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Recurrent adamantinoma metastatic to the spine: Clinical and imaging considerations ☆

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ABSTRACT

Metastatic adamantinoma involving the spine is an extremely rare occurrence. In this case report, we present a patient with recurrent adamantinoma of the tibia which was found to have metastasized to the spine. The metastatic involvement was diagnosed pathologically, status post CT guided percutaneous core needle biopsy, performed after the patient returned with concerning symptoms and imaging findings suggestive of metastasis. The patient was ultimately treated surgically with vertebrectomy and reconstruction. A thoughtful review of this disease process is explored, emphasizing the pathology, imaging characteristics, and pertinent differential diagnostic considerations. While uncommon, knowledge of this rare disease process and its presentation can improve future patient diagnosis and outcomes.

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Introduction

Adamantinomas are very rare primary malignancies that typically occur in the tibial diaphysis of young patients. These lesions typically represent less than 1% of all primary bone neoplasms and have been seen to metastasize in less than 10% of all reported cases [1]. A low-grade malignancy, adamantinoma has rarely been identified to metastasize distantly, primarily to the lung, osseous structures, lymph nodes, pericardium, and liver. Metastatic involvement of the spine is an extremely rare entity with only 1 case documented in the literature to date [2]. To put this into perspective, spinal metastases occur in approximately 40% of patients who succumb to cancer, and at least 5–10% of all patients suffering from cancer will present with symptomatic spinal metastases at some point during their disease course [3]. Among all of the osseous struc-

tures which are affected by metastatic disease, the vertebrae are the third most common location, trailing only the liver and lung [3]. In this case report, we present a patient with recurrent adamantinoma of the tibia with metastasis to the spine. This case report presents a diagnostic approach based on clinical, laboratory, and imaging characteristics.

Case report

A 33-year-old woman with a past medical history of diabetes mellitus and hypothyroidism initially presented as a transfer from an outside hospital with a pathologic left tibia fracture, status post fall 2 days prior. Additional history obtained at the time of presentation included reports of a non-painful palpable abnormality along the mid anterior aspect of the left shin. On admission, she was complaining of sharp pain in the leg and an inability to bear weight. Radiographs of

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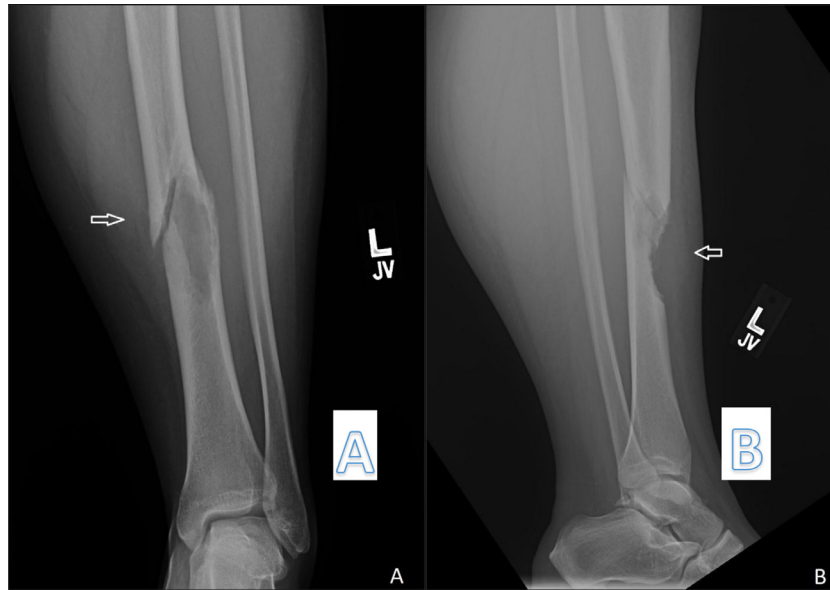


Fig. 1 – Radiographs of the left tibia at initial presentation. (A) AP radiograph with a multilocular, slightly expansile, osteolytic cortical lesion and a mildly displaced pathologic fracture. (B) Lateral radiograph demonstrates the aforementioned findings, with increased conspicuity of an anterior soft tissue component.

