IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 13, Issue 9 Ver. VIII (Sep. 2014), PP 23-27
www.iosrjournals.org

Primary Intramuscular Hydatid Cyst within Brachialis Muscle: A Case Report and Review of Literature

Dr Nirpex Tyagi[1], Dr Nitin Garg[2]
[1,2]*(Department Of General Surgery,People's College Of Medical Sciences & Research Centre,India)*

Abstract: *Hydatid disease is a cyclo-zoonotic parasitic infection mainly affecting liver and lung. Primary hydatid cyst in the muscle is very rare and is usually secondary, resulting either from the spread of cysts spontaneously or after surgery.*
Here, we report a case of primary hydatid cyst within brachialis muscle, with emphasis on the fact that it should be included in the differential diagnosis of peripheral cystic mass to avoid invasive investigations like FNAC. It results in better patient management because leakage of the cyst contents may not only result in recurrence and dissemination, but may also cause anaphylaxis and subsequent surgical resection difficult.
Key words: *Hydatid Cyst; Echinococcosis; Intramuscular*

I. Introduction

Hydatid disease or Echinococcosis has a worldwide distribution and causes health problem in endemic countries like Australia, Argentina, Africa, Middle East and the Mediterranean region. The disease is also endemic in India with high prevalence in Kashmir, Andhra Pradesh, Tamil Nadu and Central India [1-2].

Being a zoonotic disease, it is caused by the larval tapeworm of genus Echinococcus granulosus, Echinococcus multilocularis and Echinococcus oligarthus. However, most human cases occur due to infection of E.granulosus [3].

Life cycle (Figure 1) :- Adult tapeworms live in the small intestine of carnivore like dog that is a definitive host. The eggs pass in the faeces and cause environmental contamination. The main intermediate hosts are herbivores, usually sheep, but occasionally humans. When eggs are swallowed, oncospheres released in the gastrointestinal tract penetrate the intestinal mucosa and are disseminated via venous and lymphatic channels. They disseminate to various organs, where they implant and develop hydatid cysts. In humans, liver is the most common site of cyst development (60%), followed by lungs (20%)

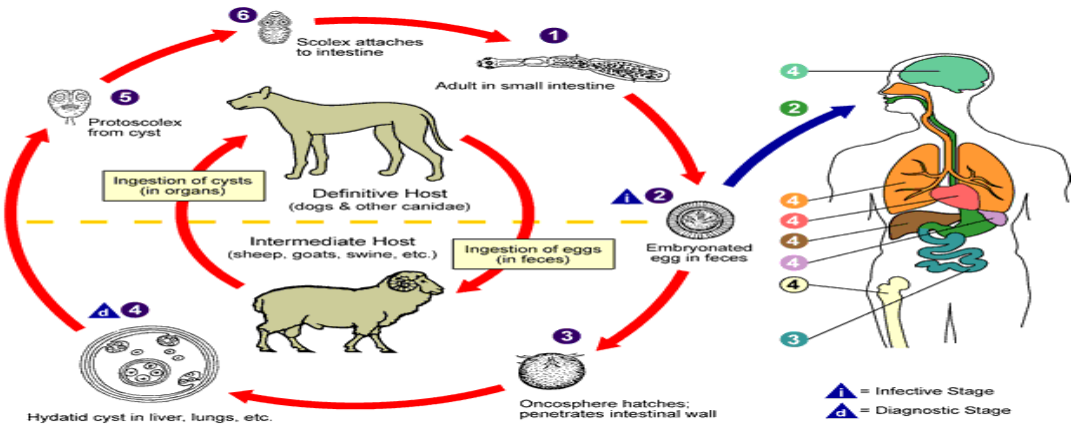

Figure-1:- Life cycle of E.granulosus

By passing the liver and pulmonary capillaries, embryo may enter in greater circulation and may reach to other organs such as kidney, spleen, brain, heart, bone, orbit or muscle and develop there. So affection to organs other than liver and lung can be primary or secondary following liver and lung involvement.

Localisation at unusual sites in body can have an atypical presentation, which can pose a diagnostic challenge and mislead the treatment including inadvertent use of interventional investigations that may complicate the outcome and prognosis.

