CASE REPORT

Osteofibrous Dysplasia-like Adamantinoma Mimicking Bone Metastasis in a Patient With Rectosigmoid Cancer

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ABSTRACT

A patient with underlying rectosigmoid cancer presented with solitary bone lesion at left tibia. Eventhough, solitary bone metastasis particularly in the appendicular skeleton is rare in rectosigmoid cancer, it remained as the most likely initial diagnosis. However, after further characterisation by various imaging modalities and subsequent biopsy, the lesion proved to be an osteofibrous dysplasia like-adamantinoma (OFD-like adamantinoma), which is a subtype of adamantinoma. Being a rare primary bone tumour, adamantinoma and its subtypes are infrequently thought of in the initial working diagnosis of a patient with known malignancy who presents with solitary bone lesion. We present here a case of OFD-like adamantinoma in a patient with underlying rectosigmoid cancer, which mimic a metastastic bone lesion.

Keywords: Osteofibrous dysplasia-like adamantinoma, Bone metastases, Rectosigmoid cancer, Bone scintigraphy, SPECT-CT

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INTRODUCTION

The presentation of bone pain in a patient with underlying malignancy usually points towards bone metastases until it is proven otherwise. In the process of validating the presence of bone metastases, various imaging methods are employed for localisation, characterisation as well as assessing the true extent of disease. These include both radiological imaging [e.g. plain radiographs, computed tomography (CT) and magnetic resonance imaging (MRI)] and nuclear medicine imaging (e.g. bone scintigraphy, SPECT-CT and PET-CT). However, in some patients, the diagnostic dilemma on the true nature of the lesion remains, particularly in the presence of solitary lesion. We present a patient with underlying rectosigmoid cancer who presented with solitary left leg lesion. Despite undergoing a plain radiograph, MRI and bone scintigraphy with SPECT-CT, the diagnostic dilemma remained until a biopsy was performed. A lesion which was initially thought to represent a metastatic lesion, proved to be an OFD-like adamantinoma. To our current knowledge, there are no other published cases of OFD like-adamantinoma occurring in a patient with underlying rectosigmoid cancer.

CASE REPORT

A 45 year old lady presented with left leg pain for one month which was continuous and throbbing in nature. The pain was exaggerated by walking and relieved by rest. There is no history of trauma. There were also no constitutional symptoms such as fever, night sweats or history of contact with TB patients. She has underlying rectosigmoid adenocarcinoma (Stage 3), which was diagnosed 2 years ago for which she underwent open anterior resection. On examination of the left leg, there was no swelling, no tenderness, no warmth and no skin changes overlying the site of pain. X-ray of left tibia was done and it showed a lytic lesion at distal third of tibia. MRI was done to further characterize this lesion. It showed abnormal marrow signal at the distal 3rd of the left tibia (diaphysis) that is heteregenously isointense to muscle in T1W, hyperintense in T2W and TIRM/FS sequences and enhanced avidly in the post contrast sequences (Fig. 1). The differential diagnoses based on these features were metastasis or focal osteomyelitis. In view of underlying malignancy, working diagnosis at this point of time was metastasis. Subsequently, a bone scan with SPECT/CT was performed to assess the extent of involvement and to rule out other sites of bone metastasis. Three hours after intravenous administration of 19.4mCi of Tc99m Methylene Diphosphonate, a whole body planar bone scan was performed followed by SPECT/CT. The planar bone scan showed solitary intense focus of increased

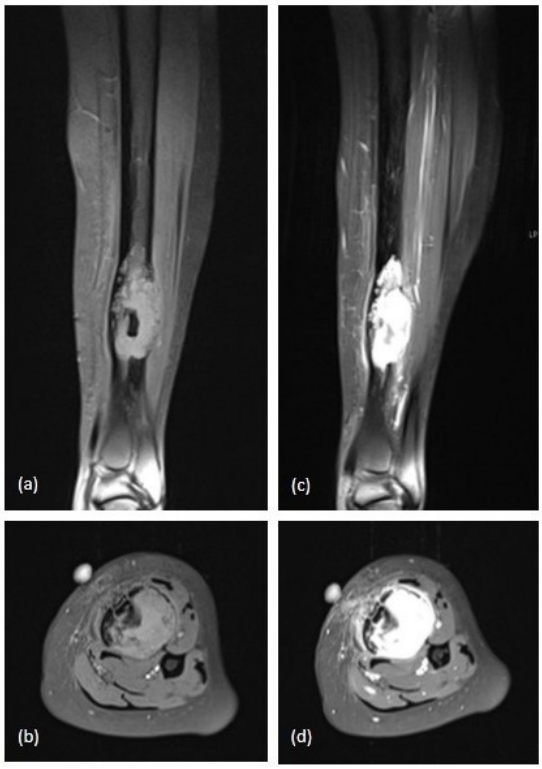

Figure 1: MRI (a) Coronal, (b) axial T1-weighted images showing the lesion at the diaphysis of left distal tibia. MRI (c) Coronal, (d) axial T1-weighted images after gadolinium administration showing intense enhancement of the lesion.

