The Foot 27 (2016) 10–15

Contents lists available at ScienceDirect

The Foot

journal homepage: www.elsevier.com/locate/foot

Case report

Metatarsal leiomyosarcoma masquerading as acute osteomyelitis – A diagnostic trap unveiled by vigilant clinical, radiologic and pathologic analysis

Louis Tsun Cheung Chow *

Department of Anatomical and Cellular Pathology, Prince of Wales Hospital, Hong Kong

HIGHLIGHTS

- The present article represents the first reported case of metatarsal leiomyosarcoma.
- The present and the only other reported case of foot leiomyosarcoma show immunohistochemical staining pattern consistent with vascular smooth muscle derivation.
- Application of h-caldesmon and calponin staining in bone biopsy specimen is helpful in the differentiation of smooth muscle tumor from acute or chronic osteomyelitis with reactive fibroblastic proliferation.

ARTICLE INFO

Article history:
Received 28 April 2015
Received in revised form 31 August 2015
Accepted 4 September 2015

Keywords:
Metatarsal leiomyosarcoma
Acute osteomyelitis
Bone
Leiomyosarcoma
Foot

ABSTRACT

Due to overlapping clinical and radiological features, the differentiation between osteomyelitis and bone tumor can be challenging. A 48-year-old lady presented with intermittent left foot pain for a few months. Plain radiographs showed an osteolytic lesion affecting the proximal diaphysis of the left fourth metatarsal bone, with thinning and irregularities of the cortex and focal periosteal reaction. Ultrasonography revealed diffuse subcutaneous edema in the dorsum of the left foot, cortical irregularities along the mid-shaft of the left fourth metatarsal bone, and surrounding periosteal collection. Computed tomography showed medullary expansion along the shaft and base of the left fourth metatarsal bone with cortical irregularities and defects suggestive of cloaca, and focal mild periosteal new bone formation. The clinico-radiologic diagnosis was acute osteomyelitis with periosteal collection. During open biopsy, the finding of intramedullary fleshy tissue in the absence of significant inflammatory edema and purulent discharge, and subsequent negative culture result prompted a review of the histologic slides which was initially reported as benign fibroblastic tissue proliferation. Careful analysis of the histomorphology disclosed a spindle cell sarcoma for which ray amputation of the fourth and fifth metatarsal was performed. The final diagnosis was grade 1 leiomyosarcoma and the patient remained well 33 months after the operation.

1. Introduction

Osteomyelitis defined as inflammation of bone and marrow leading to bone destruction is caused by infectious microorganisms in the vast majority of cases [1]. A pathophysiologic classification of value in clinical application segregates osteomyelitis into three subtypes [2]. Those resulting from hematogenous seeding are seen typically in pre-pubertal children and elderly patients. Osteomyelitis may occur from direct inoculation of microorganisms into intact bone following trauma, bone surgery or joint replacement and thus can involve any bone and person of any age groups. Lastly, vascular insufficiency mostly a result of diabetic vasculopathy predisposes the spread of soft-tissue infection in the foot to adjacent bones.

The great similarity with significant degree of overlap in clinical and radiologic features renders the differentiation of hematogenous osteomyelitis from bone tumors difficult [3–6]. In this article, we report a lady with primary leiomyosarcoma of the left fourth metatarsal bone misdiagnosed clinically and radiologically as acute osteomyelitis. The subtle clinical, radiologic and histologic features

* Corresponding author at: Department of Anatomical and Cellular Pathology, Prince of Wales Hospital, 30–32 Ngan Shing Street, Shatin, Hong Kong.
Tel.: +852 26322335; fax: +852 26374858.
E-mail address: louistcchow@gmail.com