Here, our aim is to report a case of primary hydatid cyst in brachialis muscle because of its rare presentation, to emphasize the fact that hydatid cyst should remain in differential diagnosis of cystic lesions in unusual locations and to avoid diagnostic fine-needle biopsy which may result in leakage of cyst contents.

II. Case Report

A 16 year old female presented in the Department of Surgery of our hospital with chief complaints of swelling over left arm since 6 months and pain since 15 days. Swelling was gradually progressive. There was no H/o fever, cough, abdominal pain, or trauma. No H/o any associated comorbidities neither similar episode in the past. She also had H/o close confinement with cattles and sheep.

On local examination a single swelling of size aprox. 4 x 3-cm over medial aspect of left midarm, soft & nontender, fluctuant, nontransilluminant, and slightly mobile in both vertical and horizontal dimension with smooth surface and borders. Skin over the swelling was normal. There was no erythema, ecchymosis, increased warmth, or regional lymphadenopathy (**Figure 2**). The elbow and shoulder had full range of motion. Distal pulses were equal in the both upper extremities. Other general and systemic examination was within normal limit.

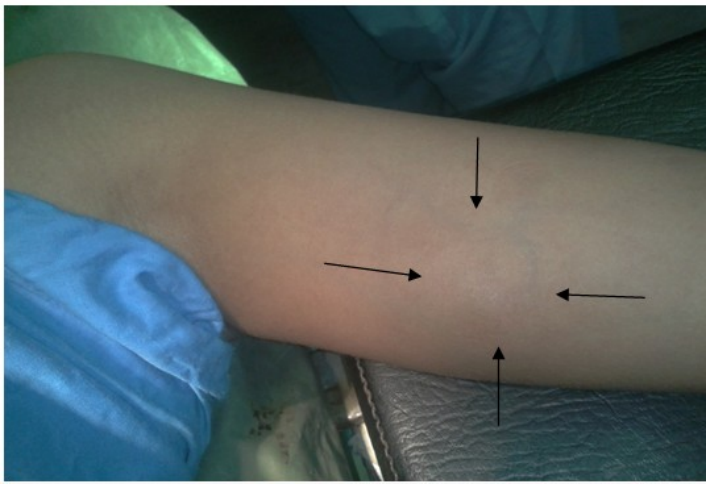

Figure-2:- Swelling over left arm

Complete laboratory data was normal. Indirect hemagglutination test for E.granulosus was positive. No calcification seen on plain radiograph. Ultrasonography (USG) revealed a hypoechoic lesion of size 4x1.9 cm in left arm seen involving anterior fibres of left brachialis muscle extending upto subcutaneous soft tissue plane. In visualised swelling a small anechoic cystic lesion of size 7x8 mm seen having a small calcified mural nodule suggestive of possibility of musclar hydatid cyst. (**Figure 3**).

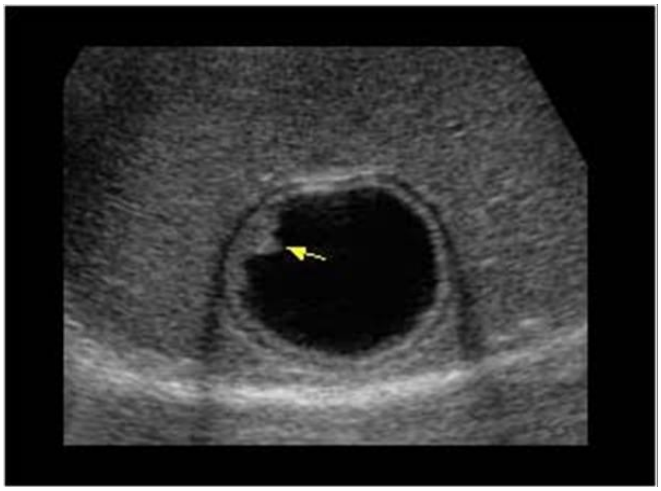

Figure-3:-USG showing cystic lesion in left arm with small

USG Abdomen and X-ray Chest were normal indicating that this was a case of solitary primary hydatid cyst of brachialis muscle of left arm. The patient was started on oral Albendazole 1 week prior to surgery and planned for surgical excision.

