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Case Report

Extensive humeral hydatid cyst with extraosseous involvement: a case report

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ABSTRACT

Hydatid cysts caused by *Echinococcus* species mainly involve visceral organs. Rarely skeletal involvement occurs. The spine and pelvis account for half of the cases of osseous hydatidosis, making humeral hydatid cysts one of the rarest conditions. We report a case of humeral hydatidosis in a 42-year male who presented to us with pain and swelling over the left upper arm, radiological investigations suggestive of osseous cystic lesions were inconclusive and diagnosis of hydatidosis was confirmed with a biopsy subsequently. Surgical excision and curettage of bone and surrounding soft tissue were done, and the void was filled using PMMA cement followed by antihelminthic chemotherapy. No complication or recurrence has been found postoperatively at the end of 6 months of follow-up, with preservation of limb function. To keep orthopedic surgeons aware of this morbid condition, due to its low prevalence, and advanced presentation which is likely misdiagnosed, and even after the early intervention, there is a high recurrence rate which makes this one of the rare cases of humeral hydatidosis notifiable.

Keywords: Hydatid cyst, Hydatosis, Cystic echinococcosis, Muscular echhinococcosis, Parasitic disease

INTRODUCTION

Hydatid disease is a parasitic disease, endemic in Australia, South America, the middle east, and central Asia. It is primarily a zoonotic disease, with the highest prevalence in India, reported from Saurashtra, Andhra Pradesh, and Tamil Nadu. Hydatidosis is common among cattle rearers in the Saurashtra and Kutch areas of Gujarat state.[1-3] Hydatid disease primarily occurs in the liver (65-75%) and lungs (25-30%), it occurs in bone only in 0.5-2.5% of all patients mimicking locally malignant lesions radiologically.[4] Osseous cystic echinococcosis (CE) most commonly affects the spine (35%) and pelvis (21%) followed by long bones like the femur (16%).[5] Among all the bones, the humerus is one of the rarest to be involved by CE. Osseous CE presents with pain and swelling associated with other non-specific symptoms, causing a major diagnostic dilemma and mimicking other entities. Even after radiological evaluation, cystic lesion of osseous CE is difficult to distinguish from malignancy and needs histopathological confirmation. Many cases of hydatidosis resembling tuberculosis (TB) have been noted. Lack of osteoporosis and sclerosis in host bone and absence of disc space and vertebral body damage, help to differentiate osseous CE from TB spine.[6] Due to presentation at an advanced stage in the majority of cases, lesions extend into surrounding soft tissue and neurovascular structures, making it difficult to eradicate CE, so along with surgical management, chemotherapy is given to prevent recurrence.[6]

CASE REPORT

A 42-year-old male presented with pain and swelling over the left upper 3rd arm with no complaints of fever/weight loss/prior trauma. On local examination, diffuse swelling

and tenderness were present with pain and limited range of motion for 5 years. Radiological investigations were suggestive of an osteolytic, multicystic lesion involving proximal 2/3rd humerus and causing cortical disruption at the greater tubercle leading to surrounding muscular spread but no intraarticular involvement of the shoulder joint (Figures 1 and 2). Histopathological confirmation of hydatid cyst was done by Biopsy. The routine hematological investigation was suggestive of eosinophilia and raised ESR. USG and CT abdomen was done to rule out visceral hydatidosis. No other hydatidosis location was found. Surgical intervention in the form of intralesional extended curettage with marginal resection of soft tissue was performed via a utilitarian shoulder approach. Metaphysis and diaphysis bone cortex was removed with surrounding tissue and chemical sterilization was done with hypertonic saline to the bone canal, the void filling was done with 80 gm of Polymethylmethacrylate (PMMA) antibiotic cement (Figure 3). Post-operatively, antihelminthic therapy was prescribed as albendazole 400 mg 3 times a day for 1 year. At the end of 18 months of follow-up, the patient has a complete range of movements in his left shoulder with discomfort on terminal abduction, not his routine activities. No postoperative complication or recurrence is found to date (Figure 4).

