HYDATID DISEASE OF BONE MASQUERADING AS MALIGNANCY: A CASE REPORT

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ABSTRACT:

Hydatid disease continues to be a significant health problem in under developed and developing parts of the world where farming and agriculture is common but no veterinary control exists. It is also encountered in developed communities where it is common in the immigrants or in people with a history of travel to endemic areas. Hydatid disease of the musculoskeletal system is one of the rarest but extremely debilitating, difficult to diagnose and even more difficult to manage clinical manifestations of the disease. We present a case report of hydatid disease of femur to highlight this rare but important manifestation of this parasitic zoonosis.

INTRODUCTION:

Hydatid disease is a parasitic zoonosis caused by the Echinococcus tapeworm. Dogs or other carnivores are the definitive hosts, whereas sheep or other ruminants are intermediate hosts. Humans are aberrant intermediate hosts, and become infected by ingesting food contaminated with eggs of the parasite. The embryos released from these eggs enter the portal circulation via the bowel mucosa where most of these embryos get trapped in the hepatic capillaries. Some of these embryos die while others grow into hydatid cysts. Only few of them pass through the capillaries to reach other organs [1].

Osseous hydatidosis is very rare (0.5%–2% of cases).The most commonly involved bones are the spine (35% of cases), pelvis (21%), femur (16%), tibia (10%), ribs (6%), skull (4%), scapula (4%), humerus (2%), and fibula (2%)[1]. When the embryo travelling in systemic circulation lodges in the bone it causes primary hydatid disease of bone. Pericyst, which is composed of modified host cells and forms a dense and fibrous protective zone, does not form in bone with resultant thinned walled cyst formation. The other factor that contributes to the cyst enlarging in an irregular branching is the compact ,rigid composition of the bone . The cyst thus follows the path of minimal resistance and does not acquire its normal spherical shape. The parasite replaces the tissue between osseous trabeculae due to the slow growth of multiple vesicles. The disease ultimately reaches and destroys the overlying cortex with subsequent spread of the disease to surrounding tissues . The osseous hydatid cysts does not usually calcify , as compared to the extraosseous hydatid cysts which usually show calcification[2].

Diagnosis of bone hydatid disease is challenging and is primarily based on imaging findings. In the spine, the main differentials are infection (osteomyelitis) and inflammation (tuberculosis spondylitis), however, it usually does not demonstrate any osteoporosis or sclerosis, and there is lack of damage to the vertebral discs. Extension beyond the spine into the adjacent tissues and the ribs ,in case of thoracic spine, is also commonly seen.

Hydatid disease in other bones appears as variable sized lytic lesions, some of which are confluent, resulting in thinning and erosion of overlying cortex[3, 4]. Pathological fractures and extension beyond the bony confines into the adjacent soft tissues is also seen [1]. In some cases, curvilinear calcification of the capsule may be seen .

These radiographic features can often lead to the misdiagnosis of infection(osteomyelitis), inflammation(tuberculosis), bone cysts or primary bone tumors (benign or malignant). Pluchino et al report that using plain x ray, osseous changes are found in 27% of cases and ‘‘moth eaten areas’’ with surrounding sclerosis are common. But Liu et al reported their starting diagnostic results by x ray; 15 cases are diagnosed as tuberculosis, 6 cases as metastases, 2 cases as a chordoblastoma,1 case as a psoas abscess, 2 cases as bone cysts, 2 cases as bone giant cell tumours, and in the remaining 9 cases no specific diagnosis was conclusively made[2].

As a result of the non-specific radiological findings and low index of suspicion, osseous hydatidosis may prove a diagnostic and therapeutic challenge.

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CASE PRESENTATION:

History: A middle aged female patient presented with chronic pain in right thigh for about 6 months , which was followed by severe acute pain and limitation of movement of the same limb for 3 days. She was a house wife with history of cattle rearing. On examination, there was warm, tender swelling of the distal third of the thigh with limited, painful movement of the knee joint. The initial blood tests showed normal haemoglobin, total and differential leucocyte counts and peripheral blood smear.

Plain radiograph of the limb was advised.

Figure 1: Xray showing pathological fracture with ill-defined lytic and sclerotic lesions in the distal femoral diaphysis with endosteal scalloping . No definite soft-tissue component identified.

An MRI femur was subsequently ordered, which was reported as highly aggressive lesion of distal diaphysis of right femur with soft tissue extension and displaced pathological fracture. The differentials of primary bone sarcoma and osteomyelitis were given.

This was followed by bone scan and CT Chest, Abdomen and pelvis to look for possible metastasis. Bone scan revealed abnormal tracer uptake in distal right femur at the sight of pathological fracture with the rest of the skeleton appearing normal scintigraphically. CT chest, abdomen and pelvis showed no abdominal, pelvic or pulmonary primary malignancy and no evidence of any hepatic, pulmonary, adrenal or peritoneal metastasis.

An excisional biopsy was eventually performed and was subjected to histopathological examination, which confirmed that the lesion was primary hydatid cyst of the bone.

The patient underwent screening for systemic involvement by the disease . Ultrasound abdomen and pelvis was normal as was the chest radiograph. The patient was referred to the Department for control of infectious disease where she was started on the therapeutic dose of the oral antihelminthic drug Albendazole. She used Oral albendazole off and on for 6 months with no significant clinical improvement . Xray was repeated and MRI on 2/8/21 revealed worsening of the disease process.

