

## Recurrent Hydatosis at the Site of Non-union Humerus Fracture

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### ABSTRACT

Hydatid disease is still endemic in several regions of the world and is caused by two species of tapeworms, *Echinococcus granulosus* and *Echinococcus alveolaris*. It primary involves liver and lung, and bone involvement is relatively rare (0.2–4%), where it is most commonly seen in the spine. The skeletal involvement is usually due to secondary extension such as hematogenous spread. The disease has usually a silent manifestation until a complication exists; so, many cases are diagnosed intraoperatively. Treatment of hydatid disease because of its bone involvement and spillage of fluid with subsequent contamination seeding is difficult, so it has a high mortality rate and many cases will recur. Therefore, we can prevent these occurrences if we treat hydatid disease completely and in the primary stage. Adjuvant medical treatment, if the diagnosis is known, prevents systemic spread and recurrence. Here, we present a primary recurrent hydatosis at the site of non-union humerus fracture. We have pointed out osseous hydatosis as one of the important differential diagnoses in destructive bone lesions and the necessity of its radical resection.

**Key words:** Echinococcosis, humerus, hydatid, non-union, recurrent

### INTRODUCTION

Hydatid disease, also known as echinococcosis or hydatidosis, is a parasitic infection caused by the larval stage of two species of the tapeworms, *Echinococcus granulosus* (cystic hydatid disease) and *Echinococcus multilocularis* (alveolar hydatid disease). The former is endemic in several areas, mostly in sheep-raising communities such as South America, Mediterranean countries, Russian Federation, Middle East, and Central Asia.<sup>[1]</sup> The complete life cycle involves two hosts. The definitive host is usually a dog and the adult worm of the parasite lives in the proximal small bowel of the definitive host.<sup>[2]</sup> The intermediate host of the larval stage of *E. granulosus* is usually sheep, but tapeworms can also occur in cattle, pig, and other domestic livestock. Humans may become intermediate hosts through contact with a definitive host (usually a domesticated dog) or by ingestion of contaminated water or vegetables. The ovum loses its protective chitinous layer as it is digested in the duodenum. The released hexacanth embryo, or oncosphere, passes through the intestinal wall into the portal

circulation. The liver and lungs filter the eggs in 70% and 20% of the cases. Not filtered out by these organs, the eggs are carried into the general circulation to involve the brain, kidney, bones, and other tissues.<sup>[3]</sup>

Hydatid disease may develop in almost all parts of the body. The overall incidence of organ infestation is greatest in the liver (50–77%) and lungs (8.5–43%).<sup>[4]</sup> Incidence of bone echinococcosis is much lower, about 0.5–4% of the total reported cases. Osseous hydatosis is usually silent for decades. Primary bone manifestation is much rarer than secondary one. In bone involvement, pericyst formation does not occur, thereby allowing aggressive proliferation in an irregular branching fashion along the line of least resistance, especially the bone canals.<sup>[5]</sup> Early diagnosis is uncommon and is primarily based on X-ray findings. The most common radiological manifestation of skeletal hydatid disease is a lucent expansion lesion with cortical thinning.<sup>[6]</sup> In the spine, the appearance is of an irregular destruction of vertebral body and, sometimes, intervertebral space narrowing.<sup>[7]</sup> Computerized tomography (CT) scan can be useful not only in excluding the involvement of surrounding soft tissue, but also for planning surgery and assessing cyst viability. CT images can also recognize rupture of the endocyst by showing a folded detached endocyst, but the typical films are rarely found.<sup>[8]</sup> Magnetic resonance imaging (MRI) is the most useful technique for diagnosing hydatid cyst disease. It is so useful in depicting the extent of the disease, especially in soft tissue and spine as it gives a full image of the vertebral axis of the spinal canal.<sup>[9]</sup> Here, we present a primary recurrent hydatosis at the site of non-union humerus fracture. We have pointed out osseous hydatosis as one of the important differential diagnoses in destructive bone lesions and the necessity of its radical resection.

## CASE REPORT

A 39-year-old female presented with non-union of the left humerus fracture following trauma. The surgeon found the cystic lesion intraoperatively. Curettage with evacuation of the cyst was done and the fracture was fixed with dynamic compression plate. Analysis of resected specimen reconfirmed the diagnosis and the patient was referred for chemical therapy. After 2 years, the patient was re-admitted with the diagnosis of non-union humeral fracture.

