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Case Report

Hydatid Cyst in Proximal Femur: A Case Report

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Abstract

Hydatid cysts, caused by the larval stage of the tapeworm Echinococcus Granulosus, are rare in the musculoskeletal system. We present a unique case of a hydatid cyst found in the proximal femur / hip joint of a patient with an implanted hip prosthesis. A 75-years old male with a history of cemented Hemi-arthroplasty (Thompson prosthesis) presented with hip pain and limited (painful) range of motion of the hip. Radiologically osteolysis was appreciated, suggestive of implant loosening. Surgical intervention was done. Per-operative cystic lesions were found. Clinical diagnosis of Hydatid disease was made and the cysts were successfully treated with Hypertonic saline and albendazole. Histopathological examination confirmed the diagnosis of Hydatid cyst. This case highlights the importance of considering Hydatid cyst as a rare, differential diagnosis in patients even with a prosthetic joint, residing especially in endemic regions of the world, which would require multidisciplinary approach for their management.

Keywords: *Echinococcus Granulosus, Proximal Femur, Hip prosthesis, Peri-prosthetic, Hip arthroplasty*

Introduction

Echinococcus, particularly E. Granulosus (bone cyst), is the causative agent of hydatid disease of the bone. All continents except Antarctica have the disease, but it is endemic in parts of northern and eastern Africa, South Asia, Central Asia, the Middle East, South and Central America, and Europe. Locality associated with the prevalence of sheep farming in these regions. The primary hosts of Echinococcus are dogs and foxes, while intermediate hosts include sheep, pigs, and humans. [1]

Hydatid cysts are commonly present in the liver, and skeletal presentation is rare, with a prevalence of 0.5 to 2 percent. The spine (35%) is the most common site for hydatid disease of bone, followed by the pelvis (21%), the femur (16%), and the tibia (9%).[2] The disease is difficult to diagnose due to its delayed clinical manifestation and the initially nonspecific symptoms it presents with. Its resemblance on radiological screening to bone tumors or bone cysts can result in a misdiagnosis. The mean age at diagnosis of the disease is 52.[3]

The cysts invade the cancellous trabecula and medullary canal, causing hematogenous seeding that result in the bone manifestation. The polycystic kind of cyst observed in bone develops when the cysts grow and give rise to daughter cysts. The damage is generated by three mechanisms: (i) mechanical expansion and compression of the surrounding tissues; (ii) ischemic occlusion of the nutrition arteries; and (iii) cellular proliferation of osteoclasts around the compressed bone tissue. Primary bone lesions are infrequent and are assumed to be the result of hematogenous dissemination from another soft tissue site. [4]

Case Presentation

75 years old, male, Diabetic, presented to our clinic with complaints of progressive left hip & thigh pain, primarily startup pain with limited range of motion. The patient had a history of left hip Hemiarthroplasty with a Thompson (Cemented) prosthesis for a traumatic neck of femur fracture (Secondary to road traffic accident) about 8 years back. Physical examination revealed tenderness in the left thigh and groin region and painful terminal range of motion in the left hip joint. No signs of systemic infection or allergies were noted. Laboratory investigations showed no significant abnormalities, including normal inflammatory markers.

Initial radiographs displayed signs of loosening of prosthesis, with halos at the cement-bone interface and cement-prosthesis interface especially at Gruen zones 5, 6, 8, 9 and 12 which revealed cystic (halo) lesions adjacent to the prosthesis in the (Figure 1 & 2). Aseptic implant loosening was suspected.

Figure 1: *Plain AP view radiograph with cement loosening.*

Figure 2: *Plain radiograph lateral view of left hip joint with cemented Thompson prosthesis in-situ with loosening.*

After obtaining informed consent, the patient was initially planned for revision cement-less total Hip arthroplasty. Intraoperatively, jelly like multiple cysts were found in the Vastus lateralis muscle, which raised a high suspicion of Hydatid disease. (Figure 3)

The surgery was put on a standstill and prompt arrangement of albendazole & hypertonic (3%) solution was made and surrounding structures were walled off with the hypertonic solution-soaked abdominal sponges to prevent further dissemination. The cysts were washed with Albendazole.

