Case Report

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Radiology Section

Multifaceted Approach in Diagnosing and Managing Pelvic and Hip Echinococcosis: A Case Report

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ABSTRACT

Echinococcosis, a zoonotic disease caused by *Echinococcus granulosus* or *Echinococcus multilocularis*, forms hydatid cysts in various organs, occasionally affecting the skeletal system. The present case study delves into a rare instance of pelvic and hip echinococcosis, underscoring the diagnostic and treatment challenges posed by such atypical manifestations. Hereby, the authors present a case of 42-year-old male, who presented with persistent lower back pain for four months. A combination of haematological tests, radiographic imaging and histopathological examination was employed to diagnose echinococcosis. Imaging techniques included X-rays, ultrasound, Non Contrast Computed Tomography (NCCT) and Magnetic Resonance Imaging (MRI), which revealed multiple cystic lesions with characteristics indicative of hydatid disease. Normal haematological tests showed no acute inflammation or infection. Radiographs displayed several expansile lytic lesions in the pelvis and femur, with double-walled cystic formations and interior septations being crucial for diagnosis. MRI showed the largest cyst in the perivesical space. Histopathology revealed laminated hydatid cyst walls and reactive alterations without cellular atypia. Echinococcosis should be considered when diagnosing cystic skeletal lesions, especially in endemic areas. This challenging condition requires a multidisciplinary strategy that integrates radiographic, serological and surgical methods, emphasising the need for increased awareness and diagnostic precision in atypical presentations of echinococcosis.

Keywords: Diagnostic imaging, Pelvic hydatid disease, Skeletal cysts, Zoonotic infections

CASE REPORT

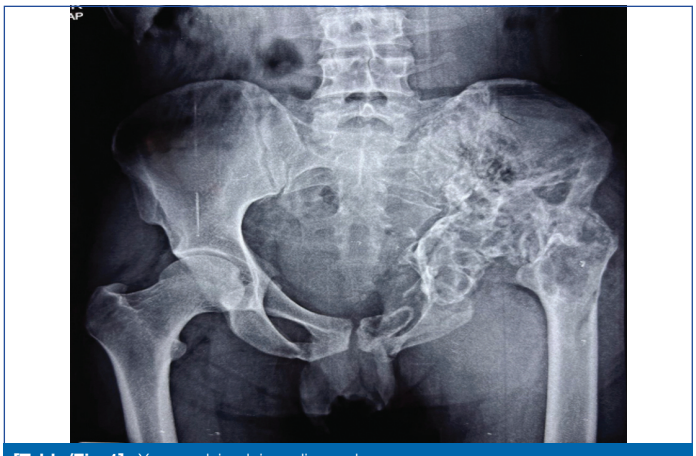
A 42-year-old male from Baran, Rajasthan, India, presented to the Orthopaedic clinic with a chief complaint of chronic lower back pain radiating to the left thigh and leg. This pain was unrelieved by rest and had persisted for four months. The patient was previously medication-naïve and reported no change in symptomatology with positional adjustments or periods of inactivity. He has no significant past medical history. The patient is a smoker and chews tobacco but has no history of drug use. Additionally, there is no relevant family medical history.

Comprehensive laboratory investigations were conducted, including a Complete Blood Count (CBC), C-reactive Protein (CRP) assay, Erythrocyte Sedimentation Rate (ESR), serum urea, creatinine and hepatic enzyme levels, specifically Alanine Aminotransferase (ALT) and Aspartate Aminotransferase (AST). These investigations yielded results within normal physiological parameters, indicating no acute inflammatory or systemic pathologies.