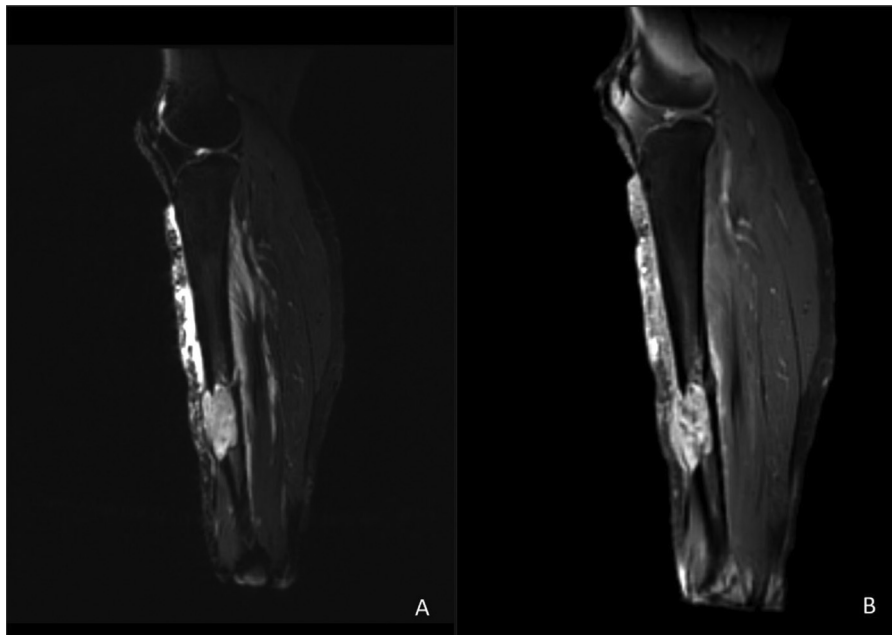


Fig. 2 – Initial MRI of the left tibia. (A) Sagittal STIR MRI of the left tibia demonstrates a smoothly lobulated, cortically based lesion with hyperintense STIR signal. There is evidence of an exophytic component as well as adjacent edema within both the nearby bone marrow and superficial soft tissues. (B) Fat suppressed post-contrast enhanced T1 sagittal MRI of the left tibia shows avid, heterogeneous internal enhancement.



Fig. 3 – Initial CT of the spine. (A) Sagittal and (B) axial images of the lower thoracic/upper lumbar spine demonstrate extensive vertebral body destructive changes with the involvement of posterior elements. Additionally, there is a bulky epidural component that extends into the paraspinal soft tissues. Multilevel involvement of the neural foramina and spinal canal are noted which result in significant spinal canal narrowing and cord compression.

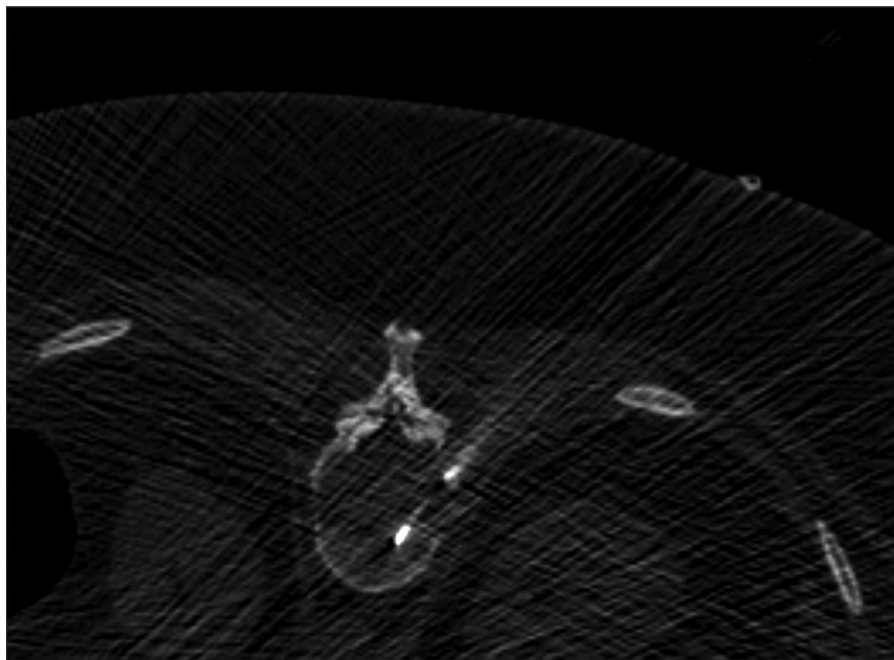


Fig. 4 – CT-guided percutaneous biopsy. (A) Axial image at the level of the T12 vertebral body demonstrates a lytic, destructive osseous and soft tissue lesion with biopsy device in a satisfactory position, utilizing a posterior parapedicular approach.