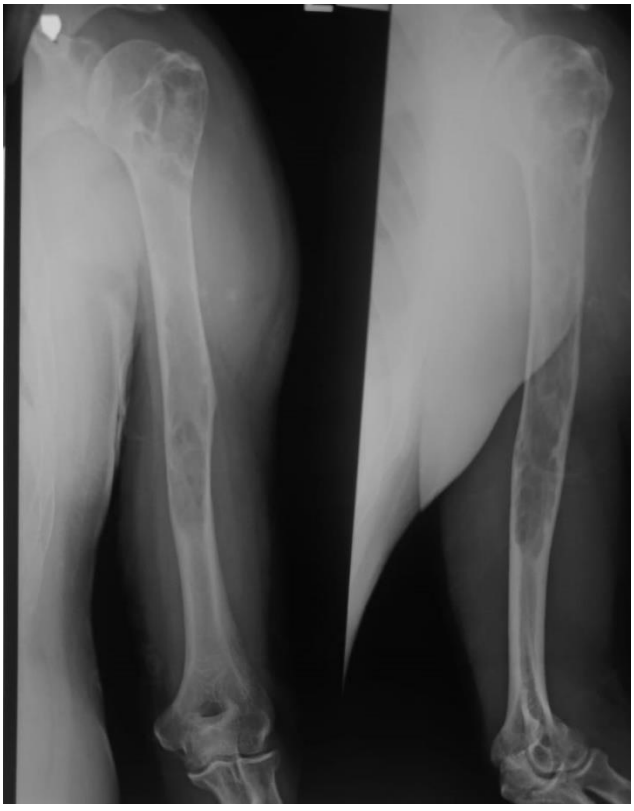

Figure 1: Plain radiograph of the left humerus: At the time of presentation the patient had complaint of left upper arm pain, on radiological evaluation it was found to have osteolytic lesions with cortical thinning involving more than half of the humerus.

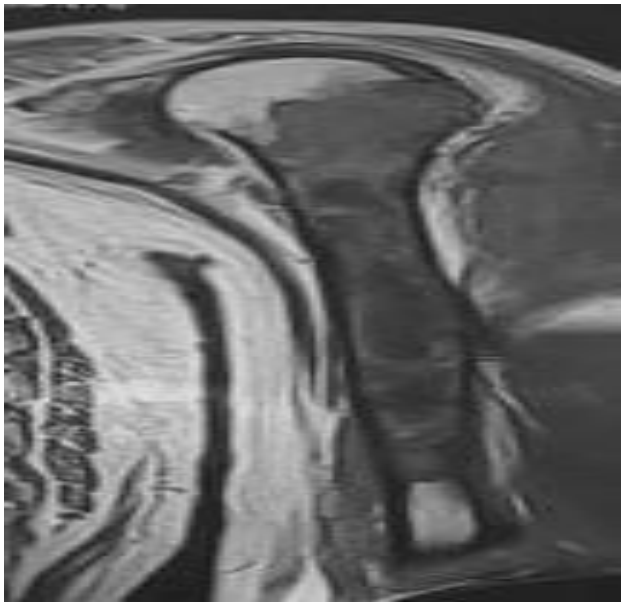

Figure 2: MRI of left shoulder: on further evaluation, T2 weighted MRI showed multicystic lesions involving metaphysis and diaphysis of the humerus with a cortical break at the greater tubercle and the surrounding tissue involvement suggestive of the diagnosis of hydatidosis. After which histopathological confirmation of hydatidosis was done.

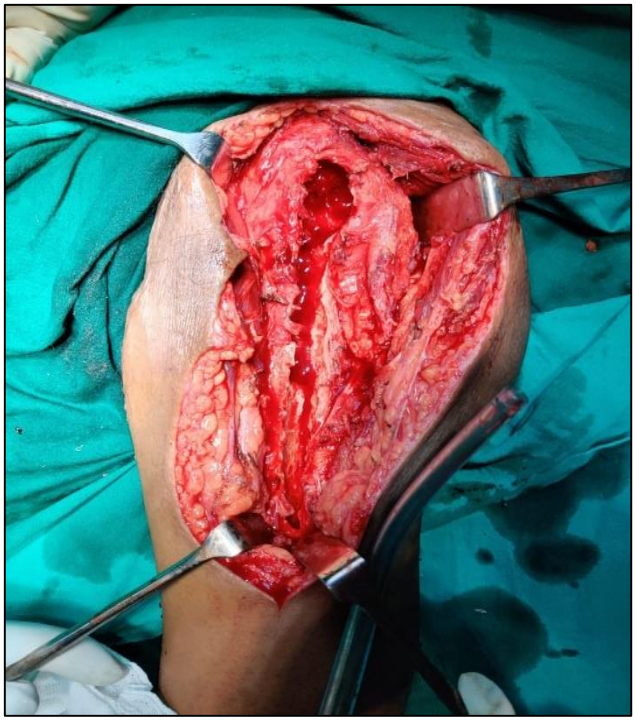

Figure 3: Intraoperative findings: Via utilitarian shoulder approach, the proximal humerus and shaft were exposed, along with all the daughter cysts, infected part was removed. Extended curettage was done using phenol and the void filling was done using PMMA cement. High-speed burr was used to curettage under challenging areas.