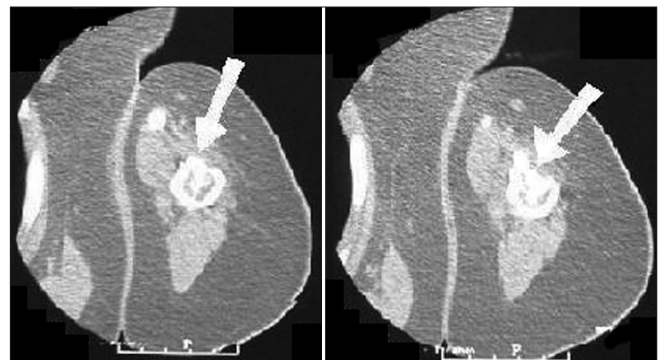
In physical examinations, motion of fractured site decreased and pain was eminent, but neuromuscular examination was normal. The laboratory findings were not specific and the routine serological tests were normal. Before operation, radiological plans including X-ray [Figure 1], CT scan [Figure 2], and MRI [Figure 3] showed a cystic lesion in the proximal part of the left humerus in the presence of non-union.

We administered albendazole (10 mg/kg/day) and praziquantel (40 mg/kg/day) for 2 weeks before surgery. During surgery, we found a highly viscous, yellowish liquid which was aspirated. After curettage, we cleaned the wound with a solution of H<sub>2</sub>O<sub>2</sub> and hypertonic saline solution to prevent recurrence of cysts. Because of massive resection, we used autographs from fibula and iliac bones, and the fracture was fixed with LCP [Figure 4].

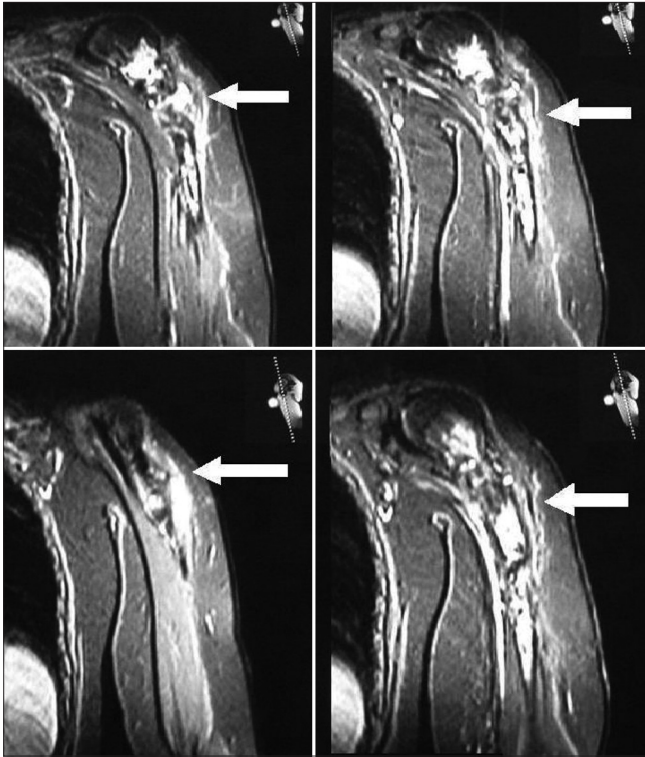
The histopathologic tests revealed the characteristic trilamellar hydatid cyst wall and scolices of *E. granulosus* scattered amidst fragments of bone and marrow. An ultrasound



**Figure 1:** Pre operation X-ray



**Figure 2:** Pre operation CT

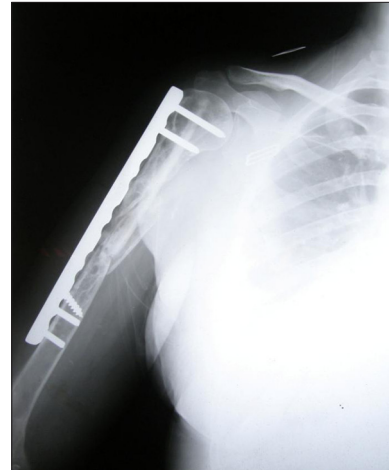


**Figure 3:** Pre operation MRI: Cystic lesion in the proximal part of the Humerus

and CT scans of the brain, breast, and abdomen were performed later, which revealed no cysts in these organs. After the operation, we performed serological investigations which showed negative results for complement fixation test (CFT) and latex agglutination test (LAT). The patient was discharged on albendazole (10 mg/kg/day) and praziquantel (40 mg/kg/day) to prevent recurrence of cysts. At the last follow-up (9 months), the patient was doing well. She was pain-free and completely asymptomatic. X-ray did not show any local recurrences.