Fig. 5 – (A) 3D MIP demonstrate intense hypermetabolic activity within multiple vertebral body levels in the lower thoracic and upper lumbar spine.

the left tibia obtained at the outside facility demonstrated a lytic osseous lesion involving the mid anterior tibial cortex with an associated soft tissue mass and minimally displaced pathologic fracture [Fig. 1].

A pre-operative MRI of the left tibia was performed which revealed a heterogeneously enhancing lesion, with an associated soft tissue lesion and edema, centered within the anterior cortex [Fig. 2]. A differential diagnosis was provided at this time and included: adamantinoma, osteofibrous dysplasia, Langerhans cell histiocytosis, and less likely Ewing's sarcoma. The patient was subsequently taken to the operating room for open biopsy of the left tibia lesion which on pathology proved to be an adamantinoma, classic type. Immunohistochemical stains performed with appropriate controls showed the tumor cells were positive for Pancytokeratin-MNF-116, CK-19, CK5/6, p63, D2-40, Epithelial Membrane Antigen (focal), Vimentin, Smooth Muscle Actin (SMA) and negative for CK7, TLE-1 and Desmin, consistent with the above diagnosis. The patient subsequently returned to the hospital for radical resection with allograft reconstruction. Concern for tumor recurrence on surveillance imaging required several additional surgeries, including further bone and soft tissue resections with additional reconstructions.

Approximately 5 years after the initial presentation, the patient presented to the emergency department with complaints of back pain and muscle spasms. No focal neurologic symptoms were present. A CT of the spine was performed which showed extensive vertebral body destructive changes at T7-T10 and T12 with the involvement of posterior elements and extensive epidural tumor resulting in severe cord

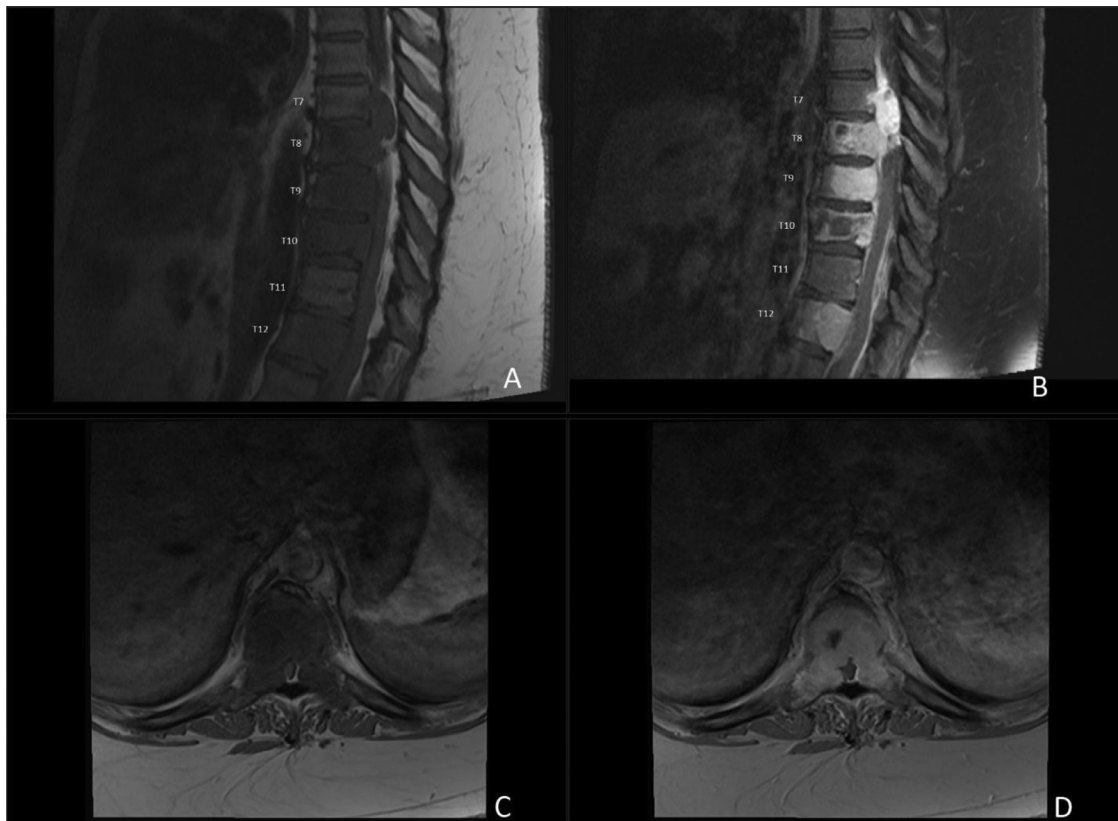


Fig. 6 – MRI of the spine. (A) Sagittal precontrast T1 of the lower thoracic spine shows abnormal hypointense T1 signal within several vertebral bodies. Notably, there is a large epidural component extending posteriorly at the T7-T8 level. (B) Postcontrast sagittal T1 demonstrates avid enhancement of the tumor. (C, D) Axial postcontrast T1 images demonstrate extensive enhancing epidural component which results in severe canal narrowing and cord compression.



