

# INTRAOSSIOUS HYDATID CYST OF THE SACRUM AND PELVIS BONES: AN UNCOMMON OCCURRENCE

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## ABSTRACT

Skeletal hydatidosis results from the deposition of the larval form of the Echinococcus, a genus of tapeworm. The incidence of bone disease is extremely low as most larvae are trapped by the liver and lung upon release of the embryo into the portal bloodstream. Bone hydatid disease is often asymptomatic and its diagnosis is usually made at an advanced stage when lesions have become extensive. The interpretation of imaging studies can prove very confusing as there are no characteristic features and this often leads to misdiagnosis. We present a case of an 83-year-old man who was admitted to BPKIHS, Dharan complaining of pain and swelling in the left hip.

**Keywords:** *Echinococcus; Cestoda; Diagnostic Errors*

## INTRODUCTION

A parasitic tapeworm, Echinococcus, causes hydatid disease. The definitive host is usually a dog, and sheep are the most common intermediate hosts. Humans may become intermediate hosts through contact with a definitive host or through ingestion of contaminated water or vegetables.<sup>1,2,3</sup> In humans, hydatid disease involves the liver in approximately 75% of cases, the lung in 15%, and other anatomic locations in 10% of the cases.<sup>1,2,4</sup> Osseous hydatid disease is an infrequent entity that represents 0.5–2.5% of all hydatidosis. The vertebrae are the most commonly affected bones (50%), followed by the pelvis (25%) and the long bones (15–25%).<sup>5</sup>

In osseous hydatid, pericyst formation does not occur, thereby allowing aggressive proliferation in an irregular branching fashion, along the line of least resistance, especially the bone canals.<sup>6,7</sup> The parasite replaces the osseous tissue between trabeculae due to the slow growth of multiple vesicles. With time, the parasite reaches and destroys the cortex, with subsequent spread of the disease to the surrounding tissues.

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## CASE REPORT

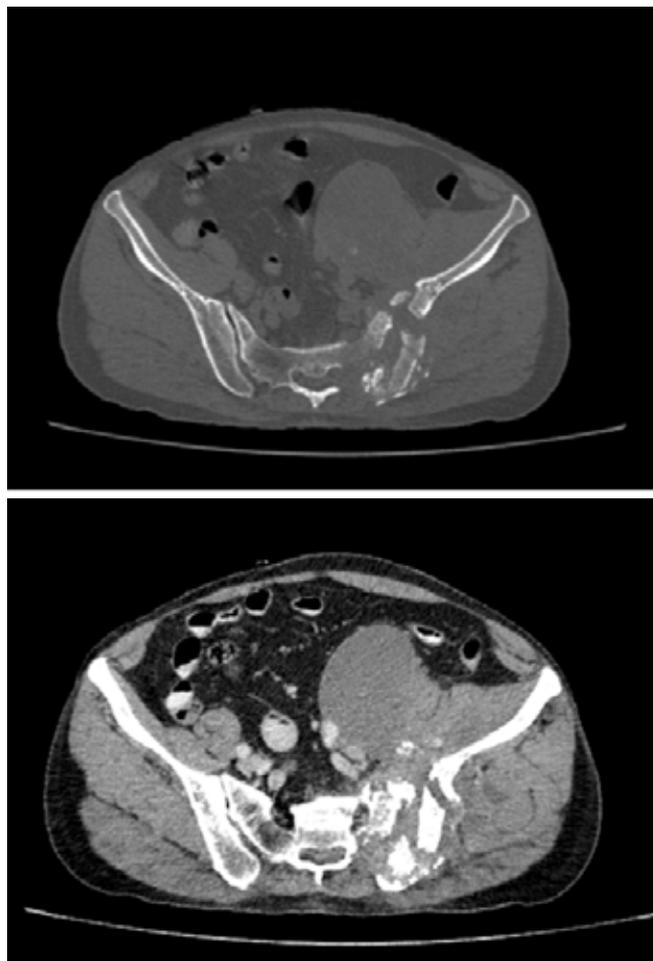
An 83-year-old male presented to the OPD with a history of pain and swelling in the left gluteal and hip regions for 3 months. The pain was insidious in onset and progressive in nature and increased on walking. He had no abdominal pain or any abdominal discomfort, no chest pain or, cough. There was no history of tobacco smoking or any substance or alcohol abuse. The patient had had a retropubic prostatectomy done 7 years back.