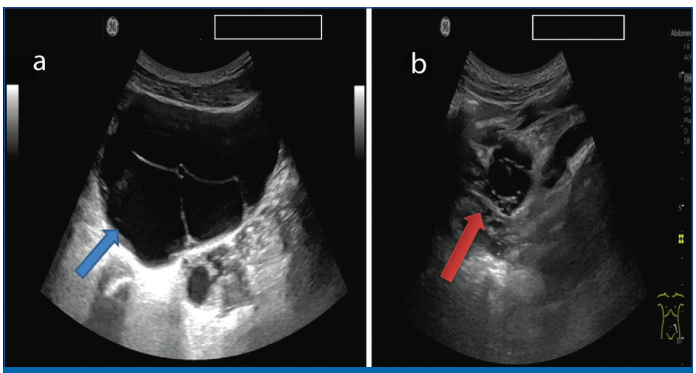
The consulting orthopaedist ordered anteroposterior and lateral pelvic radiographs and referred the patient to radiology to investigate the cause of the neuropathic pain, assess osseous involvement and detect any degenerative changes in the pelvic and lumbar regions.

The plain radiographic imaging of the pelvis revealed multiple expansile lytic lesions accompanied by cortical thinning. These lesions were observed in several locations on the left-side of the pelvis like the sacralala, the Sacroiliac (SI) joint, the iliac bone, the ischium, as well as, both the superior and inferior pubic rami. Additionally, similar lytic lesions with expansile characteristics were noted in the left femur, specifically involving the femoral head and neck [Table/Fig-1].

The ultrasound images of the pelvis demonstrated a distinct cystic structure with internal septations, characterised by a double-walled appearance, seen in the pelvic cavity [Table/Fig-2a] and in the intramuscular plane of the left thigh [Table/Fig-2b]. This is indicative of a hydatid cyst at stage Cystic Echinococcosis 2 (CE2) [Table/Fig-2]. The internal architecture of the cyst shows septations, a feature that suggests the presence of daughter cysts within the parent cyst, which is typical of echinococcal infections. These septations contribute to the complexity of the lesion and are important diagnostic features. The lesion is located within the pelvic cavity and extends into the intramuscular plane of the left thigh, indicating an aggressive nature of the cyst and potentially suggesting invasive growth beyond its original confines.

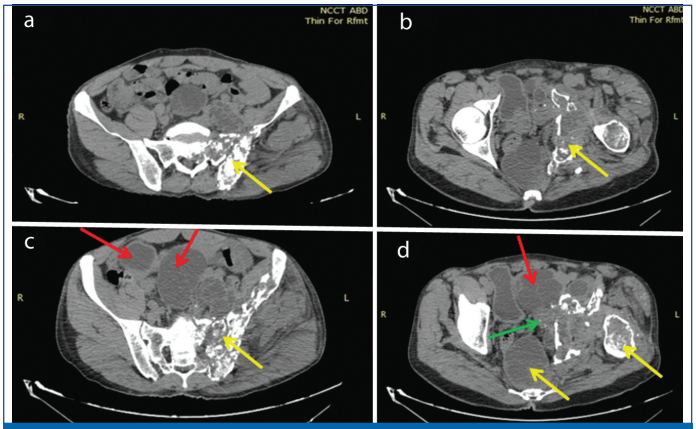


[Table/Fig-1]: X-ray pelvis plain radiograph.



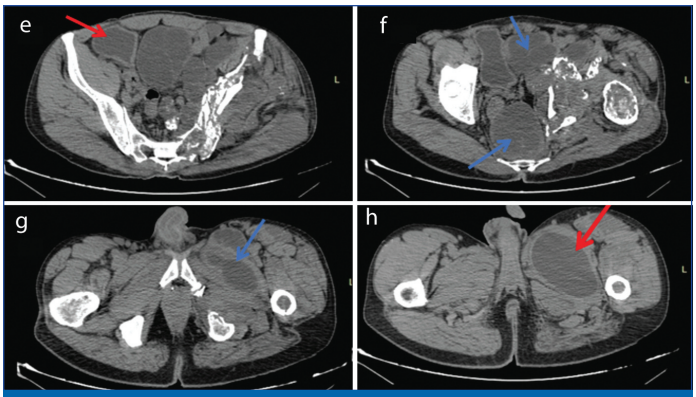
[Table/Fig-2]: B-mode Ultrasonography (USG).

