Pelvic Hydatid Disease: A Case Report and Review of Literature

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Learning Points for this Article:
Pelvic hydatidosis is a rare but important differential diagnosis of tubercular hip arthritis, especially in areas with high prevalence of hydatid disease.

Abstract

Introduction: Cystic echinococcosis of the bone is rare and difficult to treat due to frequent recurrences, especially in certain locations such as ilium and hip, where radical surgery is difficult to achieve.

Case Report: We present a case of a 35-year-old female of the Indian subcontinent who had complaints of pain in left hip and limp since 1 year. The first clinical and radiological diagnosis was tuberculosis of the hip. However, higher imaging modalities revealed the diagnosis of hydatid disease of hip. The patient underwent surgical debridement and anti-helminthic chemotherapy. At 3-year follow-up, the patient was disease-free.

Conclusion: Hydatid disease of the hip and pelvis, although rare must be kept in the differential diagnosis of pathologies of hip-like tuberculosis. Debridement and excision of hip joint give good functional outcome, while also minimizing morbidities that are usually associated the use of custom-made prosthesis or complex arthroplasty.

Keywords: Hydatid disease, pelvis, Echinococcus.

Introduction

Hydatid cysts commonly affect the liver and the lung [1]. Cystic echinococcosis of the bone is rare with an incidence of 0.5-4% among patients with hydatid disease [2]. Bone hydatidosis is difficult to treat and carries high morbidity due to frequent recurrences, especially in certain locations such as ilium and hip, where radical surgery is difficult. We report a case of hydatid disease of hip and pelvis treated by debridement and Girdlestone arthroplasty (excision of the hip joint).

Case Report

A 35-year-old female of the Indian subcontinent, from a rural background, was referred to our center complaining of pain in left hip and limp since 1 year. The pain was insidious in onset and was associated with difficulty in squatting and sitting cross-legged. The patient had anorexia and undocumented weight loss. There was no other significant past medical history. Family history revealed that her husband had been previously treated for pulmonary tuberculosis 10 years back.

The hip was tender on palpation and associated with fullness in the iliac fossa. The hip was flexion deformity of 30°, with only jog of movement at the hip joint, and true shortening of 3 cm.

Plain radiographs (Fig. 1) showed obliteration of joint space with the destruction of femoral head and acetabulum. Osteolytic lesions were seen in the supra-acetabular region and superior pubic ramus. Laboratory investigations revealed an elevated erythrocyte sedimentation rate (ESR) of 52 mm/h and eosinophilia, with absolute eosinophil count (AEC) of 930/mL (30-350/mL).

Based on this, a provisional diagnosis of tuberculosis of the hip was made. Baseline liver function tests (LFT) before starting anti-tubercular therapy (ATT) revealed mild hyperbilirubinemia (T. Bil -2.1 mg/dL) and elevated...
Cystic echinococcosis is a persistent zoonosis in rural livestock raising areas. Countries in the temperate zone are endemic areas. The definitive hosts are dogs, foxes, and other carnivores. The tapeworms live in the small bowel and infected ova are shed in feces. When ingested by intermediate hosts such as man, sheep, or cattle, the larvae enter the portal circulation. Sometimes, larvae reach the lungs and other areas of the body. The life cycle is completed when the definitive hosts consume infested viscera of the intermediate host.

Echinococcus granulosus. The larval stage of E. granulosus is the most common cause of hydatid disease in man. The definitive hosts are dogs, foxes, and other carnivores. The tapeworms live in the small bowel and infected ova are shed in feces. When ingested by intermediate hosts such as man, sheep, or cattle, the larvae enter the portal circulation. Sometimes, larvae reach the lungs and other areas of the body. The life cycle is completed when the definitive hosts consume infested viscera of the intermediate host.

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**Figure 4:** (a) Cystic swelling on the anterolateral aspect of the involved hip and proximal thigh, (b) combined anterior inguinal approach and lateral approach used to expose the hemipelvis, (c) injection of 10% povidone-iodine solution before dissection and excision of cyst, (d) hydatid sand sent for biopsy.

**Figure 5:** Patient sitting cross-legged and squatting without difficulty at follow-up.

(China, Central Asia, Mediterranean region, Australia, and parts of South America) have the greatest prevalence of cystic echinococcosis [4]. Andhra Pradesh and Tamil Nadu have the highest prevalence of hydatid disease in India [5].

Age of presentation is usually around 40 years [6, 7]. Tekin reported a female predominance [8]. However, Palanivelu reported a male predilection of the disease (5:1) [7].

The liver and lungs are the most common sites (~90%) of cysts in humans. The kidneys, spleen, and muscles are occasional sites (2-3% each). The heart, brain, and bones are extremely rare sites (<1% each) for cystic echinococcosis [9]. Vertebra accounts for the majority (75%) of osseous hydatidosis. Pelvis accounts for 16-28% of osseous hydatidosis [10].

