

Primary intramuscular hydatid cyst over the back: A rare presentation of echinococcosis

R Sonarkar, A R Raikwar

From Department of Surgery, NKP Salve Institute of Medical Sciences, Nagpur, Maharashtra, India

Correspondence to: Dr. A R Raikwar, NKP Salve Institute of Medical Sciences, Digdoh, Hingna Road, Raikwar Hospital, Jayastambh Square, Paratwada, Amravati, Nagpur, Maharashtra, India. Phone: +91-9422917873. E-mail: ankurhallow@yahoo.co.in

Received - 30 January 2018

Initial Review - 28 February 2018

Published Online - 16 April 2018

ABSTRACT

Hydatid disease is often manifested as a slowly growing cystic mass, with hepatic and pulmonary involvement being most common. Unusual sites of involvement frequently cause diagnostic problems, and hence, delay in diagnosis and many serious complications. In endemic areas, cystic lesions anywhere in the body, in appropriate patient group, should be evaluated for hydatid disease. We report an unusual case of primary hydatidosis of the back muscle, in which a wide excision was performed without causing any damage to the cyst wall. Injudicious approach in the management of these rare presentations may be the root cause of severe anaphylactic shock and systemic dissemination. Intramuscular hydatid cysts grow gradually and may mimic a soft tissue tumor; thus, the diagnosis of soft-tissue hydatid cysts needs a high index of suspicion.

Key words: *Echinococcus granulosus*, Hydatid cyst, Intramuscular

Hydatid disease is endemic in most cattle rearing areas, particularly Australia, New Zealand, Mediterranean countries, India, Africa, and South America [1,2]. It is often manifested by a slowly growing cystic mass, with hepatic and pulmonary involvement being most common [3]. Unusual sites of involvement frequently cause diagnostic problems, and hence, delay in diagnosis and many serious complications. Primary skeletal muscle involvement is very rare [4,5]. We present a rare case of hydatid disease of muscles in the middle-aged male, who presented to the general surgery clinic.

CASE REPORT

A 28-year-old male presented with chief complaints of swelling over back region for the past 2 years. There were no gastrointestinal or urinary symptoms or any history of fever. There was the previous history of surgery over the same site 4 years back of which no documentation was available. There were no neurological symptoms.

General physical examination of the patient was unremarkable. Local examination of swelling on the back region revealed a cystic swelling of size 4×4 cm approx., non-tender, immobile, non-ballotable, fluctuation present over the swelling in the right 10th, 11th, and 12th paravertebral region without any deformity of the adjacent spine. The swelling do not increase in size on coughing or by Valsalva maneuver.

All the blood investigations were within normal limit. Ultrasonography (USG) of local region was suggestive of cystic swelling with internal echo suggestive of an abscess. The X-ray

of spine did not reveal any abnormality. Fine needle aspiration cytology was also performed in view of primary diagnosis as a cold abscess in which, clear fluid was drained. Further, investigation (ESR) was done which comes out to be Negative. We were not able to conduct serological procedure/imaging due to the financial issues. Provisional diagnosis was kept as a lymphatic cyst. The patient was subjected to surgery after a proper workup. The cyst was excised en masse by posterior approach. About 4×4 cm approx., multilocular cyst was found within paraspinal muscles of the back. The operative area was packed with sponges soaked in 1% cetrimide for 7min (Fig. 1). The excised specimen on opening showed the characteristic laminated membranes (Fig. 2). The sample was sent for histopathology which was suggestive of infection of hydatid cyst. The patient responded to treatment well and was followed up for 1 year. Even after 1 year, the patient was well with no fresh complaints.

DISCUSSION

Hydatid disease or Echinococcosis (or hydatidosis) is a parasitic disease caused by infection with larva (metacestode) of the cestode *Echinococcus*. Four species of the genus *Echinococcus* are known to cause infection in humans: *Echinococcus granulosus* (cystic hydatid disease), *Echinococcus multilocularis* (alveolar hydatid disease), *Echinococcus vogeli*, and *Echinococcus oligarthus* (both causing polycystic hydatid disease) [6]. *E. granulosus* is a 5 mm long tapeworm and requires two hosts for completion of its life cycle. The carnivores (dogs, wolves, etc.) are the definitive hosts, harboring mature tapeworms in their intestines. The herbivores act

as intermediate hosts who ingest the eggs from which the embryos escape, penetrate the intestinal mucosa, enter the portal circulation, and are then trapped in liver [7], and a few may escape into the systemic circulation. The life cycle is completed when the carnivores eat the infected offal of the intermediate host. Human beings are the accidental hosts and dead end in the life cycle of tapeworm.