The NCCT of the abdomen and pelvis demonstrated multiple lytic bone lesions involving the left sacral ala, left iliac bone, left ischium, and both the superior and inferior pubic rami on the left-side, as well as, the left femoral neck. The yellow arrow in the images highlights active hydatid disease with bone dissemination (incidence of bone echinococcosis). Accompanying these osseous abnormalities were numerous double-walled cystic formations with internal septations, with some showing internal calcification within the pelvic cavity and extending into the intramuscular compartment of the left thigh. The red arrow indicates hydatid cyst stage CE3, while the green arrow indicates CE4 stage of the hydatid cyst [Table/Fig-3a-d].



[Table/Fig-3a-d]: NCCT abdomen and pelvis.

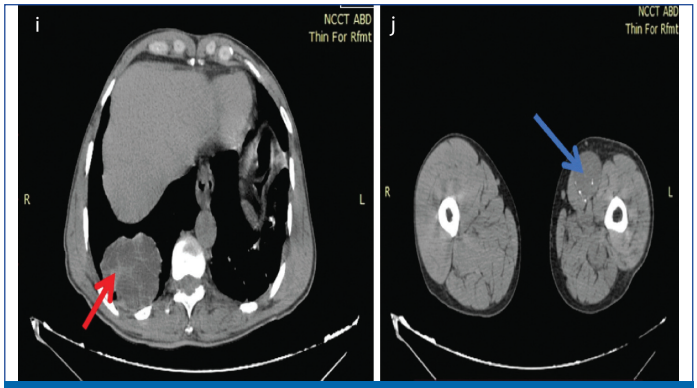
Furthermore, [Table/Fig-3e-h] of the NCCT abdomen revealed additional multiple variably sized well-defined double-walled cystic lesions, some of which showed internal septations. These lesions were distributed within the pelvic cavity, right inguinal region, perirectal area, along the left iliac vessels, within the musculature of the left thigh, and in the gluteal region (hydatid cyst). The red arrow indicates CE1 stage, and the blue arrow indicates CE3 stage of the hydatid cyst [Table/Fig-3e-h]. These findings were consistent with the lytic changes observed in the left sacrum, left pelvic bones, and left femoral head and neck.



[Table/Fig-3e-h]: NCCT abdomen and pelvis.

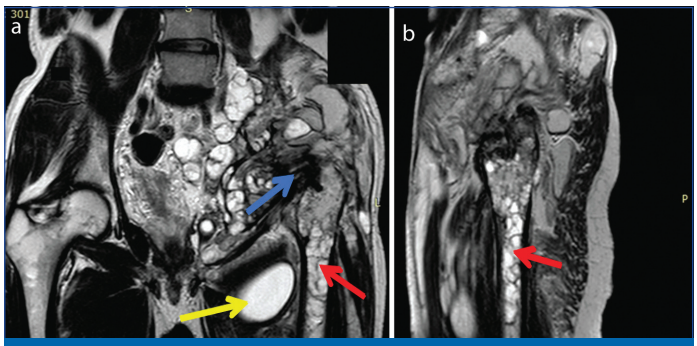
The NCCT [Table/Fig-3i,j] revealed a large, well-defined, double-walled cystic lesion with internal septations in the basal segment of the right lower lobe of the lung (red arrow: CE3 stage of hydatid), and another lesion within the left thigh musculature, which also showed a well-defined, double-walled cystic structure with internal septations and areas of internal calcification (blue arrow: CE4 stage of hydatid) [Table/Fig-3i,j]. These findings suggest disseminated cystic lesions affecting the left hemipelvis, proximal femur, pulmonary parenchyma, and left thigh musculature, indicative of a systemic condition with musculoskeletal and pulmonary involvement. The presence of septations and calcifications is consistent with a parasitic infection, potentially echinococcal disease.

