Primary Iliac Bone Hydatid Disease : An Unusual Presentation

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Introduction

Human hydatid disease is caused by *Echinococcus granulosus* [1]. Primary bone involvement is unusual, but typically occurs at more vascularized sites, such as the vertebrae, long bone epiphyses and ilium. The larval form reaches the bone, penetrates the spongy tissue and grows in the direction of least resistance, infiltrating and damaging the tissue like a tumour [2]. The prognosis is poor when bone is involved, even in patients who undergo extensive medical and surgical treatment [3].

We present an unusual case of recurrent hydatid disease that masqueraded as aneurysmal bone cyst and presented as a draining sinus in the left flank. The biopsy done from the sinus tract revealed hydatid cyst.

Case Report

A 35 year old male, presented with pain in left hip joint along with difficulty in walking of two months duration. Physical examination of the left hip joint revealed limited internal rotation without any pain. On plain radiography of the pelvis, osteolytic lesion was present in the left iliac bone. Curettage and bone grafting (autologous+bovine) was done in 2006. Histopathology then showed collection of giant cells around vascular spaces suggesting a diagnosis of aneurysmal bone cyst.

Patient presented again with left flank swelling and a draining sinus in Oct 2007. Radiograph of pelvis showed recurrent multiple erosive lesions in left ilium (Fig. 1) and computed tomography (CT) scan pelvis (Fig. 2) showed expansile lytic lesions with cortical breach at multiple sites involving left iliac bone, acetabular rim, anterior and posterior pillars, ischial tuberosity and inferior left pubic rami along with multilobulated intrapelvic collection displacing and distorting pelvic structures. Patient was diagnosed as a case of recurrent iliac bone cyst with an overlying abscess with

sinus discharge. Trucut biopsy was however inconclusive. Excision biopsy of sinus tract and iliac cyst was done. Biopsy showed hydatid cysts (laminated membrane and brood capsules with scolices) (Fig. 3) and florid foreign body giant cell reaction (Fig. 4) with small areas of infarcted bone and normal trabecular bone around. Patient was subsequently taken up for wide excision.

Discussion

Man is an accidental host in the life cycle of *Echinococcus granulosus*. Human infestation occurs when the ova are swallowed. Cysts are found in the liver (55–60%), lungs (30%), kidneys (2.5%), heart (2.5%), bones (2%, mostly in spinal column), muscles (1%), brain (0.5%) and in other organs (1.5%). The lesions in bone may mimic a tumour.

In man, infection is usually acquired in childhood. The symptoms present several years after exposure and it may take five to 20 years before a diagnosis is made. Of 532 cases of echinococcosis reviewed in Lebanon, 12 were of pelvic hydatid disease [4]. Primary pelvic hydatid cyst may present with abdominal symptoms related to compression of the rectum and urinary tract [5]. In some cases, primary pelvic hydatid cyst causing sciatica and foot drop have also been reported [6]. Our patient, with a draining sinus in his flank region, was unique and such a presentation is rare in literature. It is of utmost importance that a correct 'pre-operative diagnosis' is made, since all precautions must be taken to prevent dissemination and seeding of the surgical field. Deaths have been reported due to anaphylactic shock resulting from spillage during excision or biopsy after a mistaken diagnosis of a retroperitoneal tumour [7]. So in endemic regions, because of the diversity of its presentation, hydatid disease should always be in the differential diagnosis list of any growing destructive

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Fig. 1 : Multiple osteolytic lesions left iliac bone and femoral acetabulum

Fig. 2 : CT scan showing osteolytic lesions

mass. Diagnostic techniques such as radiography, ultrasonography, computed tomography, magnetic resonance imaging and immunologic tests are of value [8]. Complete surgical excision is the treatment of choice for osseous hydatid disease. Isolated medical therapy with mebendazole or albendazole is not adequate for controlling the process, but can be added to surgery when complete excision of the cyst is not possible or doubtful [9].

Conflicts of Interest

None identified

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Fig. 3: Laminated membrane and free brood capsules

Fig. 4 : Foreign body giant cell reaction around laminated membrane

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