Fig. 7 – (A) Sagittal STIR and (B) T2 of the lower thoracic spine show increased signal within both the osseous and soft tissue components.



Fig. 8 – (A) AP and (B) Lateral radiographs of the left tibia/fibula show post-operative changes related to tibial osseous graft reconstruction with placement of an intramedullary rod and interlocking screws. There is evidence of callus formation/healing at the proximal and distal tibial graft sites. The mid fibula diaphysis has been resected.

compression [Fig. 3]. Before performing a biopsy, whole-body PET/CT was performed for staging purposes which showed intense hypermetabolic activity within lower thoracic and upper lumbar spine vertebral body levels [Fig. 5]. A percutaneous CT-guided biopsy of T12 was subsequently performed, [Fig 4] revealing metastatic adamantinoma.

Post-biopsy and pre-operative MRI of the spine performed showed extensive bone marrow replacing lesions extending from T7-T12 with associated multilevel pathologic fractures. These lesions were T1 hypointense, T2/STIR hyperintense, and demonstrated avid post-contrast enhancement. Additional epidural involvement was noted, spanning T6-T12 with severe cord compression and edema [Fig. 6]. The patient was again taken to the operating room for a posterior lumbar decompression (T7 and L1), partial vertebrectomy (T7-T9 and T12), posterior fixation and fusion (T5-L3), and nerve root ligation (x2 at T8 on the left and T9 on the right). The patient's postoperative course was uncomplicated and she was ultimately discharged from the hospital on postoperative day 10.

Post-operative radiographs of the left tibia demonstrate post-surgical changes related to tibial osseous graft reconstruction with placement of an intramedullary rod and interlocking screws. There is evidence of callus formation/healing at the proximal and distal tibial graft sites. The mid fibula diaphysis has been resected [Fig. 8]. Post-operative radiographs of the thoracolumbar spine show posterior fusion hardware spanning T5-L3. Anterior wedging is appreciated at the T10-T12 levels [Figs. 9 and 10].

Discussion

Adamantinomas are extremely rare primary malignant neoplasms which predominantly affect patients between the ages of ten and fifty, with a slight male preponderance [4]. A

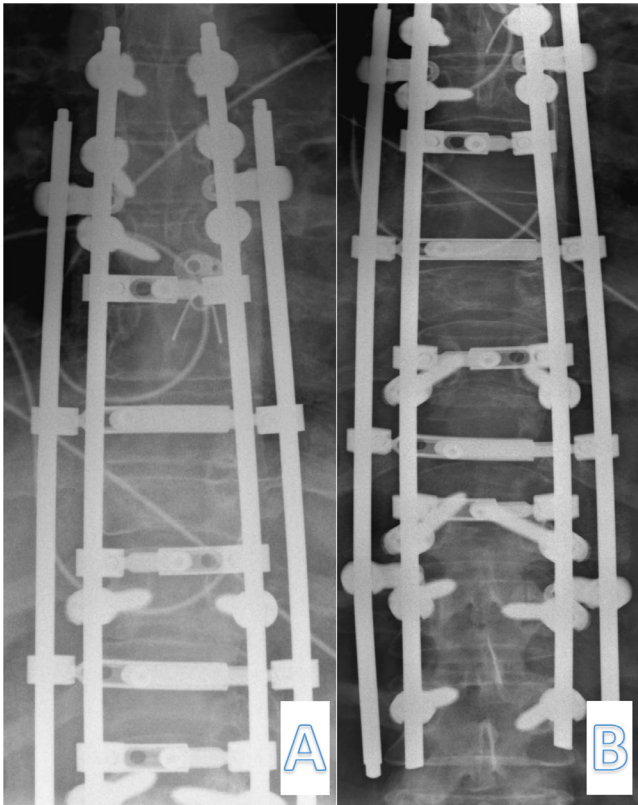


Fig. 9 – (A) Upper and (B) Lower AP radiographs of the thoracolumbar spine demonstrate posterior fusion changes spanning T5-L3. Anterior wedging is appreciated at the T10-T12 levels.

locally aggressive tumor, adamantinomas have been reported to metastasize in approximately 10% to 20% of cases with the most common sites being the lungs and less likely lymph nodes. Metastasis to a secondary skeletal site is exceedingly rare, particularly to the spine where there is only one prior report (reference) [2,4]. Primary adamantinoma of the spine is also rare, with six cases described in the literature. Whether primary or metastatic, growth within the spine has been described as slow and with a propensity for local recurrence despite treatment [5]. In a case report by Dini et al, a primary adamantinoma of the spine was described which involved multiple levels of the lower cervical/upper thoracic spine, including the lateral masses and transverse processes [6].