At presentation, his blood pressure was measured to be 130/80 mmHg, pulse rate was 74 beats per minute (bpm), respiratory rate was 18 per minute, and the temperature was 36.0°C. On physical examination, there was a lump, located over the lateral and the posterior aspects of the left hip, which was non-pulsatile, minimally tender to touch with no change of color or any increase in temperature of the overlying skin.

A complete blood count showed total white blood cells (WBC): 7700 cells/mm<sup>3</sup> with neutrophil 60%, lymphocyte 21%, monocyte 9% and eosinophil 10%, hemoglobin (Hgb) 10.2 gm/dL, hematocrit (Hct) 45%, platelets 268 x 10<sup>3</sup>. Serum urea was 37 mg/dl and creatinine was 0.7 mg/dl.

The patient underwent contrast-enhanced CT (CECT) of the pelvis including the bilateral hip joints. It showed an expansile lytic lesion involving S1 to S4 vertebral body on the left side, extending laterally across the left sacroiliac joint into the ilium. The lesion was destroying the involved bones. There was associated solid-cystic soft tissue posteriorly, involving the left gluteus medius and minimus muscles, and anteriorly, involving the left iliac and the psoas muscle. The solid-cystic soft tissue component showed multiple coarse foci of calcification.

Ultrasound-guided FNAC yielded clear thick fluid. A smear examined revealed fragments of the laminated membrane against the background of scattered inflammatory cells, necrotic debris, and calcification. The cytomorphic findings were suggestive of hydatid disease.



**Figure:** NCCT pelvis bone window (fig. A) and soft tissue window (fig. B) showing an expansile, lytic lesion involving sacrum, left sacroiliac joint, and ilium with solid cystic soft tissue with a focus of calcification in soft tissue.

## DISCUSSION

Hydatidosis, caused by parasitic tapeworm echinococcus, is an endemic disease in our country. The liver and lung involvement together account for at least 90% of the cases, as most larvae are trapped there upon release of the embryo into

the portal circulation. Bone hydatidosis is a very rare entity accounting for only about 0.5 to 2.5% of the total cases. The vertebrae are the most commonly affected bones (50%) followed by the pelvis (25%), and the long bones (15–25%).<sup>5</sup> Skeletal infestation of *E. granulosus* cyst occurs by hematogenous seeding. The disease in the bone starts when scolices settle there. Unlike in other organs where visceral cysts expand at a slow pace and grow mostly concentrically, the rigid structure of the bone prevents this pattern from becoming established. In the case of bone involvement, pericyst formation does not occur, thereby allowing aggressive proliferation in an irregular, branching fashion, along the line of least resistance, especially the osseous canals, forming a branched, polycystic appearance.<sup>6,7</sup> There is slow resorption of the trabeculae, without cortical expansion. This occurs as a result of pressure. With time, the parasite reaches and destroys the cortex with subsequent spread of the disease to the surrounding tissue as was seen in our case. There is no inflammatory reaction of bone, however, secondary infections which lead to sclerosis, abscess formation, and draining sinuses may occur.

Diagnosing bone hydatid disease is challenging, as cases do not have a specific clinical feature and imaging characteristics. Lesions are usually osteolytic and can involve cortical bone and extend to the soft tissue.<sup>8</sup> CT scans show well-defined single or multiple cystic lesions that may cause cortical thinning without contrast enhancement. It may also show a pathological fracture, cortical destruction, and soft tissue extension with calcification.<sup>9</sup>

MRI is the most helpful diagnostic technique, especially in cases of soft tissue involvement or the spine. It is not possible to differentiate hydatid cyst appearance from malignancy based solely

on imaging findings.<sup>10</sup> An accurate diagnosis may be aided by eosinophilia (25 to 35% of all cases) and positive results of complement fixation tests, intradermal injection of hydatid fluid (Casoni test), and indirect hemagglutination tests over time. Therefore, hydatid bone disease should be considered in the differential diagnosis of osteolytic lesions, especially in endemic areas since the diagnosis of primary bone hydatid disease is difficult and requires a high index of suspicion.

### CONFLICT OF INTEREST

None

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None

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