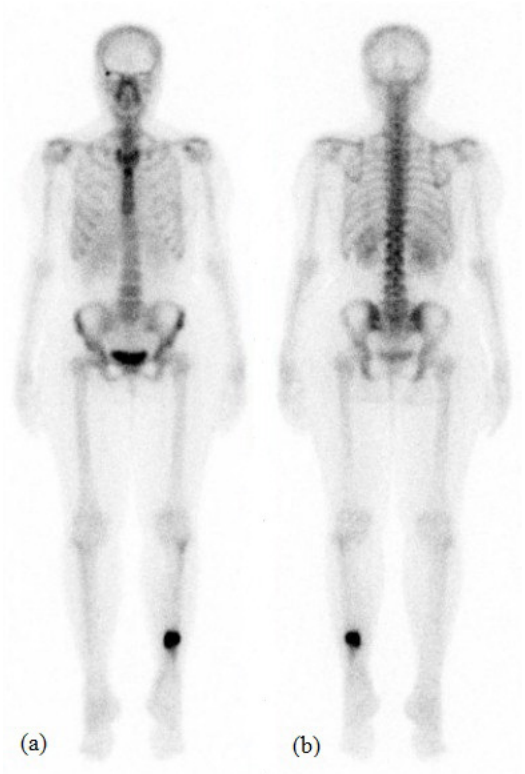

Figure 2: (a) Anterior and (b) Posterior images of planar bone scintigraphy showing solitary intense focus of increased tracer accumulation at the diaphysis of left distal tibia.

tracer accumulation at the diaphysis of left distal tibia (Fig. 2) which correspond to a lytic lesion with cortical destruction, periosteal reaction and pathological fracture on SPECT/CT images (Fig. 3). The scintigraphic features were suggestive of primary bone tumour or focal osteomyelitis rather than metastasis. Due to the diagnostic dilemma, a trucut biopsy of the left tibia was done and microscopically showed fragments of irregular, curvilinear trabeculae of woven bone rimmed by osteoblasts. The fibrous component consists of bland spindle cells and a matrix that varies from a myxoid component to moderately fibrous component. Scattered small groups of cytologically bland epithelial cells were highlighted by CK AE1/AE3 immunohistochemistry, thus consistent with OFD-like adamantinoma (Fig. 4). Following this diagnosis, she underwent excision of the OFD-like adamantinoma. A fibula bone graft with screw and plate fixation was done. Patient recovered well after the procedure.

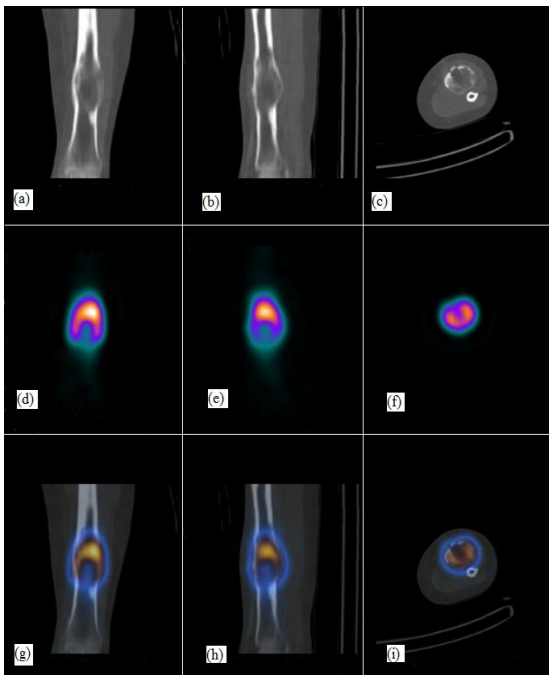

Figure 3: (a) CT, (b) SPECT and (c) fused SPECT-CT images showing a focus of increased tracer accumulation at the diaphysis of left distal tibia which correspond to a lytic lesion with cortical destruction, periosteal reaction and pathological fracture.