http://dx.doi.org/10.1016/j.foot.2015.09.003
0958-2592/

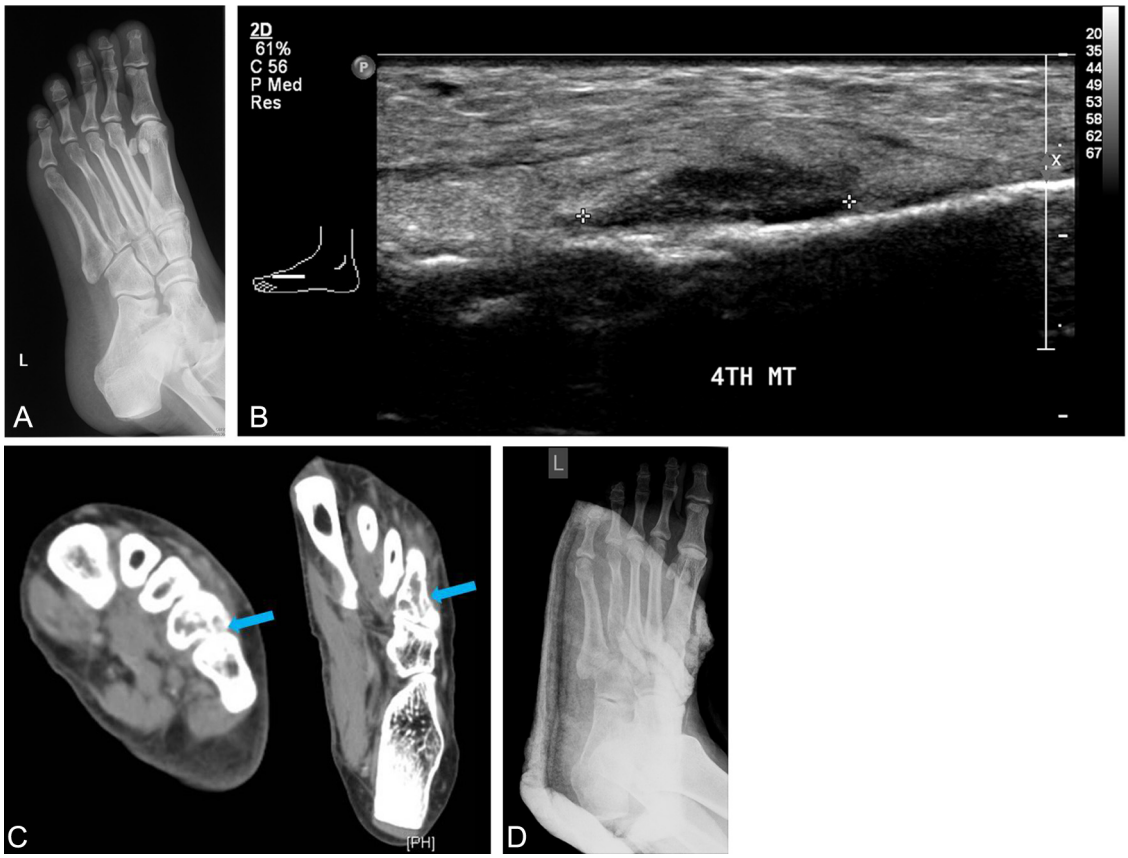

Fig. 1. (A) Plain radiograph of the left foot showing an expansile osteolytic lesion in the diaphysis of the fourth metatarsal bone. There is periosteal reaction in the proximal diaphyseal region. (B) Ultrasonography showing diffuse subcutaneous edema in the dorsum of left foot. There are cortical irregularities along the mid-shaft of left fourth metatarsal bone, associated with surrounding periosteal collection. (C) Coronal (left) and transverse (right) CT images showing medullary expansion along the shaft and base of the left fourth metatarsal bone with cortical irregularities (right arrow) and defects suggestive of cloaca (left arrow). (D) Plain radiograph of the left foot showing minimally displaced fracture in the anterior aspect of the osteolytic lesion in the diaphysis of the fourth metatarsal.

allowing the avoidance of such fallacy are discussed. As primary leiomyosarcoma of foot bones is extremely rare, the relevant literature is reviewed.

2. Case report

2.1. Clinical presentation

A 48-year-old sales lady whose job required substantial standing and manual work, enjoying good past health with no history of diabetes, presented to a peripheral hospital because of intermittent left foot pain for a few months. Physical examination revealed a slightly fluctuant swelling with mild redness, hotness and tenderness in the dorsum of the left foot over the third and fourth metatarsal region. There was no fever, no limitation of movement of the toes and ankle. The white cell count and C-reactive protein (CRP) were normal while the erythrocyte sedimentation rate (ESR) was slightly elevated. Plain X-rays showed an osteolytic lesion affecting the proximal diaphysis of the left fourth metatarsal bone, with thinning and irregularities of the cortex and focal periosteal reaction (Fig. 1A). Ultrasonography was performed under the clinical suspicion of acute osteomyelitis with abscess formation, showing diffuse subcutaneous edema in the dorsum of left foot. There were cortical irregularities along the mid-shaft of the left fourth metatarsal bone, associated with surrounding periosteal collection (Fig. 1B). Computed tomography (CT) showed medullary expansion along the shaft and base of the left fourth metatarsal bone with cortical irregularities and defects suggestive of cloaca. There were focal mild periosteal new bone formation and surrounding soft tissue swelling around the mid-shaft (Fig. 1C). The opinion of the radiologist was acute osteomyelitis of the left metatarsal shaft with periosteal collection and cellulitis along the dorsum of the left foot.

After performing Mantoux tuberculin skin test which was negative, the medical microbiologist suggested bone biopsy for histologic and further microbiologic analysis. Intra-operative radiograph during open biopsy showed minimally displaced fracture over distal shaft of the left fourth metatarsal bone. There was surrounding soft tissue swelling but the dorsal cortex of the fourth metatarsal bone was normal. However, fleshy tissue was seen in the medulla through the cortical window opened at the base. There was no purulent discharge. The medullary fleshy tissue was taken for histologic and microbiologic studies including smear and culture for *Mycobacterium tuberculosis* and fungus. A short leg slab was applied after wound irrigation, closure and dressing. The patient made an uneventful recovery and in retrospect offered a history of increase in left foot pain during home leave before biopsy, consistent with the operative finding of pathological fracture (Fig. 1D).

The histopathology was reported as benign fibroblastic tissue proliferation but in view of the fleshy medullary tissue, absence of purulent discharge and negative bacterial culture, the orthopedic surgeon was sceptical about the benign diagnosis and the histologic slides were sent to our hospital for evaluation.

2.2. Pathologic features of the biopsy

Review of the slides showed that the lesion consisted of interlacing fascicles of closely packed spindle cells supported in a dense