Longitudinal incision was made directly over the swelling and tissues dissected uptil the cyst wall. The surrounding field was packed off to prevent contamination. We carefully started to dissect the cyst from the surrounding muscle outside the pericyst. But it ruptured inadvertently at one place revealing the daughter cyst (**Figure 4,5**). Nevertheless the cyst wall was excised in whole. The wound was irrigated with hypertonic saline solution (scolicidal agent) and a suction drain kept.

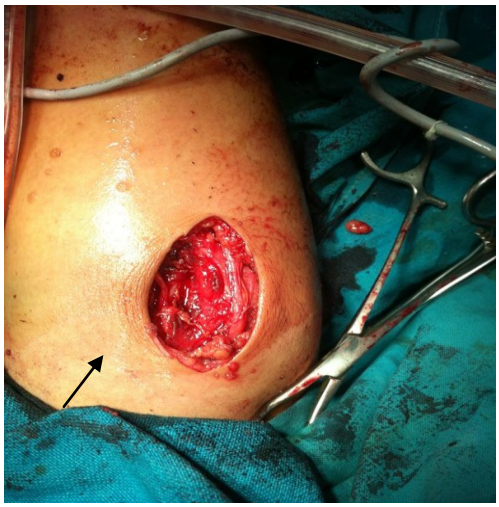

Figure-4:- Postoperative wound

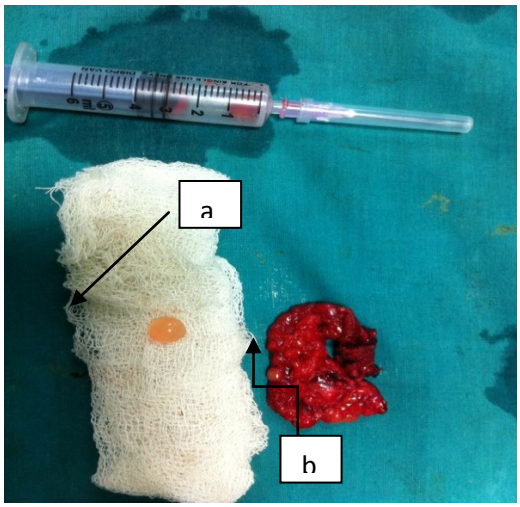

Figure-5:- Excised specimen (a.daughter cyst, b.cyst wall)

Post-operative period was uneventful, drain removed after 48 hrs and patient discharged on a course of oral Albendazole for 28 days. Histopathology confirmed the diagnosis:-

- **Gross:**– Irregular tissue measuring 4x2x2cm with a translucent cyst measuring 0.7cm diameter.
- **Microscopy:-** Cyst wall consisting of lamellar eosinophillic structure with loss of germinal lining epithelium. Inside cyst, scolices are seen in the form of invaginated tubules & cuticular lining. Surrounding fibro-connective tissue shows acute inflammatory infiltrate (**Figure 6**).

Figure-6:- Microscopic appearance . (Inside cyst, scolices are seen in the form of invaginated tubules & cuticular lining. Surrounding fibro connective tissue shows acute inflammatory infiltrate)

The case has been followed up for 1 year and there is no recurrence till now, either local or systemic.

III. Discussion

Primary hydatidosis of skeletal muscle is rare and incidence is not clear. Soft-tissue hydatid cysts occur in 2.3% of cases reported from endemic areas [4] and the prevalence is only 0.5-4.7% [5-6]. It is rare because cyst uses oxygen for growth while muscles usually contain lactic acid, and growth of cysts is further hindered by the muscle's contractility. Hydatid cysts tend to grow around the muscles of the neck, trunk and roots of the limbs, perhaps because there is greater vascularisation and less muscular activity in these regions. They develop very slowly and act as space occupying lesions, producing symptoms related to pressure on the surrounding tissues [7].

Diagnosis of Echinococcosis should be considered when slow growing soft tissue masses are present in the patients coming from rural areas especially in endemic countries.

Because an intramuscular hydatid cyst may mimic any soft tissue tumour, preoperative clinical diagnosis of intramuscular hydatid disease is difficult [8] clinically and radiologically. Before surgical excision or biopsy, diagnosis of Echinococcosis should be excluded to avoid leakage of cyst contents [7]. The differential diagnosis in these cases must include malignant soft tissue tumours such as myxoid liposarcoma, soft tissue abcesses, and chronic hematoma [9].