## DISCUSSION

*Echinococcus* is usually found in the liver and lung. Incidence of bone echinococcosis is about 0.5–4% of the total reported cases, and is most commonly seen in the spine (from 50 to >60%), followed by the pelvic and hip (20%), femur and tibia (15%), humerus (15%), ribs and scapula (10%), phalanges (5%), and hand (5%).<sup>[5]</sup> Hydatid disease of the bone is often asymptomatic for a long duration and is usually detected only after a sudden fracture, secondary infection, or neurovascular lesion caused by compression.<sup>[10]</sup> Neurological



**Figure 4:** Post operation X-ray

problems such as paraparesis and paraplegia are common when the infection involves the spine.

A preoperative diagnosis is made in half of the patients.<sup>[6]</sup> A definitive preoperative diagnosis is often difficult without histologic examination. The Casoni intradermal test and CFT are not specific. Also, eosinophilia is not always seen. Immunological assays are now being used for detection of specific antibodies, circulating antigen, and immune complexes. However, there is no test that is highly sensitive and specific, particularly for cystic hydatid disease.<sup>[11]</sup> Enzyme-linked immunosorbent assay (ELISA) using crude hydatid cyst fluid has a high sensitivity but low specificity. Purified antigen or other techniques (immunoblot analysis, detection of Ig4 antibodies, immunoelectrophoresis, etc.) are used, in which the specificity improves, but the sensitivity gets worse. Therefore, it is suggested that at least two methods are combined in the serological diagnosis of cystic echinococcosis. Fine-needle aspiration may lead to dissemination of the infection and can be responsible for an anaphylactoid reaction.<sup>[12]</sup> The diagnosis is often made on the basis of the radiographic appearance of the lesions and the pathological changes in the bone. CT and MRI have also been added to the arsenal of investigations.<sup>[13]</sup> CT facilitates an earlier diagnosis and location of hydatid disease in bone.<sup>[14]</sup> MRI allows easier recognition of the cystic changes and their relationship to skeletal structures using the different imaging techniques. But these findings may be confused with those of other lesions.

At the therapeutic level, the behavior of skeletal lesions is comparable to that of a malignant

tumor. It is difficult to cure the disease, so the prognosis is generally poor. The only definitive treatment when bone is involved is a complete resection of the involved area. The resection must be complete with a wide healthy margin to prevent recurrences. However, surgical treatment is also associated with considerable mortality, morbidity, and recurrence rates of 70–80%.<sup>[15]</sup> Internal fixation can be improved by the use of acrylic cement or bone grafts. Medical treatment can be added to surgery in order to prevent or at least minimize the risk of dissemination. When surgical treatment is not possible, isolated medical treatment with mebendazole or albendazole can be used.<sup>[16]</sup> Adjuvant medical treatment may be given preoperatively if the diagnosis is known, and postoperatively in order to control the disease locally, avoid systemic spread, and prevent recurrence. The results of chemotherapy are controversial, and the available data about the effectiveness of these drugs in bone infections are still limited.<sup>[17]</sup>

Associated toxicity for mebendazole includes alopecia, hepatitis, and reduction of insulin in diabetes, and that for albendazole includes reversible neutropenia, hepatitis, and cholestatic jaundice. Praziquantel has recently been suggested as a drug to be given additionally once a week to increase the albendazole concentration and therapeutic effects when administered in combination.<sup>[18]</sup> Most authors recommend a combination of surgical resection, bone grafting, and postoperative treatment with mebendazole or albendazole and praziquantel.<sup>[19]</sup> The unusual aspect in this case was the presenting recurrent hydatosis at the site of non-union humerus fracture without involving the liver and lung.

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