Magnetic resonance imaging of the pelvis and femur [Table/Fig-4a,b] demonstrated multiloculated, well-defined intraosseous cystic lesions, appearing as T2 hyperintensities within the left sacral ala, iliac



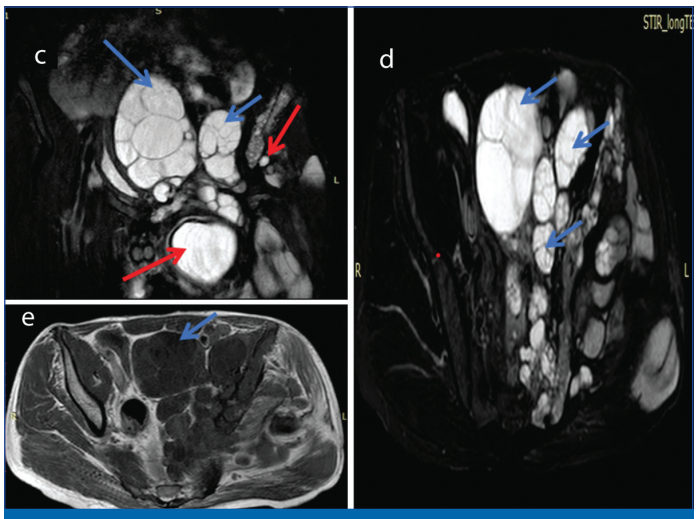
[Table/Fig-3i,j]: NCCT abdomen and pelvis.

bone, ischium, and superior and inferior pubic rami. These lesions extended into the left femoral neck and mid-diaphyseal region, measuring up to 12 centimeters in length (red arrow indicates bone echinococcosis). A pathological fracture was noted in the left femoral neck (blue arrow). Within the pelvic cavity and musculature of the left thigh, multiple well-defined double-walled cystic lesions exhibited T2 hyperintensity (yellow arrow indicates CE1 stage of hydatid) [Table/Fig-4a,b]. The extensive disease burden and associated pathological fracture require careful multidisciplinary management.



[Table/Fig-4a,b]: MRI of abdomen and pelvis.

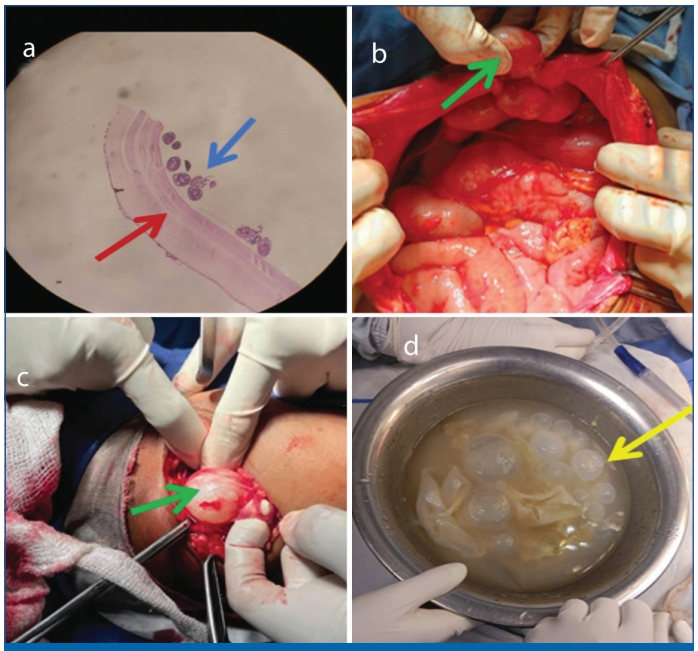
Further imaging [Table/Fig-4c,d] T2 Short Tau Inversion Recovery (STIR), [Table/Fig-4e] T1 Weighted (T1W) showed multiple well-defined T1W hypointense and T2 STIR hyperintense cystic lesions in the pelvic cavity and, left pelvic and gluteal musculature, including the largest cyst measuring 50×81×121 mm in the perivesical space. Of these cysts, some had double-walled multilocular cysts with internal daughter cysts (blue arrow indicates CE3 stages), while others were unilocular, with double membrane walls (red arrow indicates CE1 stage of hydatid). These images suggest active disseminated echinococcosis [Table/Fig-4c-e].



