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Case Report

Ensuring Accurate Diagnosis: The Imperative of Biopsy in Culture Specimens—A Rare Bone Pathology

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Introduction:
A plethora of pathologies can cause bone destruction. The acquisition of bone for culture and histological examination is critical for definitive diagnosis and the identification of causative pathogens, including their antibiotic susceptibilities. We present a rare case of intra-osseous hydatid cyst in a 56-year-old male. He initially presented to us with a 3-year history of right hip pain and a discharging sinus around the iliac crest and anterior superior iliac spine region. His history includes a debridement in which the specimen was sent only for cultures, not a biopsy. He was diagnosed to have an intra-osseous hydatid cyst based on the radiographic findings and underwent extensive debridement with tissue samples sent for histopathological confirmation, culture and sensitivity. In this case report, we present his case and review the literature.

Conclusion: Hydatid bone disease is an uncommon parasitic infection with features that make it difficult to distinguish it from other tumour-like lesions. Early diagnosis is challenging but crucial to decrease morbidity. We therefore reiterate the principle of evaluating every culture specimen for histological assessment to prevent misdiagnosis and inappropriate treatment.

Keywords: Hydatid disease, Hydatid Cyst, Biopsy, Core Needle Biopsy, Primary cell culture.

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Introduction:

Musculoskeletal infections are a major contributor to morbidity and can be difficult to manage. Bone infections occur when pathogens invade the bone, which can be pyogenic, non-pyogenic, or, in rare instances, parasitic[1]. Echinococcus granulosus, a tapeworm commonly found in Mediterranean regions, releases eggs that contain oncosphere larvae. These larvae can enter the human intestines, travel to organs such as the liver and lungs, and form hydatid cysts[2]. Hydatid disease in the bones, or osseous hydatidosis, is uncommon due to the rigidity of bone, which typically prevents cyst formation[3].
The acquisition of bone for culture and histological examination is critical for definitive diagnosis and the identification of causative pathogens, including their antibiotic susceptibilities. Bone specimens may be obtained percutaneously through uninfected skin or during surgical intervention. In cases where bone sampling is not feasible, it should be acknowledged that cultures from adjacent soft tissues may yield discrepant results, while swab cultures frequently overestimate the diversity of pathogens[4]. We present a rare case of a solitary intraosseous hydatid cyst in the ilium, with an unusual clinical presentation, initially misdiagnosed as a pyogenic infection due to nonspecific diagnostic investigations, and review the literature.

CASE REPORT:

A 56-year-old male patient with no comorbidities presented to our unit with a 3-year history of right hip pain and a discharging sinus around the iliac crest and anterior superior iliac spine region. There was no history of trauma. The patient did not report any family history of a similar disease. Before presenting to our unit, he had already undergone a debridement at another hospital, where culture specimens yielded methicillin-sensitive Staphylococcus aureus, and he was treated with antibiotics for over 12 months with no clinical response. He had also undergone a 6-month course of anti-tuberculous therapy, which the patient discontinued due to side effects and failure of clinical response.

He was referred to our sarcoma service following plain radiographs and an MRI by a general orthopaedic surgeon, where a concern of a neoplastic lesion in the hemipelvis was raised, which involved the right iliac blade and acetabulum. Radiographs of the pelvis demonstrated a large destructive lesion with multiple lucencies and foci of sclerosis involving the entire ilium extending into the acetabulum. There was sclerosis of the right sacral ala, too. Severe arthropathy of the right hip joint was also noted. No chondroid or osseous matrix was noted. The rest of the pelvis was normal. (Figure 1). MRI demonstrated multiple ill-defined destructive lesions involving the right ilium and sacral ala with multiple cysts in the bone and soft tissues, as well as a solid component. No chondroid or osteoid component was noted. (Figure 2) A provisional diagnosis of intraosseous hydatid cyst was made.

Following medical optimisation, he underwent extensive debridement of all the dead tissues and lavage with hydrogen peroxide and hypertonic saline through a utilitarian surgical approach to the hemipelvis in a semi-lateral decubitus position. During debridement, numerous cysts were noted extruding from the pelvic bones and soft tissues, which were pathognomonic of hydatid disease and sent for histological confirmation (Figure 3, 4). Samples were also sent for standard and TB (tuberculosis) cultures, along with gene expert analysis as a standard procedure.

Following debridement, standard closure was performed, with drain removal from the wound, and the patient was mobilised, weight-bearing as tolerated, the next day. Postoperatively, his pain improved significantly, and upon confirmation of hydatid disease on histology, the infectious diseases team was involved, and the patient was commenced on high dose albendazole for 12 months. He also had a systemic screening for the involvement of any other solid organs and was found to have an isolated disease. Other serological tests, including haemagglutination tests, were also performed purely for academic purposes.

On his last follow-up at 3 months following surgery, he continued his treatment without any adverse effects. His sinuses had healed completely, and he was mobile with a walking stick and continues to be under regular combined follow-up by our infectious diseases team.