DISCUSSION

In a patient with underlying malignancy who presents with bone pain, bone metastasis remains as the top differential diagnosis. This is particularly true in malignancies such as prostate cancer, breast cancer and lung cancer, which commonly metastasize to the bone. In patients with colorectal cancer, bone metastasis is less common, especially in the absence of other organ involvement such the liver and lung. However, review of literature shows that bone metastasis in colorectal cancer is actually more common than was initially thought (1). Bone metastasis in colorectal cancer usually involves the pelvic region and spine due to presence

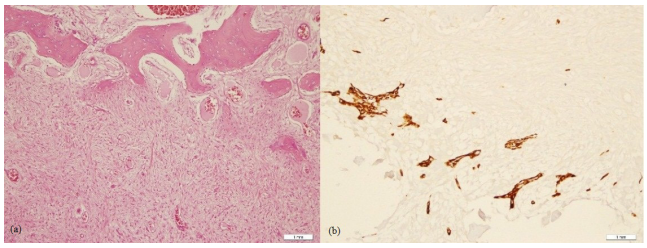

Figure 4: (a) H & E stain showing osteofibrous component which is made up of storiform-oriented, bland spindle cells intermingled with areas of irregular, curvilinear woven bony trabeculae rimmed by osteoblasts (x10). (b) Immunohistochemical stain (CK AE1/AE3) showing the epithelial islands and scattered individual spindle cells.

of paravertebral venous plexus of Batson. Involvement of appendicular skeleton is rare, in particular solitary involvement. There are a few published case reports of patients with rectosigmoid cancer who presented with solitary tibial lesion and histopathology proved them to be metastatic lesions (2,3).

Our patient presented with solitary tibial bone involvement and the initial working diagnosis was bone metastasis despite its rarity. Due to the diagnostic dilemma based on the clinical and imaging features, our patient was also subjected to a biopsy, but in contrast to the other cases mentioned above, the histopathology showed OFD-like adamantinoma.

Adamantinoma is a rare tumour, accounting for less than 1% of all primary bone tumours and has a strong preponderance for tibia. There are two distinct types of this disease entity which is the classical type and differentiated type (OFD like-adamantinoma). Both types not only share similar imaging features with each other but also with osteofibrous dysplasia, which makes histological assessment mandatory as there is significant variation in the clinical course of these three disease entities. Clinically, osteofibrous dysplasia is a non-malignant fibro-osseous lesion, whereas classic adamantinoma is an aggressive malignant tumour with metastatic potential. On the other hand, OFD-like adamantinoma has a relatively indolent course (4,5).
Histologically, the differentiation between classic adamantinoma and OFD-like adamantinoma depends on the proportion of epithelial and osteofibrous components that are present. OFD-like adamantinoma has predominance of osteofibrous tissues with small groups of epithelial cells which are usually detected by immunohistochemical analysis. Even though there are wide array of speculations regarding the origin of adamantinomas, all the histologic subtypes of adamantinomas show strong positive staining with pan-cytokeratin antibody which points towards epithelial origin of these tumours (5).

Plain radiography is usually the first imaging modality of choice with the classical feature of a predominantly osteolytic, intracortical lesion at the diaphysis of anterior tibia. MRI is done for further delineation of the lesions and typically shows low-intensity on T1-weighted images and high-signal intensity on T2-weighted images. On the other hand, bone scintigraphy is a sensitive imaging modality that is frequently used to detect bone metastasis but there is limitation to its specificity as many other benign conditions can also have similar increased uptake (hot spot) as metastatic lesions. The advent of SPECT-CT has enabled more accurate characterization of the lesions detected on bone scintigraphy, as it correlates anatomical information from CT with functional information from SPECT. In our patient, the focal increased uptake on bone scintigraphy was suspicious for metastasis. However, upon performing SPECT-CT, our differential diagnosis was narrowed down to either primary bone tumour or focal osteomyelitis based on the combination of morphoscintigraphic features.

CONCLUSION

In a patient with underlying malignancy, the detection of a solitary bone lesion should prompt further investigation. Imaging such as MRI and bone scan with SPECT-CT can help to further characterize the lesion. The latter will also provide whole body assessment and exclude other occult metastases. However, in instances where the diagnosis remains uncertain due to non-specific features, pathological confirmation is mandatory to avoid futile treatment.

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