A rare case report of femur hydatid disease with neglected pathological fracture

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Abstract

Osseous hydatid disease is a very rare parasitic infestation which comprise 1-2% of hydatidosis. Spine is the most common seat of osseous hydatidosis and long bone infestation is reported very less in literature. It mimics neoplasm and atypical infections. Hence it should be differentiated by investigations and strong suspicion in a predisposed individual. This is a rare case report of femur hydatid disease with pathological neglected fracture.

Keywords: rare case, femur hydatid, pathological fracture

Introduction

Hydatid disease is caused by a Cestode worm Techno Coccus Granulosus /multilocularis which completes its life cycle requiring a carnivorus and herbivorus mammals. The adult worm inhabitates the small intestine of dog and shreds ova in feces which enters the portal venous system of mammals when they eat the contaminated food. In portal circulation they reach liver and lung where they form cysts, which contain worm heads/scolices in their wall. When the dog feeds on infested mammal it enters dog small intestine and complete its life cycle. In Indian subcontinent prevalence of the disease correlates with human to cattle ratio.

Fig 1: Life cycle of Echinococcus granulosus
**Hydatid disease of bone**

It occurs when blood borne scolex passes the filters of the liver and lung entering the systemic circulation to reach a bone [1]. It forms a cyst but with lesser aggression in bones, which eventually weaken the cortex entering para-osseous compartments. Because of weakening of bony trabeculae due to constant pressure by cyst, cortex fails leading to a pathological fracture in many instances.

Patients present with pathological fracture without significant constitutional symptoms and may rarely present with pain and swelling over a bone. In severe infestations there are reports of amputations in the literature. There are many reports about recurrence after debridement with sinus tract formation. It is a disease compatible with life but difficult to eradicate.

Radiologically there will be a multilocular cystic lesion in the shaft of long bone which spreads through medullary cavity without sclerosis [2]. Peri-cyst formation is absent in bony Hydatid disease unlike liver or lung cysts [2]. MRI is used to differentiate it from metastasis/primary bone tumors.

**Case report**

A 65 year old gentleman with rural background presented to our out-patient department with pain and inability to walk from one month following slip and fall from standing height. Patient did not seek any medical attention and underwent local treatment with oil massage and manipulation but didn’t improve.

On examination he had shortened, externally rotated lower limb with generalised swelling of thigh. X-ray showed fractured midshaft of left femur with over-riding of sclerosed bone ends which were showing scalloping.

Blood investigations were not in favour of malignancy and osteoporosis. Patient was taken up for surgery and fracture fragments were distracted gradually with help of LRS post-operatively over 14 days.

Patient was on IV antibiotics post-operatively and culture & HPE reports on 4th post-op day showed hydatid cyst infected with gram-positive cocci in pairs and clusters (Staphylococci). Hence IV antibiotics continued for 2 weeks along with oral Albendazole 400mg HS for 4 weeks. There was sero-sanguinous discharge from fracture site wound for about a week. Fluid aspirated from fracture site didn’t show Hydatid scolices and discharge stopped with regular dressings. On follow-up after 3 months from surgery, there was sufficient callus formation at fracture site in X-ray and there was no discharge from the fracture site.
Discussion

Hydatid disease of bones is a rare disease with only reported cases of less than a hundred of femur. It is a disabling bone disease with dramatically reduced quality of life due to less available chemotherapeutic agents and no definitive surgical management other than debridement and fixation. As bone is relatively less vascular compared to other soft tissues chemotherapeutic agents should be given in higher doses for long duration. Albendazole at 10mg/kg body weight has been advised for 2 months, but alone it can’t cure the bone disease without proper debridement and fixation. Until recently treatment of osseous hydatid disease has been entirely surgical, the aims being removal of the cyst and surrounding bone, replacement of bone defects with bone grafts or a prosthesis, avoidance of secondary infection, and prevention of recurrence. Unfortunately these goals are rarely achieved completely in this relentless disease. Booz (1972), Hooper and McLean (1977) and Duran et al. (1978) have advocated thorough mechanical curettage to remove macroscopic cysts and “chemical” sterilisation of the scolices using formalin, 0.5% silver nitrate or hypertonic saline. Any major defect should be filled with autogenous bone graft. Most “scolidal” agents do not kill all microscopic daughter cysts, and therefore recurrence is likely, and surgery is often only palliative. Effective chemotherapy would therefore be of great benefit. Following surgery, Albendazole (10 mg/kg/day) for six cycles of 25 days each is needed for preventing recurrence and it is essential to monitor the liver function during treatment for hepatotoxicity. Albendazole will not improve the strength of the weakened bones, therefore surgical excision should be combined with bone grafting or prosthetic replacement, where this is indicated and safe. The variable success of Mebendazole was thought to be due to the insolubility of Mebendazole with its consequent poor absorption leading to low concentrations in serum and cysts.

Conclusion

Bone hydatid disease is notorious for recurrence with high chances of infective non-union. It should be suspected when a patient presents with lytic lesion and should be considered as an osseous tumor and imaging should be done accordingly. CT is a better imaging modality to define margins. Unlike tumors, needle biopsy is contra-indicated in hydatid disease as anaphylaxis and extra-osseous spread are the devastating complications. It should be excised along with normal margins and cavity should be irrigated with scolicidal agents thoroughly. As recurrence is very common bone can be stabilised with external fixator and patient can be given albendazole for a minimum of 2 months with antibiotic coverage if secondarily infected depending on culture reports. In second stage if swabs are negative for parasite, cavity can be managed with either bone graft and PMMA with intramedullary nail fixation of femur.

References