DISCUSSION:

Hydatid cysts are mostly found in the liver and lungs, but they can also occur in other organs, including the myocardium, brain, eye, spleen, and bone. The diagnosis of bone hydatid cysts is usually based on radiological images and tissue biopsy.

In orthopaedics, it is crucial to follow a precise and accurate methodology when managing pathological lesions, as they can be malignant or benign, and mismanagement can lead to increased morbidity and even mortality. A comprehensive multidisciplinary approach is therefore necessary, and clinicians must maintain a high level of suspicion for uncommon diagnoses. It is therefore routinely practised and advised that any samples taken for culture and sensitivity testing must also be sent for histological analysis, and vice versa, to avoid errors, as in this case. Had a sample been sent for histology at the first surgery by the previous physicians, the diagnosis would have been made in a timely fashion and further morbidity reduced with the commencement of appropriate treatment.

One such rare bony pathology is hydatid disease, which can present with non-specific clinical and radiological features, making diagnosis challenging; hence, histological sampling and analysis are further important. Cystic echinococcosis is more prevalent in regions such as the Middle East, Central Asia, Western China, and the Mediterranean, particularly in developing countries[5]. The most common form of this disease affects the liver, though it can occur in other soft tissue organs as well[2]. Bone involvement is unusual due to the unique structure of bone and periosteum, and several

theories have been proposed to explain how the infection spreads to bone. One hypothesis suggests a mechanical process leading to ischemic osteitis, ultimately resulting in bone destruction without an inflammatory response[6]. Invasion of bone by the blood-borne scolex prevents the typical formation of a peri cyst, and instead there is aggressive growth along the path of least resistance through bone canals, leading to slow destruction and replacement of normal bone tissue between trabeculae by the expansion of multiple vesicles[7,8]. This gradual pathological process, combined with common but subtle symptoms such as nonspecific pain, swelling, and, in severe cases, pathological fractures, makes early diagnosis challenging. As a result, the condition is often misdiagnosed and can be mistreated.

Radiological findings are also nonspecific; plain radiographs may reveal a cystic or irregular, lytic lesion, while CT (computed tomography) may show localised, ovoid bone destruction. Occasionally, a "double-layered arcuate calcification" is observed, resulting from the calcification of the cyst wall[8,9]. MRI is more reliable, potentially showing a multilocular appearance, the cyst wall and its relationship to surrounding tissues; however, these findings are still not highly specific[7]. Serological tests are important, though they may be negative if the cyst has calcified, aged, or become dead, and the diagnosis should be confirmed by biopsy, even if a culture is performed. There is no effective conservative treatment for hydatid bone disease. Without aggressive surgical treatment and extended adjuvant therapy with Albendazole, high recurrence rates are likely[10,11].

In our case, the provisional diagnosis of hydatid bone disease was made radiologically and confirmed both intraoperatively with the presence of multiple cysts and postoperatively with histological analysis.

CONCLUSION:

In conclusion, hydatid bone disease is an uncommon parasitic infection with non-specific clinical and radiological features, making it difficult to distinguish it from other tumour-like lesions. Early diagnosis is challenging but crucial to decreasing morbidity. We therefore reiterate the principle of evaluating every culture specimen for histological assessment to prevent misdiagnosis and inappropriate treatment.

Declaration

Informed consent was obtained from the patient.

The authors declare that they have no conflict of interest.

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Consent to participate – yes

No AI was used

Figure 1: AP (antero-posterior) radiograph showing a large destructive permeative lesion involving the right ilium and sacral ala with multiple lucencies.

