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Intramuscular hydatid cyst: Rare cases from Nepal

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Abstract

We present two cases of slowly growing painless lumps in the upper and lower limb. The diagnosis was made preoperatively based on physical examination, ultrasonography, and FNAC. Hydatid cyst may present with swelling in soft tissue, especially in the endemic region. Hydatid cyst should be considered in the differential diagnosis of swelling in the limb along with sarcoma. We report two rare cases of a hydatid cyst from a rural part of Nepal.

Keywords

Echinococcosis; Hydatid cyst; Intramuscular hydatid.

Introduction

Echinococcosis is caused by the tapeworm genus *Echinococcus* which normally lives in the intestine of carnivores. Four species of Echinococcus are infective to humans i.e. *E. granulosus*, *E. multilolocularis*, *E. vogeli*, and *E. oligarthus* [1]. Adult worm of *Echinococcus granulosus* inhabiting in the intestine of the definitive host (usually dogs, or related species) produce thousands of eggs per day which are expelled in the stool and released in the environment. The eggs become an infective source to the intermediate host (sheep, goats, camels, horses, cattle, etc.) while humans are the accidental host. Following egg ingestion, oncosphere larva hatches from an egg and penetrates the intestinal mucosa entering the bloodstream or lymphatic system. The oncosphere larva migrates to the liver or visceral organs through the portal system. Within a few days, it develops into multiple layered hydatid cysts [2]. It is more prevalent in Nepal as agriculture and livestock breeding is common practice here.

Echinococcosis commonly affects the liver and lung comprising around 55-70% and 18-35% respectively [3]. However, it rarely infects muscle and should be considered in the differential diagnosis of muscle swelling along with abscess, sarcoma, or hematoma.

Case 1

A 17-years-old girl was presented swelling in the right upper thigh with slow growth. The swelling was progressively increasing in size and was painful. Clinical examination revealed intramuscular swelling occupying the lateral region of the right thigh which was immobile in any plane. It measured 16 X 10 cm. With the differential diagnosis of intramuscular abscess, sarcoma, hematoma, and hydatid cyst, investigations were sent. Total blood count revealed a slight increase in eosinophil count. Ultrasound examination revealed multiple well-defined rounded cystic structures with honeycomb appearance in muscle planes of the right lateral thigh, suggestive of hydatid cyst. The serology for hydatid cyst was not available thus we were unable to confirm the diagnosis with the investigation. Chest X-ray and abdominal ultrasound were done to rule out liver and lung involvement. Excision of the cyst was planned under local anesthesia. An elliptical incision was made over the protrusion on the thigh. Tensor fasciae lata muscle and vastus lateralis muscles were exposed, which was densely adherent to the surrounding muscle, and the mass was removed *en bloc*, as a white cystic ovoid mass. The wound was irrigated with povidone-iodine solution and the wound was closed.

Examination of the cyst had multiple small fluid-filled daughter cysts. Pathological examination of cyst confirmed our preoperative diagnosis of Hydatid cyst. The postoperative course was uneventful. On the second postoperative day, the patient was discharged on Albendazole 400 mg for six weeks. After 3 months, the follow-up concluded she was disease-free.

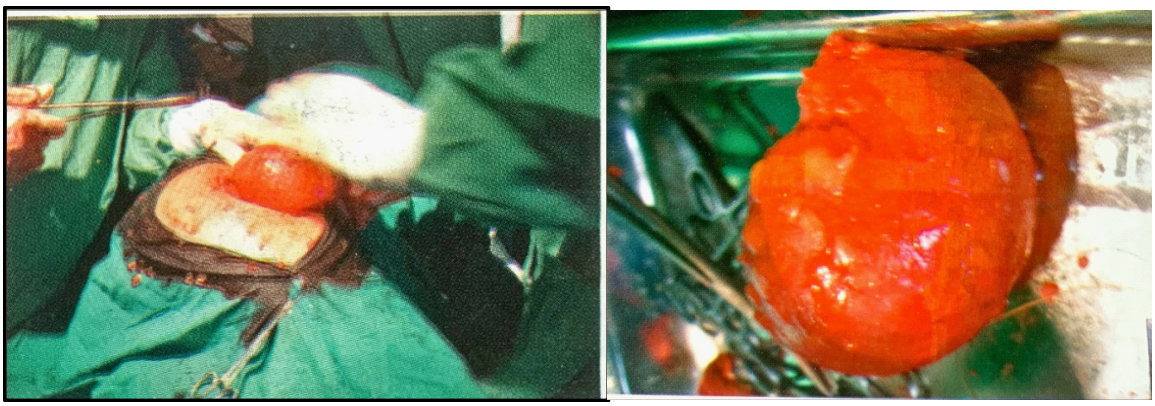

Figure 1: Intra-operative finding of the Cyst. Figure 2. Cyst after being removed.