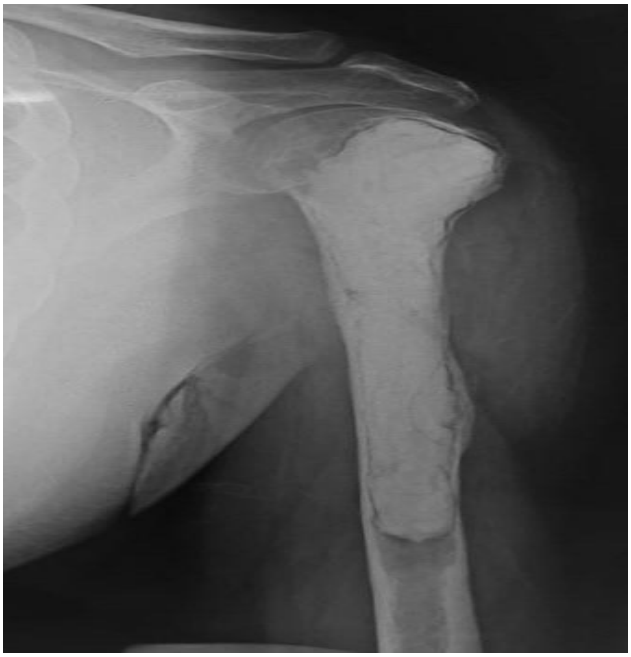

Figure 4: Follow up: Plain radiograph was done every 6 months to look for recurrence. The patient was on anti-helminthic therapy for 12 months and at 18 months of follow-up patient doesn't have satisfactory range or movement without any complications or recurrence.

DISCUSSION

Skeletal involvement by *Echinococcus* granulosus is rare as it occurs in only 0.5-2.5% of all hydatid cases. Among the osseous CE, the spine is commonly involved; especially the thoracic region is most frequently involved in about 35-50%. Osseus CE is most commonly the primary infection and not the result of extension from neighboring soft tissue.[1] Primary bone involvement occurs due to the settlement of blood-borne scolexes, the proliferation of cysts occurs in the bone canal and mimics locally malignant lesions radiologically.[7] Primary involvement of bones is rare and humerus involvement has hardly been reported. In our case with primary bone involvement, cortical thinning was a present and cortical break over the greater tubercle was present which resulted in surrounding soft tissue involvement. There was no intraarticular extension or neurovascular involvement. Soft tissue involvement necessitates wide excision with safe healthy margins.

The presentation of the disease is in the advanced stage and so severe that the condition has been called "white cancer".[8] The lesion in the bone may lie dormant for up to 10-20 years.[9] Hydatid cyst of bones remains asymptomatic over a long period and usually present with secondary infection or pathological fractures or after compressive myelopathy in vertebral lesions. After pathological fractures in long bones due to hydatid, non-union is common.[10] When CE affects long bones, it generally affects metaphyseal regions and later on extends to diaphysis and nearby bone. It is difficult to distinguish hydatidosis from other disease entities after radiological evaluation, and without histopathological evidence, even serological tests have limited value. In early stages, bony involvement mimics osteomyelitis and in later stages when cysts progressively enlarge and fill the medullary cavity, erosion of bone occurs and leads to osteolysis, which radiologically mimics aneurysmal bone cysts, giant cell tumors, fibrous dysplasia and other neoplastic lesions.[3] Thus histopathological investigation is confirmatory.

In India where osseous tuberculosis is so common and due to similar clinical presentation and radiological appearance, hydatidosis can be misdiagnosed as TB. Even a few cases have been reported where patients were put on anti-tubercular treatment which was later on diagnosed as bony hydatosis.[11] The only native treatment is complete surgical resection with wide healthy margins.[9-13] This is difficult to achieve but incomplete removal results in recurrence. We did surgical excision by removing all surrounding involved soft tissue. Extensive curettage was performed and chemical sterilization using hypertonic saline was done to remove any dormant cysts. To fill the void and bony defect of about 20 cm, PMMA antibiotic cement was used. PMMA is preferred as it increases the temperature of polymerizing cement which has a necrotizing effect and kills daughter cysts. The use of PMMA has reported not only excellent results but also reduced recurrence rate.[9]

Postoperatively, antihelminthic chemotherapy can be given using various drugs like albendazole, mebendazole, and praziquantel. Albendazole has been found to have superior efficiency among other antihelminths.[12] WHO recommends 10-14 mg/kg/day for 4 weeks out of 6 weeks and treatment should be continued for more than 3 months, and in some cases up to 1 year.[8] Long-term follow-up after surgery with chemotherapy is needed to rule out recurrence.

Humeral hydatidosis is very rare and due to delayed presentation with the associated risk of surrounding neurovascular involvement, our goal should be aimed at maintaining limb function with the prevention of recurrence. Even though low prevalence, the differential diagnosis of bony hydatidosis should be kept in mind while dealing with the bony cystic lesions, as delayed and advanced presentation of this disease may aggravate the risk of amputation, recurrence, and sepsis.

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