**Table 1: Summary of hip and pelvic hydatid disease**

| Author year | Age/sex | Lesions | Initial diagnosis | Management | Outcome |
|-------------|---------|---------|------------------|------------|---------|
| Wirbel et al. [19] 1995 | 49/- | Acetabulum, proximal femur, pubis and ischium | Traumatic central fracture dislocation | 5-year chemotherapy followed by partial pelvic resection and prosthesis replacement | Recurrence that required revision surgery with custom-made prosthesis |
| Belzunegui et al. 1997 [15] | 54/f | Hemipelvis and proximal femur | Giraldstone arthroplasty followed by 10-year chemotherapy | Recurrence managed on chemotherapy |
| Natarajan et al. 2002 [20] | 28-55 years 2 females 1 male | Pathological fracture of femur | Wide excision, customized mega prosthesis and chemotherapy | No recurrence and good function |
| Siwach et al. 2009 [17] | 51/f | Femur (pathological fracture) and pelvis | Hindquarter amputation and chemotherapy | Death due to sepsis |
| Rath et al. 2009 [20] | 35/M | Iliac bone, acetabulum, trochanteric, and pubic rami | Aneurysmal bone cyst, recurrence was also labeled as ABC | Wide excision | - |
| Nekapelka et al. 2010 [21] | 35/f | Hip joint and ileum | Propionibacterium infection | 2 years of medical therapy followed by cemented THR | Recurrence that required customized hemipelvic replacement |
| Notarnicola et al. 2010 [24] | 53/f | Proximal femur (pathological fracture) | Idiopathic coxalgia | Total hip replacement | Recurrence dislocation and disassembly for which revision done with Wagner type prosthesis |

Osseous hydatidosis may lie dormant for decades. It is usually detected only after a complication such as a pathologic fracture, neural deficit, and infection or fistulation of the abscess [10].

Physical findings are usually unremarkable except in a few cases. There can be a deformation of limbs, a cold or fistulized abscess, or a vertebral kyphosis. Since the disease has nonspecific features, one must maintain a high index of suspicion, especially in shepherders, veterinarians, or butchers.

The most common radiographic feature of osseous lesions is multiple expanding lucent lesions without clear boundaries, together forming a huge area of osteolysis with the classic waffle appearance [11]. This is accompanied by cortical thinning and a lack of periosteal reaction unless there is a superadded infection.

Abdominal pelvic ultrasonography and radiography of the chest is required to look for a visceral hydatidosis. CT scan is the best method for diagnosis and follow-up of osseous hydatidosis. Magnetic resonance imaging is often used for defining the local extent of the lesions in the soft tissues [12]. The hydatid vesicles are seen as a hypodense signal in T1-weighted images and a hypersignal in T2-weighted images [13].

The differential diagnosis includes tuberculosis, chronic osteomyelitis, and tumors such as aneurysmal cysts, osteoclastoma, chondrosarcoma, osteosarcoma, and metastases [14].

Pelvic hydatid disease is dangerous due to the clinical latency of the disease, which allows parasitic invasion of the sacral, iliac, and hip joint, thus making eradication of parasite almost impossible. However, wide excision or marginal excision is preferred to decrease the parasitic load. Belzunegui et al. have shown good functional outcome after Girdlestone arthroplasty [15]. Aggarwal reported satisfactory outcome with the combination of chemotherapy and surgery [16]. Siwach et al. reported a case of extensive hydatid disease of pelvis and femur for which a hindquarter amputation was performed [17]. Prothesis is fraught with danger because of the risk of bacterial infection [18]. Wirbel reported a case where replacement was done with unsatisfactory results (Table 1) [19].

**Conclusion**

Hydatid disease of the hip and pelvis, although rare must be kept in the differential diagnosis of pathologies of hip-like tuberculosis. Raised eosinophil count should prompt further investigation into the cause of eosinophilia, especially if associated with deranged LFTs. Debridement and excisional arthroplasty give good functional outcome, while also minimizing morbidities that are usually associated with the use of a custom-made prosthesis or complex arthroplasty.

**Figure 6: (a) Cystic swelling on the anterolateral aspect of the involved hip and proximal thigh, (b) combined anterior inguinal approach and lateral approach used to expose the hemipelvis, (c) injection of 10% povidone-iodine solution before dissection and excision of cyst, (d) hydatid sand sent for biopsy.**
Clinical Message

In endemic regions, hydatid disease of the bone must be considered in the differential diagnosis of atypical aggressive infections. Debridement and excision along with appropriate medical anti-helminthic treatment are necessary to treat the disease.

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