The differential diagnosis of adamantinomatous lesions, in general, includes chondromyxoid fibroma, fibrous dysplasia, osteofibrous dysplasia, Ewing sarcoma, and osteosarcoma [2,7,8]. Radiographic and CT imaging characteristics of adamantinoma include a multilocular or slightly expansile and lytic, cortically based lesion with scattered areas of lysis interspersed with sclerosis and a relative lack of periosteal reaction [9,10,11]. MRI findings of adamantinoma are largely nonspecific, but include a solitary lobulated lesion which may or may not contain a fluid-fluid level(s). There is typically increased T2 weighted signal with avid post-contrast enhancement [12]. In regards to the patient that we are presenting, the primary tibial lesion (and spinal metastases) did not demon-

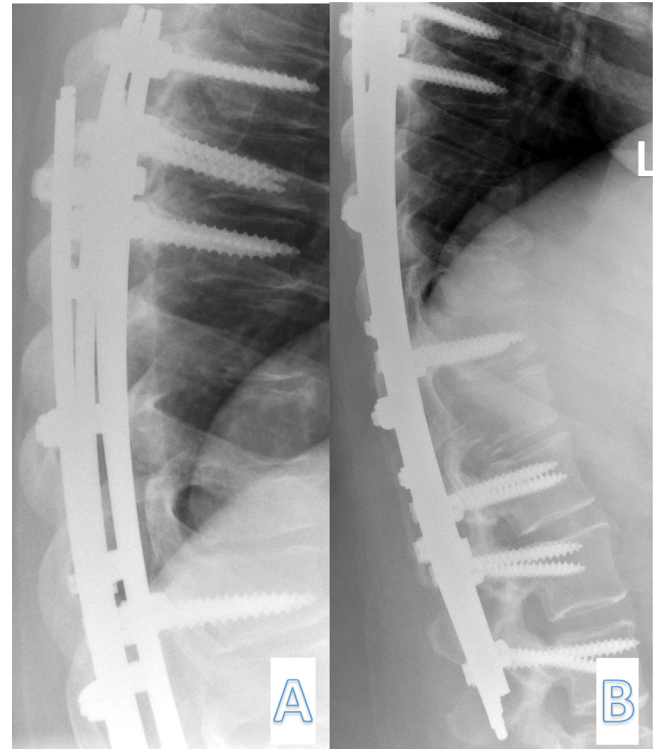


Fig. 10 – (A) Upper and (B) Lower lateral AP radiographs of the thoracolumbar spine demonstrate posterior fusion changes spanning T5-L3. Anterior wedging is appreciated at the T10-T12 levels.

strate fluid-fluid levels. The lesions did, however, show typical intrinsic T1 prolongation, T2/STIR hyperintensity, and avid post-contrast enhancement Figure 7.

Treatment of adamantinoma is most effective with wide surgical excision with/without bone graft reconstruction. In the case of tibial adamantinoma, amputation is rarely indicated [13]. The probability of local recurrence of this lesion is directly dependent upon the success of index surgical resection, ideally including clear margins. Adamantinoma is highly resistant to both chemotherapy and radiotherapy [2,14]. However, there is an overall insufficient amount of data to comment on the optimal treatment of metastatic adamantinomatous lesions involving the spine [15].

Conclusion

We present a patient with pathologically proven recurrent adamantinoma of the tibia with metastatic involvement of the spine. The diagnosis was established by taking into consideration imaging findings, pathologic specimens, and clinical context. While metastatic spinal involvement of adamantinoma is exceedingly rare, it behooves the clinician to remain vigilant of this entity when findings of adamantinoma are made on cross-sectional imaging and additional lesions are identified and/or suspected. Treatment discussions should be made on a case-by-case basis with thoughtful consideration of other

potential comorbidities, especially if surgical management is entertained.

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Nil.

Edited patient consent

The report makes use of anonymized images from which the individual cannot be identified, do not contain any identifying marks and are not accompanied by text that might identify the individual concerned.

Patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in this report. The patients understand that their names and/or initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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