Hydatid cyst consists of a fluid-filled cavity lined by three layers: Pericyst (host reaction), ectocyst (laminated membrane),



Figure 1: Hydatid cyst over the back infrascapular region

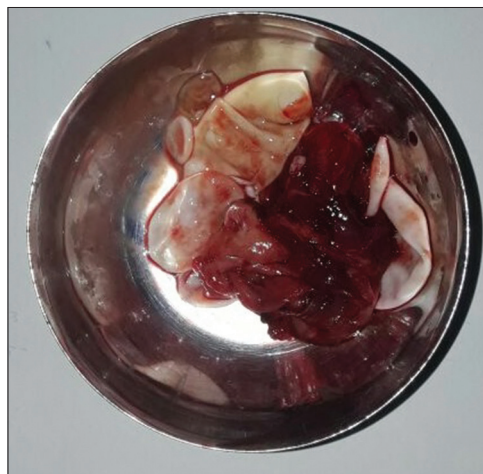


Figure 2: Laminated membrane of Hydatid cyst

and endocyst (germinal layer) which gives rise to the laminated membrane and secretes hydatid fluid. The hydatid cysts may potentially inhabit any part of the body. The liver (59–75%) and lungs (27%) are most commonly involved organs, with other organs such as kidney (3%), bone (1–4%), brain (1–2%), heart, spleen, pancreas, breast, and muscles being very rarely affected [8]. Most patients with muscle hydatid will have associated hepatic cysts [4]. There have been a few case reports citing hydatid cysts in adductor muscles of the thigh [9], pectoralis major muscle [10], gluteal and neck muscles [4], and rectus and spinal muscles [5]. Our patient was diagnosed postoperatively as, primary hydatid cyst of the paraspinal muscles of the back (without any evidence of Echinococcosis in other anatomic sites of body) which has been reported only once in the medical

literature [11]. The echinococcal cysts grow very slowly and generally remain asymptomatic. 5–20 years elapse before cysts enlarge sufficiently to become symptomatic [12].

Clinical manifestation depends on the anatomic site involved and usually manifest as a slowly growing palpable mass. Symptoms due to compression of adjacent structures are not uncommon [3] anaphylactic reaction due to cyst rupture and secondary infections are the most common complications [11]. Protean manifestations are not unknown with epilepsy and raised intracranial pressure (cerebral cysts) [13], choledochal cyst [14], pulmonary hypertension [15], and portal hypertension [15] due to hydatid cysts being reported in literature. The case being reported by us presented with low backache and a soft left lumbar swelling.

Serology and imaging are the main tools for establishing diagnosis. Various serological tests detect specific serum antibodies or circulating antigens and include ELISA for Echinococcus IgG (sensitivity 95% and specificity 94%) [11] and immunoelectrophoresis, hemagglutination tests. Plain X-ray, USG, computed tomography (CT), and magnetic resonance imaging (MRI) are the various imaging modalities utilized to visualize the cyst. The characteristic features of the hydatid cyst on CT and MRI help in establishing the diagnosis with high accuracy [11,12]. The case under discussion had a Type II cyst (Lewall) [13].

Therapy for extrahepatic Echinococcus disease is dependent on size, location, and manifestation of the cyst and the health status of the patient. Asymptomatic small cysts can be managed with anthelmintic drugs. Surgery is the mainstay of treatment for large, symptomatic cysts [14]. Radical or conservative approach can be adopted depending on the clinical situation. Total cystectomy carries least risk of recurrence but is associated with high morbidity. Simple deroofing and enucleation of the cyst are appropriate for intraperitoneal cysts firmly embedded in the viscera. Care is taken to prevent spillage. Various methods are available to manage the residual cavity [14].

