [2–4]. The case of discussion revealed no malignant cells but had a substantial amount of osteoid formations on the second review of the excised tumor mass, and hence an amendment to the initial histology report was made.

The pelvis is an intricate anatomical region with multiple vital organs in close relation [6]. Telangiectatic osteosarcomas are inherently expansile in nature and with a potential for growth to large sizeable masses that are largest in the pelvis [2, 7]. Due to their malignant behavior, the gold standard of treatment is wide marginal excision with or without reconstruction when dealing with extremity TOS [2]. In the pelvis, however, wide-margin excision is potentially impossible due to the intimate anatomical relations and it is prone to cause morbidity, and therefore early diagnosis could lead to a smaller simpler excision with minimal residual functional fallout [4, 8]. Fortunately, at resection for our case, the mass could be excised whole with a small amount of attached mesenteric tissue at analysis with no complications noted.

Delayed or misdiagnosis is not uncommon when dealing with TOS of the ilium [3, 4]. Tumor surgeons propose an open biopsy as a gold standard when suspecting TOS [3]. The latter is based on the diagnosis hinging on the presence of malignant cells in the septae of the blood-filled cavities, therefore making the distinction of the condition from ABC and/ GCT [4, 5]. True and testament to the latter statement is that in our experience, the whole tumor specimen was submitted for histological analysis but making the diagnosis was still challenging at the first tissue analysis. Fortunately, our patient was found to be asymptomatic at the last follow-up assessment. However, in our case and also in the literature, a complete resection favors long-term survival, especially in the absence of metastasis as no strong scientific evidence exists for treatment protocols due to the rarity of this subtype of osteosarcoma [7, 8]. Our case had no local or distant recurrence at 3 years even though abdominopelvic sarcomas have been shown to have a high recurrence rate [7].

Conclusion

Pelvic TOS is rare. These tumors can be sizeable at presentation with intraabdominal or pelvic extension with a high chance of misdiagnosis. Fortunately surrounding soft tissue involvement seems to be a rare and a late finding when present. TOS should be considered when dealing with large osteolytic lesions affecting the ileum extending into the abdomen.

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Author contributions
MPK carried out the conceptualization, literature review, ethics application, and paper review. NM carried out the conceptualization, literature review, and write up. MJ carried out paper supervision and review.

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Not applicable.

Data availability
All relevant data pertaining to the case is available for perusal by reviewers and Editor-in-Chief.

Declarations

Ethics approval and consent to participate
Institutional ethics was approved: references number: M230557.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
Not applicable.

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