Implant was removed and the Acetabulum was packed with hypertonic saline soaked gauze. Cement was removed. The Femur was re-explored for any residual cysts, none were found. The exposed area was washed again with hypertonic solution followed by Albendazole solution and curettage of the cystic region of the bone was done.

Figure 3: *Encircled image of Hydatid cyst per-operatively.*

Figure 4: *Per-operative image of wound after clearing the hydatid cyst.*

Wagner cementless prosthesis for total hip arthroplasty was arranged for Vancouver type B3.

Surgery was completed and biopsy was taken and sent for histopathology. Patient received Albendazole (10mg/kg) therapy postoperatively and to be continued for three months. Histopathology was reported positive for Echinococcus Granulosus (Figure 6).

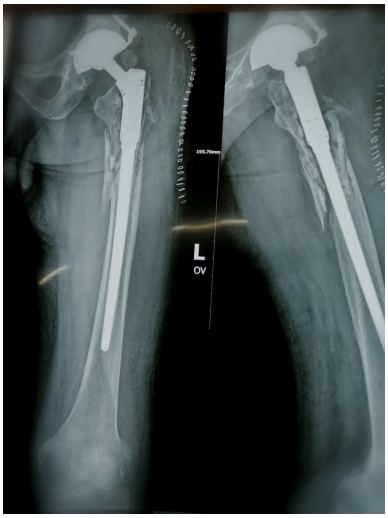
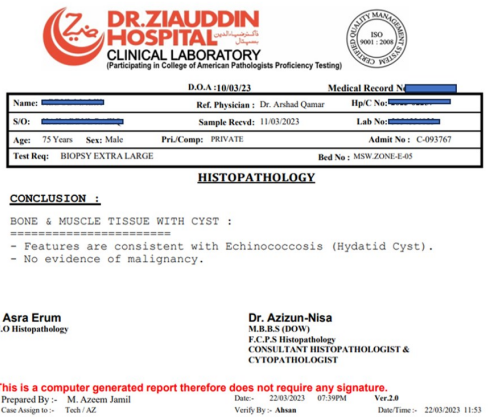


Figure 5: *Post-operative image with Total Hip replacement with a Wagner prosthesis.*



DR.ZIAUDDIN HOSPITAL
CLINICAL LABORATORY
(Participating in College of American Pathologists Proficiency Testing)

D.O.A :10/03/23 Medical Record No [redacted]

Name: [redacted] Ref. Physician : Dr. Arshad Qamar Hp/C No: [redacted]
S/O: [redacted] Sample Recvd: 11/03/2023 Lab No: [redacted]
Age: 75 Years Sex: Male Pri./Comp: PRIVATE Admit No : C-093767
Test Req: BIOPSY EXTRA LARGE Bed No : MSW.ZONE-E-05

HISTOPATHOLOGY

CONCLUSION :

BONE & MUSCLE TISSUE WITH CYST :
- Features are consistent with Echinococcosis (Hydatid Cyst).
- No evidence of malignancy.

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Figure 6: *Histopathological examination of the excised cyst revealed the characteristic laminated membrane consistent with a hydatid cyst.*

The patient's postoperative course was uneventful, with significant improvement in hip pain and restoration of joint function. At 4-week follow-up the patient was symptom free and doing well.