[Table/Fig-4c-e]: MRI abdomen and pelvis.

The histopathological examination [Table/Fig-5a] of the specimen under Haemotoxylin and Eosin (H&E), 40x magnification revealed

a laminated, acellular hydatid cyst wall typical of Echinococcus species (spp.) (red arrow). The blue arrow indicates multiple scolices of Echinococcus spp., with adjacent tissue displaying reactive changes but no atypia. During surgery, multiple cystic structures [Table/Fig-5b-d] were indicative of hydatid disease. The green arrow points to an intraoperative hydatid cyst, while the yellow arrow indicates a postoperative resected hydatid cyst found in the abdominal and pelvic cavities [Table/Fig-5a-d]. These clear cysts contained hydatid fluid and numerous echinococcal protoscolices or daughter cysts. The largest cysts were removed intact to avoid spillage and reduce the risk of anaphylaxis or secondary hydatidosis. The extracted cysts were immersed in saline for viability testing of the protoscolices and further pathological analysis. These findings indicate an advanced *Echinococcus granulosus* infestation, which was managed through meticulous surgical removal of the cysts and the administration of antihelminthic medication.



[Table/Fig-5]: a) Histopathological image (H&E; 40x); b) Cyst wall typical of Echinococcus species: c) Intraoperative hydatid cyst; d) Postoperative resected hydatid cysts (H&E stain).

Following the surgical resection of pelvic and intramuscular hydatid cysts, the recommended treatment was albendazole 400 mg, twice a day for three months. For inoperable hydatid cysts in bone, radiotherapy was advised at 2 Gy per fraction over 25-30 fractions. Radiotherapy could play a role as part of multimodal therapy in cases where surgery was not possible. Upon follow-up, the patient was stable. However, he was discharged against medical advice due to non co-operation during the postoperative stay. The patient's relatives later reported that he suddenly passed away within a month after discharge.

DISCUSSION

Hydatidosis, caused by *Echinococcus granulosus* or *Echinococcus multilocularis*, can form cysts in any body tissue [1]. Diagnostic radiological signs include cyst calcification, daughter cysts and membrane detachment, although these are not always specific [2]. Serological tests assist in diagnosis but are not infallible, and atypical cyst locations can delay treatment, increasing the risk of complications [3]. Cystic Echinococcosis (CE) in the skeletal system is rare, affecting 0.5-4% of cases [4]. Managing skeletal hydatidosis, particularly in areas such as the ilium and hip, is challenging due to high recurrence rates and significant morbidity [4].

Echinococcosis is a global zoonotic infection caused by larval Echinococcus species. It mainly manifests as CE from *Echinococcus granulosus* and Alveolar Echinococcosis (AE) from *Echinococcus multilocularis*, with CE being more common [5]. Humans become accidental hosts of *Echinococcus granulosus* through direct contact or by ingesting contaminated food or water. Ingested eggs release oncospheres that enter the bloodstream, forming cysts in various organs. The liver is the most affected organ (50-70% of cases), followed by the lungs (25%), while involvement of the brain, heart, and bones is rare. Bone echinococcosis occurs in 0.5-1% of cases, often asymptomatic, and is usually discovered incidentally through imaging for other conditions. Symptomatic cases arise from the mass effect of the cysts [6].

Radiologically, bone echinococcosis is characterised by osteolytic lesions that merge, leading to cortical thinning and destruction. Diagnostic signs include the absence of reactive osteoporosis and intralesional calcifications. Imaging may show osteolytic areas, occasional bone thickening, sclerosis, and periosteal reactions [7]. Ultrasonography (USG) is frequently used due to its accessibility and cost-effectiveness. Cysts can range from purely cystic to calcified masses. The World Health Organisation (WHO) classifies CE into several types: type CE1 (unilocular cysts) and CE2 (multilocular cysts) are active; type CE3 (cysts with daughter cysts) are transitional; and types CE4 and CE5 are inactive and calcified [8,9].