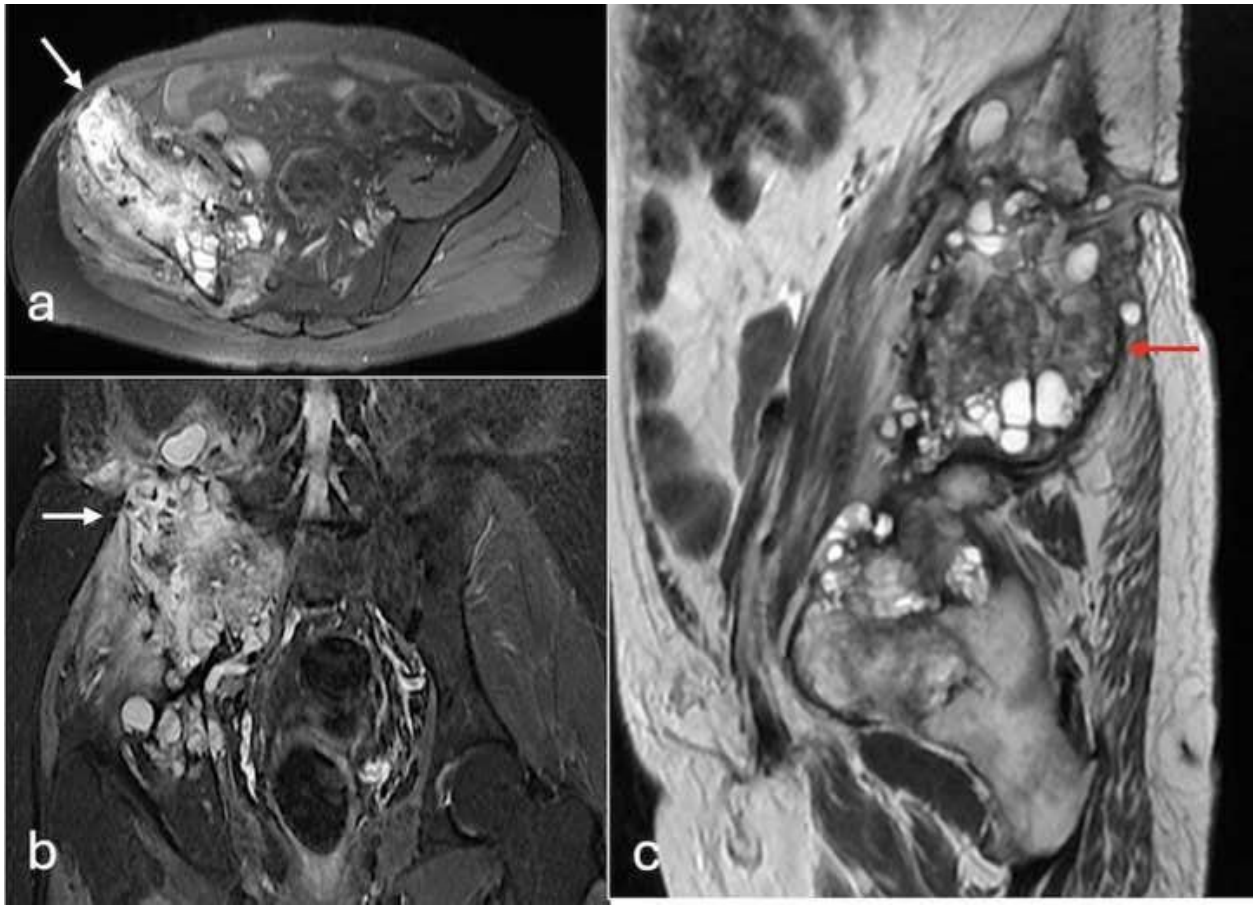

Figure 2: (a-c) – STIR axial(a), coronal(b) and T2 sag (c) showing a destructive lesion (arrow) involving

the right ilium and sacral ala with multiple cystic lesions extending into the adjacent soft tissues.

Figure 3: Multiple cysts and daughter cysts of Echinococcus granulosis from the iliac bone.

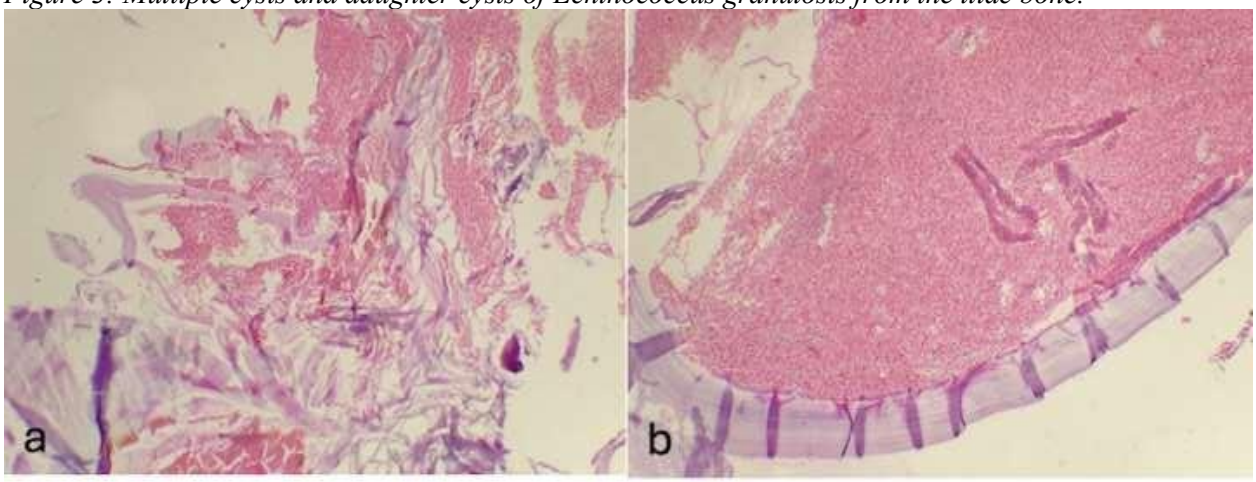

Figure 4: Haematoxylin and eosin stain (a, b) showing pink laminated gelatinous membrane suggesting hydatid cysts

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