Figure 2; lateral radiograph of thigh showing displaced distal diaphyseal fracture of right femur with multiple poorly circumscribed lytic and sclerotic areas involving the cortex and medulla of distal femoral diaphysis and patella with resultant moth-eaten appearance. Endosteal scalloping is also seen. No definite soft tissue component identified.

MRI femur showed large altered signal intensity lesion in right femur with extension into proximal tibia and patella along with intra and trans articular extension and displaced pathological fracture.

Figure 3: Large altered signal intensity lesion involving diaphysis and distal metaphysis of right femur. The lesion appears hypointense on T1WI(a), hyperintense on T2WI (b) and STIR (c) sequences and show marginal enhancement on post contrast (d) images. It is extending via the distal femoral epiphysis into the synovial cavity resulting in expansion of joint space and supra patellar bursa. There is diffuse involvement of patella. Trans-articular extension is also seen involving tibial plateau. There is associated displaced fracture of femoral diaphysis.

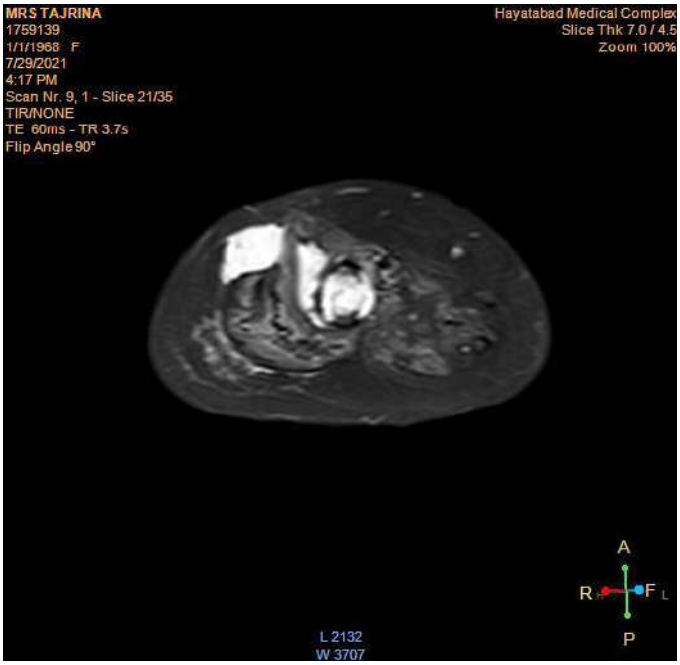

Figure 4; Axial STIR image showing extension into the subcutaneous tissue via a defect in adjacent muscles(vastus lateralis and intermedius muscles)

She was offered right hip disarticulation for the non-salvageable hydatid disease. Surgery was done on 28/8/21 which was uneventful. The surgery was covered with Praziquantel plus Albendazole .Thereafter the patient was sent home on Praziquantel for 3 days and Albendazole for 28 days as advised by the infectious and tropical disease control unit.

The recent review showed that the patient had only minimal pain. She could walk with crutches. Her maintenance treatment is one monthly oral Albendazole every year.

DISCUSSION:

This disease is a worldwide public health problem, especially in underdeveloped and developing parts of the world where farming and agriculture is practised without proper precautionary and hygiene measures [5].It is also encountered in developed communities where it is common in the immigrants or in people who have travelled to endemic parts of the world.

While hydatid disease can occur in any part of the body, primary osseous hydatid disease is very rare, reason being trapping of majority of the larvae in capillary beds of the lungs and liver. Incidence of bone hydatid disease is reported to be 0, 4%-5% from various studies and the vertebral column is involved in ≥50% of these cases[6]. Lack of awareness of medical community regarding osseous hydatidosis often results in misdiagnosis leading to poor therapeutic results as well as high rates of complications and poor financial impacts. Results of an Italian study showed that bone hydatid disease pose a significant healthcare problem with the estimate of average national financial burden being 4 million euros per year.

Familiarity with imaging features along with laboratory and clinical findings and high index of suspicion , especially in endemic parts of the world can help in early diagnosis. Hydatid disease should be included in the differential diagnosis whenever a cystic lesion is seen in any part of the body. Moreover, further research should be carried out on epidemiology, diagnosis and awareness of hydatid disease in Pakistan and other endemic regions to make proper management and eradication possible[7]. Timely diagnosis can help eradicate the disease ,while missed/delayed diagnosis on the other hand can result in dire consequences like amputation, recurrence and sepsis.

CONCLUSION:

In summary, primary hydatid disease of bone is an extremely rare manifestation of echinococcus zoonosis. The clinical course leading to diagnosis can be long and difficult because of the various and non-specific clinical and radiological features. Osseous hydatidosis remains associated with a high risk of morbidity, disability and mortality, that is why imaging, serological tests and PCR should be done immediately to lead to an early diagnosis and timely medical or surgical treatment. The long-term outcome depends on the complete resection of all parasitic lesions, which is often hampered by the infiltrative nature of the disease. We can conclude that the bony hydatid cyst should be considered in the differential diagnosis when there are cystic lesions that cause osteolysis, cortical breaks and/or pathological fractures to provide the patient with optimal treatment and rapid recovery, and then rehabilitate him for his daily and working activities.

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