Case 2

A 30-year male was presented with swelling in the left arm which had progressively increased in size over 5 months. On examination, the swelling was 3 X 4 cm in size arising from the intramuscular plane. The swelling was mildly tender. To rule out sarcoma and confirm the diagnosis ultrasonography and fine-needle aspiration cytology (FNAC) were sent. Ultrasound revealed the thick wall cystic lesion with few septations and hyperechoic nodular focus in the left arm likely hydatid cyst, while the FNAC report showed sheets of mixed inflammatory cells and degenerated cells. With the diagnostic dilemma, the patient was planned for excision of mass under local anesthesia. Biceps muscle was exposed and mass was completely removed. It was a white mass filled with fluid and pus with a daughter cyst. Diagnosis of infected hydatid cyst was made. Chest X-ray and Ultrasonography of the abdomen were done to rule out the concomitant lesion. The patient was discharged on Albendazole for 6 weeks. He had progressed well over 3 months.

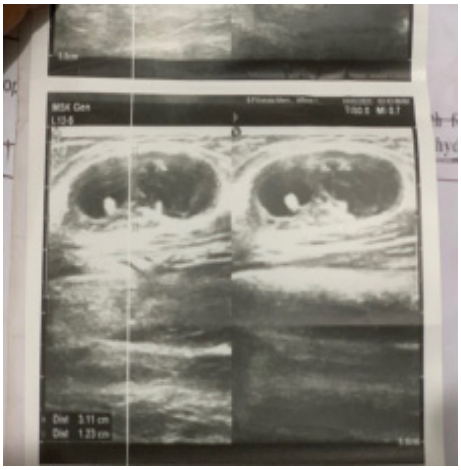

Figure 2: USG findings.

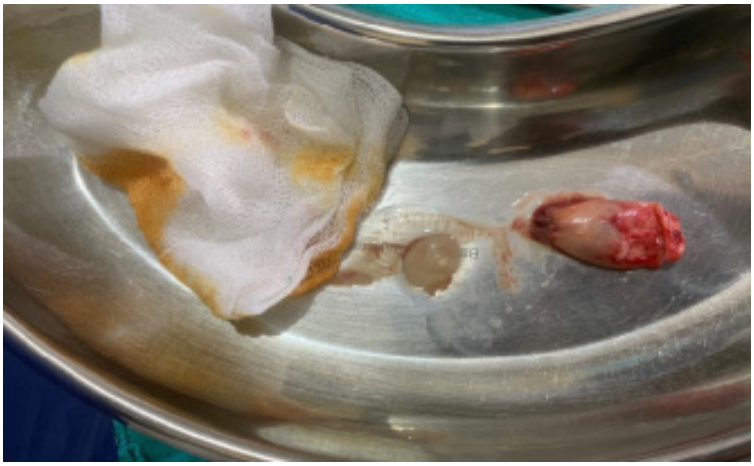

Figure 3: Cyst after the excision..

Discussion

Echinococcus granulosus cause echinococcosis which can affect liver, lungs, spleen, rarely kidney, heart, bone, eye and muscle. Fever and hypersensitivity reaction are the main manifestation of cyst rupture. Lethal anaphylaxis can occur with percutaneous treatment of cystic Echinococcus [4].

The reason for intramuscular hydatid cyst has been postulated by two theories. First is by direct implantation of the cyst in the muscle through dog bites and the other is by transport through systemic circulation from the intestine to skeletal muscle [3]. Ingestion of the ova with the development of hydatid cyst in the liver and lungs is labeled as a primary hydatid cyst. In secondary hydatid cyst, there is the proliferation of larva at the primary site later spreading to the other site. It tends to grow in those muscles with increased vascularity and decreased activity [5]. In our case, it can be due to traumatic events leading to implantation of the cyst in the muscle labeling as primary hydatid cyst but it still remains unsure.

The patient usually presents with a slow-growing mass in the muscle with or without pain during movement. Cyst increases by 1 to 5 cm per year in diameter [6]. It can be found on any site via primary inoculation or secondary spread. The liver is affected in about two-thirds of patients, lung in about 25% of cases [7]. In our case, patients presented with slow growing mass without any involvement of liver or lungs in both of the cases.

Ultrasonography, MRI, CT scan, and Serology are a few diagnostic tools. Based on USG, the cyst has been classified into 5 different groups based on Gharbi criteria. Type 1 is pure cystic, Type 2 is a cyst with germinative membrane, Type 3 is a multi-cystic lesion with septate, Type 4 is a degenerative cyst with the false solid view, and Type 5 is a false solid cyst with calcification. USG finding of daughter cyst separated membranes, and double line sign, confirm the diagnosis [8]. MRI or CT scans are more valuable tests in detecting muscle hydatidosis. It is advised to avoid biopsy or aspiration to prevent spread [3]. ELISA is a more sensitive and specific test for detecting *E. multilocularis* than *E. granulosus* with sensitivity and specificity of 95 to 100% [7].

Excision is the primary mode of treatment. However, due to the risk of recurrence, perioperative

treatment of albendazole or mebendazole is beneficial. Perioperative treatment with the drugs inactivates the protoscolices thus, it reduces the risk of recurrence. The optimum duration of the treatment is uncertain but in general, it should be initiated 4 days before surgery [1]. Since the relapse is high, long-term follow-up of the patient is advised.

Conclusion

Intramuscular hydatid cyst is rare but it should be considered in differential diagnosis of slow growing mass in the extremities.

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