Puncture-aspiration-injection-aspiration (PAIR) refers to an ultrasound-guided technique consisting of puncture and evacuation of the contents of the hydatid cyst; injection of scolecidal agents such as 95% ethanol, and reaspiration of the contents of the cyst. Although puncture of a hydatid cyst has long been discouraged because of potential complications, such as anaphylactic shock or risk of iatrogenic spreading, there is expanding literature suggesting that PAIR is effective for treatment of primary uncomplicated hepatic cysts [15] However, this technique has been used in a recent study as a modality for muscular hydatid cyst also. We performed total cystectomy in our patient and obliterated the cavity by caption age.

CONCLUSION

In endemic areas, cystic lesions anywhere in the body, in appropriate patient group, should be evaluated for hydatid disease. The present serological and imaging modalities help in establishing the diagnosis with high accuracy. Treatment varies from medical management to radical surgical excision.

REFERENCES

- Pedrosa I, Saiz A, Arrazola J, Ferreirós J, Pedrosa CS. Hydatid disease: Radiologic and pathologic features and complications. *Radiographics* 2000;20:795-817.
- Jahed AK, Fardin R, Ferzad A, Bakshandeh K. Clinical Echinococcosis. *Ann Surg* 1975;182:541-6.
- Tsaroucha AK, Polychronidis AC, Lyrantzopoulos N, Pitiakoudis MS, J Karayiannakis A, Manolas KJ, *et al.* Hydatid disease of the abdomen and other locations. *World J Surg* 2005;29:1161-5.
- Ghatashah M, Etaiwi M. Skeletal muscle Echinococcosis: A rare manifestation of Echinococcal disease. *Ann Saudi Med* 2002;22:245-6.
- De Cosare V. Unusual localization of hydatid disease: An 18 year experience. *G Chir* 1994;15:83-6.
- Khuroo MS. Hydatid disease: Current status and recent advances. *Ann Saudi Med* 2002;22:56-64.
- Sayek I, Yalin R, Sanec Y. Surgical treatment of hydatid disease of the liver. *Arch Surg* 1980;115:847-50.
- Yuksel M, Demirpolat G, Sever A, Bakaris S, Bulbuloglu E, Elmas N, *et al.* Hydatid disease involving some rare locations in the body: A pictorial essay. *Korean J Radiol* 2007;8:531-40.
- Milanese A, Camana GP, Carbone P. Echinococcus cyst in the pelvic cavity: Presentation of case. *Minnerva Urol Nephrol* 1991;43:111-4.
- Abdel-Khaliq RA, Othman Y. Hydatid cyst of pectoralis major muscle. Case report and note on surgical management of muscle echinococcosis. *Acta Chir Scand* 1986;152:469-71.
- Balik AA, Celabi F, Basglu M, Oren D, Yildirgan I, Atamanalp SS. Intra-abdominal extrahepatic Echinococcosis. *Surg Today* 2001;31:881-4.
- Parray FQ, Ahmad SZ, Sherwani AY, Chowdri NA, Wani KA. Primary paraspinal hydatid cyst: A rare presentation of Echinococcosis: A Case Report. *Int J Surg* 2010;8:404-6.
- Hageman G, Cottstein B, Witte OW. Isolated *Echinococcus granulosus* hydatid cyst in the CNS with severe reaction to treatment. *Neurology* 1999;52:1100-1.
- Utpal DC, Basu M. Hydatid cyst of common bile duct mimicking type I choledochal cyst. *J Indian Assoc Pediatr Surg* 2007;12:83-4.
- Erdo-Gan Y, Turay UY, Deniz PP, Biber C, Ergun P, Çiftçi B, *et al.* A case of pulmonary hypertension due to chronic pulmonary hydatid cyst embolism. *Turk Respir J* 2007;8:105-7.

Funding: None; Conflict of Interest: None Stated.

How to cite this article: Sonarkar R, Raikwar AR. Primary intramuscular hydatid cyst over the back: A rare presentation of echinococcosis. *Indian J Case Reports*. 2018;4(2):124-126.