Diagnosis of echinococcosis is available through various serological tests, however they may give negative results because of encapsulation of the lesion; immunoelectrophoresis is the most specific method [7]. USG is useful in diagnosis, showing the size, localization and type of the cyst. The sensitivity of USG is 95%, and if vesicular fibrils are present, the sensitivity of US increases to 100%. WHO has supported development of internationally standardized classification of ultrasonographic images in cystic Echinococcosis. For management purposes the cysts are divided into three groups – active, transitional, and inactive. The classification enables worldwide comparison of cyst types and recommended treatment for different cyst types [10].

CT scan should be performed in suspicious cases or in order to determine the technique of surgery with demonstration of the relationship to adjacent organs. Inverse crescent sign, signet ring sign, high CT density (40-160HU) and thick wall are recognized [11]. Demonstration of air bubbles within the cyst together with ring enhancement is strong indicator of infected hydatid cyst [12].

MRI is best for clear identification of involved structures and for surgical planning. It is also an effective means of making a differential diagnosis and the pathognomonic 'water-lily' sign (type-2 hydatid cyst) can be detected by MRI [13].

In this case, we didn't do FNAC as we routinely do for soft tissue swellings. Suspicion of intramuscular hydatid cyst was raised on clinical examination and later diagnosis supported by ultrasound and serology. CT or MRI were not done due to superficial nature of the swelling and no proximity to important neuro-vascular structures.

To reduce the risk of dissemination during surgery and to prevent recurrence, we took some measures like preoperative adjuvant chemotherapy with antihelminthic agent-Albendazole and continuing it post-operatively, isolating the cyst during dissection and irrigation with scolicidal agent like hypertonic saline. Cyst rupture did happen inadvertantly in our case but in one year follow-up no local or systemic recurrence has been noted, perhaps due to above precautionary measures. Many studies have supported the use of pre and postoperative course of Albendazole in reducing the risk of dissemination and recurrence [14].

Nonetheless, the cyst rupture should be avoided as far as possible because it releases viable scoleces, which may enter the circulation, disseminate to distant organs and reproduce asexually to form additional cysts. Because the fluid of cyst contains a highly antigenic protein, leakage of cyst contents may also cause anaphylactic shock [15].

IV. Conclusion

In endemic areas, any cystic enlargement of soft tissue should raise the suspicion of hydatid disease and serologic tests and USG should be performed before any invasive procedure that may adversely affect the outcome.

References

[1]. Nepalia S,Joshi A,Shende A et al. Management of echinococcosis. J assoc Physicians India 2006;54:452-456.
[2]. Akhter J,Khanam N,Rao S.clinico epidemiological profile of hydatid diseases in central India,a retrospective and prospective study. Int J Biol Med Res 2011;2:603-606.
[3]. Arora V, Nijjar IS, Gill KS, Singh G. Primary hydatid cyst of muscle-A rare site. Indian J Radiol Imaging.2006;16:239–41.
[4]. Muñoz Sánchez JA, Conthe Gutiérrez P, Arnalich Fernández F, Fernández A, García Seoane J, Ortiz Vázquez J, et al. The incidence of hydatidic disease in a general hospital. I. Epidemiological analysis of 1056 cases. Med Clin (Barc) 1982;78:421–6. [PubMed]
[5]. Merkle EM, Schulte M, Vogel J, Tomczak R, Rieber A, Kern P, et al. Musculoskeletal involvement in cystic echinococcosis: report of eight cases and review of the literature. Am J Roentgenol 1997;168:1531-4.

[6]. Tacal T, Altinok D, Yildiz YT, Altinok G. Coexistence of intramuscular hydatid cyst and tape worm. A J Rotenjol 2000;174:575-6.
[7]. Kazakos CJ, Galanis VG, Verettas DA, Polychronidis A, Simopoulos C. Primary hydatid disease in femoral muscles. J Int Med Res 2005;33(6):703-6.
[8]. Ates M, Karakaplan M. Hydatid cyst in the biceps and gluteus muscles: case report. Surg Infect (Larchmt) 2007;8(4):475-8.
[9]. Said M, Babba H, Golli M, Ganouni A. