Case Report

Hydatid disease: A rare cause of fracture nonunion

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Abstract

Hydatid disease is an infrequent parasitic infestation caused by cestode, most commonly, *Echinococcus granulosus*. Bone involvement is distinctly uncommon. We would like to share our experience of a rare case of hydatid disease of femur in a 24-year-old male who presented with nonunion of subtrochanteric fracture. Histopathology showed typical lamellated wall and dagger-shaped hooklets. In view of its rarity, hydatid disease often remains an unsuspected infection of the bone.

Keywords: Bone, fracture nonunion, hydatid disease

Introduction

Hydatid disease is an infrequent parasitic infestation caused by cestode, most commonly, *Echinococcus granulosus*. Liver and lung are most commonly affected, 28.9%–65% and 25%–48.1% cases, respectively.[1,2] Bone involvement is distinctly uncommon; 0.6%–5.6% cases of hydatid disease have osseous manifestations.[3,4] We would like to share our experience of a case of nonunion of subtrochanteric fracture of the right femur associated with hydatid disease of bone.

Case Report

A 24-year-old office worker male presented to outpatient orthopedics department of our hospital with nonresolution of his symptoms despite a previous surgical intervention. His complaints dated 6 months back when he sustained trivial trauma following which he was unable to bear weight on his right lower extremity. There was no history of fever or constitutional symptoms. An X-ray right hip and pelvis were performed which revealed a subtrochanteric fracture of the right femur with displaced fracture fragments [Figure 1]. There was no evidence of osteoporosis, lytic/blastic lesion suggestive of bone tumor. Subsequently, he underwent open reduction with intramedullary nail fixation for fracture stabilization.

After 6 months postsurgery, he presented to our outpatient with complaints of persistent local pain and swelling. He was unable to bear weight on the right lower limb. Fresh radiographs revealed lysis around the implant with resorption of the fracture ends [Figure 2]. Notably, there was no evidence of callus formation. Computed tomography scan or magnetic resonance imaging could not be performed due to financial constraints. Suspecting persistent low-grade chronic pyogenic infection, he was planned for surgical debridement with external fixation. Intraoperatively, the muscles and soft tissues were found to be indurated. Pearly white structures and granulation tissue were identified at lesional site. A through betadine wash was performed, excised tissue sent for histopathology, and external fixator applied.

The excised specimen consisted of multiple fragments including pearly white-flattened structure measuring 2.8 cm × 2 cm × 1 cm. Microscopy revealed fragments of thick eosinophilic acellular lamellated membrane corresponding to parasite exocyst [Figure 3]. Degenerating bits of thin endocyst/inner nucleated germinal layer were seen occasionally. High magnification showed characteristic refractile dagger-shaped hooklets [Figure 4]. Hence, diagnosis of hydatid

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Access this article online

Quick Response Code:

Website: www.jfmpc.com

DOI: 10.4103/2249-4863.214979

How to cite this article: Aggarwal D, Girotra V, Wadhwa N, Chadha M. Hydatid disease: A rare cause of fracture nonunion. J Family Med Prim Care 2017;6:164-6.

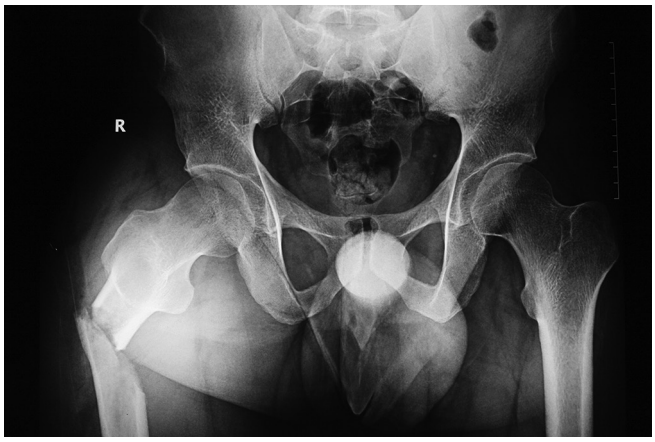

Figure 1: Preoperative radiograph showing subtrochanteric fracture of right femur. Note absence of any obvious clue suggestive of primary bone pathology

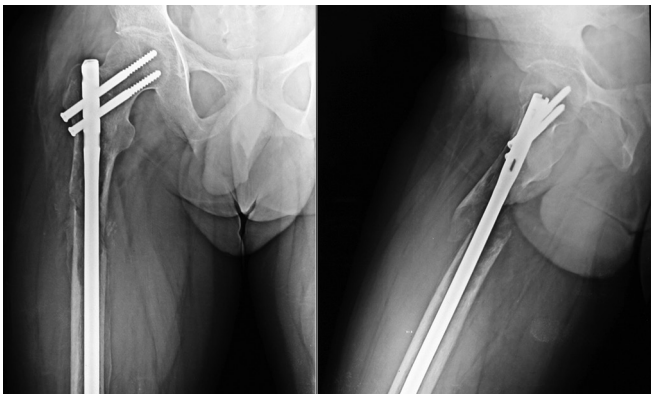

Figure 2: Postoperative films showing *in situ* intramedullary nail. There was lysis around the implant, resorption of the fracture ends, and no evidence of callus formation

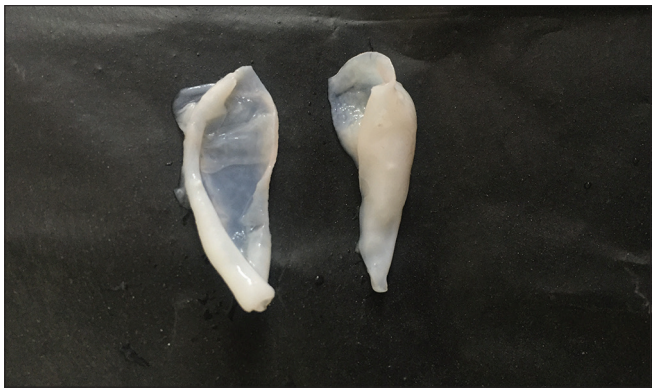

Figure 3: Part of specimen showing pearly white cyst wal

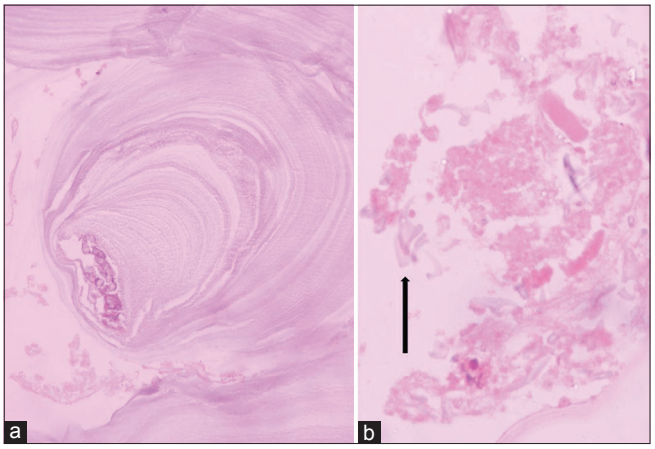

Figure 4: (a) Thick eosinophilic lamellated membrane/parasite exocyst (H and E, ×200). (b) Dagger-shaped refractile hooklets (arrow) of *Echinococcus* and tiny fragments of germinal layer (H and E, ×400)

disease of the right femur was rendered. Ensuing ultrasound abdomen and chest did not reveal liver or lung involvement. The patient was started on 10 mg/kg albendazole into two divided doses with fat rich meals, to be continued for 2 years. Since the diagnosis was established postoperatively, wait and watch policy was decided; further surgical intervention to be done only if indicated clinically.

Discussion

Hydatid disease is an uncommon zoonotic parasitic infestation. It is caused by larval form of cestode *Echinococcus*, most commonly *E. granulosus* and *Echinococcus multilocularis*, and occasionally *Echinococcus vogeli*. Humans are accidental intermediate, dead-end hosts. Infection is often acquired by the consumption of unwashed vegetables contaminated with cestode eggs. The oncospheres enter portal circulation by penetrating intestinal wall.[1,4]

By virtue of their filtration function, liver and lungs trap most larvae and hence account of majority of disease burden (77%–90%).[1,2] Hydatid disease outside these sites suggests larval escape. Although theoretically any site may be involved, bone disease is very rare. Even in endemic areas, osseous disease accounts for 0.6%–5.6% cases.[3,4] Thoracic spine is the most common site followed by lumbar and sacral. Other sites include ilium, femur, humerus, tibia, ribs, and skull bones. In our case, it is difficult to comment if the disease was missed in first surgical intervention or happened subsequently. Irrespective of timing, it interfered with bone healing, i.e., callus formation.

Hydatid disease of the bone behaves unlike other site infestation.[3,5,6] Because of its hard structure and rigidity, bone, especially the cortex, offers resistance to cyst expansion. The disease remains confined to the spongy medulla and frequently fails to achieve large cystic dimension typical of loose tissues such as lung and liver. In view of this, some authors prefer to label bone disease as hydatidosis. The gradual but unrelenting pressure compromises vascular supply and results in ischemic events. Compression fractures occur in spine and weight-bearing long bones. Grossly identifiable cysts, like in our case, are uncommon.[6-8] Such cases often have extraosseous soft tissue component permitting cyst expansion.

Clinical presentation of bone hydatid disease is nonspecific. Bracanovic *et al.* found pathologic fracture to be the most common presentation (48.8%), followed by pain (41.5%), and

paraplegia (22%). The pathological fracture was most common in spine (75%), followed by femur (20%), and tibia (5%).[3] Sinus formation and pus discharge simulating osteomyelitis too have been described. These signs and symptoms may be seen in other bone lesions such as infections, tumor metastasis or rarely in the primary bone tumors.

Radiologic findings associated with hydatid disease of the bone too are varied. Expansile lytic lesions and cortical thinning are common.[8] Reactive bone formation is usually minimal. Changes secondary to bone destruction, for example, ischemic rarefication, sequestrum formation, honeycombing, and pathological fracture often underlie clinical presentation. Extension into soft tissues may be evident as soft tissue calcifications. The differential diagnoses of this uncommon lesion include more common conditions such as giant cell tumor, solitary bone cyst, aneurysmal bone cyst, fibrous dysplasia, bone metastases, and intraosseous ganglion.[9] Death of parasite invokes inflammation and such cases are likely to be mistaken as osteomyelitis – pyogenic or tuberculous. Diagnosis of hydatid disease of the bone is seldom made preoperatively. In contrast to hepatic and pulmonary disease, serologic tests have limited value in bone hydatosis. Loudiye *et al.* found only 50% of their cases to have a positive result.[10] Histology remains the mainstay of diagnosis.

In view of its rarity, there are no standardized guidelines for management of hydatid disease of bone. Surgery remains the mainstay despite high recurrence rates. Bone prosthesis cementing may be required in cases with extensive bone destruction. Long-term (up-to 2 years) medical cover with albendazole is recommended for the prevention of recurrence and systemic cover. The clinical outcome remains unpredictable. Hence, long-term follow-up is suggested.

To conclude, we have shared our experience of an unusual association of hydatid disease of bone with fracture nonunion. Such cases are difficult to diagnose clinico-radiologically. Histopathology remains cornerstone for diagnosis of hydatid disease of bone.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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