Computed Tomography (CT) is advantageous in certain clinical scenarios, such as in obese patients, those with excessive intestinal gas, or postoperative abdomens. CT can differentiate cyst characteristics, such as water-density fluid, internal septa and wall calcifications. It is superior to USG in detecting complications like intrabiliary rupture and cyst infection. Typical CT findings include rounded or oval cystic masses with well-defined margins and without contrast enhancement. However, bone hydatidosis on CT may mimic abscesses or tumours [10,11].

Magnetic resonance imaging excels in detailing liver echinococcosis cysts, showing a T2 hypointense rim indicative of the collagenous pericyst and hyperintense daughter cysts on T2-weighted images. It surpasses other methods in identifying cystic structures and assessing the integrity and viability of the cyst wall and internal architecture [12].

Advanced imaging techniques, accurate diagnosis, and effective management are crucial for pelvic and hip echinococcosis. Bhatnagar N et al., reported a 35-year-old male with hip pain, elevated ESR (52 mm/h), and eosinophilia (930/mL). MRI and CT scans confirmed the presence of multivesicular cysts, which led to surgical debridement followed by albendazole therapy [13]. Ganjipour Sales J et al., reported a 49-year-old male with sacral pain and a history of pulmonary and osseous hydatidosis. CT and MRI revealed multiple cysts measuring up to 10×4 cm. Serological tests {Western blot and Enzyme-linked Immunosorbent Assay (ELISA)} confirmed the diagnosis. Surgical removal followed by albendazole chemotherapy resulted in stable lesions and manageable residual hip pain at a seven-year follow-up [14]. Zaizi A et al., presented six cases of musculoskeletal hydatid cysts diagnosed through ultrasound and MRI. Imaging showed typical multivesicular appearances, with serology confirming hydatid disease in two patients. They underwent pericystectomy and removal of soft-tissue masses without rupture, along with postoperative antihelmintic therapy lasting 3-12 months for treating bone localisation [15]. Iken M et al., discussed a 37-year-old male with extensive pelvic hydatid disease revealed by MRI. Positive serology (Western Blot and ELISA) and hypereosinophilia confirmed the diagnosis. Long-term albendazole therapy stabilised the lesions over seven years, but the patient remained functionally handicapped, requiring crutches for mobility [16].

Bony involvement of hydatid cysts is a rare manifestation of hydatid disease that can be mistaken for sacroiliitis and can produce severe, escalating lower back pain. It is challenging to diagnose and differentiate from close differential diagnosis such as simple/septated cysts, aneurysmal bone cysts, giant cell tumours, fibrous dysplasia, chondrosarcoma, abscesses, lytic or sclerotic bone lesions, chronic osteomyelitis, neurofibromatosis and fibrocystic disease, as well as,

tuberculosis. Therefore, a multifaceted approach to diagnosis is essential [17-19].

Patients with pelvic and hip echinococcosis often experience delayed diagnosis due to non specific symptoms, complicating management. Advanced imaging reveals extensive disease, and treatment is challenging, with residual symptoms persisting despite prolonged therapy. The rarity of the condition and its similarity to other diseases further hinder timely diagnosis.

CONCLUSION(S)

The present case study illustrates the challenges of diagnosing and treating hydatid disease, particularly in rare locations such as the skeleton. Although skeletal hydatidosis is uncommon, the case underscores the importance of a multidisciplinary approach that includes radiographic, serological and histological evidence. Effective management requires the surgical removal of cystic lesions to prevent recurrence, as well as, pharmaceutical treatment to eliminate the parasitic infection. The current case highlights the global burden of echinococcosis and the need for vigilance in endemic areas to prevent significant morbidity.

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