Discussion

Hydatid disease of bone is caused by the larval stage of Echinococus granulosus, a cestode. The worm is known as the dog tapeworm due to dogs being one of its definitive hosts. The life cycle of the worm is as follows: the adult form of the worm, is usually 2 to7mm in length and resides in the intestines of the definitive host, attached to the mucosa with hook-like structures. The gravid praglotidds lay eggs, which are excreted through the feces of the host. These eggs are consumed by the intermediate hosts, commonly sheep. The eggs then hatch in the intestines, releasing oncospheres. These oncospheres migrate through the intestinal wall into the blood stream, where they go to different organs, notably the liver and the lungs, wherein they develop into thick-walled hyatid cysts. These cysts contain protoscolices and daughter cysts, which are introduced back into the definitive host when they injest cyst-containing viscera, thus completing the cycle. Humans may also become intermediate hosts secondary to contact with the definitive host or through consuming contaminated water or food.[5]

Hydatid cysts are made up of three layers: The inner-most germinal layer containing the larval stage of Echinococcosis, the middle layer made up of laminated, acellular membrane, and the outer-most layer, Oradventitia, also known as the Pericyst. The Pericyst does not form in bones, allowing aggressive proliferation of the parasite and the formation of daughter cysts that can spread to other bones, resulting in a polycystic disease. The parasite invades the cancellous trabeculae and medullary canal and causes destruction of the bone by compression of the surrounding tissue through expansion. Other mechanisms of destruction include destruction due to ischemia resulting from compression of nearby vessels and indirectly increasing osteoclast activity around compressed bone tissue. The parasite may reach the cortex and result in erosion, leading to pathological fractures.[4]

Hydatid bone disease is difficult to detect, resulting in a late diagnosis. Delay in diagnosis is frequently associated with the parasite remaining dormant for several years before displaying nonspecific clinical symptoms. Typically, radiological tests are performed to diagnose patients. early diagnosis is mainly based on an x-ray, which may indicate bone degeneration or cysts, but the image is similar to giant cell tumor of bone and other bone pathologies, which may result in a misdiagnosis. Computed topography is a valuable tool used for diagnosis. A space occupying lesion (Oval or round in structure) with "double layered arcuate calcification" is a a typical CT presentation for Echinococcosis. However, in most cases, a conventional look is not present, complicating the diagnosis.

MRI is the best radiological tool to aid in diagnosis of Hyatid cysts because it shows the characteristic symptoms of a Pericystic wall and its multilocular nature, along with specifying the location of the lesion in respect to its surrounding structures.[5]

Immunological assays can now detect circulating antigens, specific antibodies and immune complexes. They are more accurate in ruptured cysts and frequently found negative in an ageing or calcified Hydatid cyst. Serologic examinations are classified into two groups: Antigen detection from Protoscoleces and Hydatid fluid, with the main antigenic components Ag5 and AgB25, and antibody detection in blood serum. Specific antibody examinations include the Casoni test, indirect hemagglutination, counter immuno-electrophoresis, gold-labelled antibody and ELISA. ELISA is mainly used as a confirmatory test due to its high specificity and sensitivity. Studies have confirmed final detection rate of ELISA to be 97.7%. [6] The eight tests of immunodiagnostic, including ELISA and gold-labelled methods, are the most specific and sensitive serological tests for diagnosis. These tests detect four types of antigens, with a sensitivity of up to 92.6% and specificity of 91.9%. Detecting specific IgG subclass antibodies is significant for early disease detection and prognosis following treatment. [5] Early diagnosis being uncommon due presentation of patients at an advanced stage of disease makes management challenging.

Conclusion

In this case study, we present a rare case of a Hydatid cyst found in the proximal femur / hip joint of a patient with an implanted hip prosthesis. Awareness of this uncommon presentation is crucial to consider Hydatid cyst as differential diagnosis of cystic lesions adjacent to prosthetic joints. A multidisciplinary approach involving orthopedic surgeons, infectious disease specialists, and radiologists is essential for accurate diagnosis and optimal management. Timely surgical intervention, complete cyst excision, and appropriate antehelmintic therapy are key factors in preventing recurrence and complications associated with Hydatid cysts in such unique locations.

Conflict of Interest

The authors declare no conflict of interest.

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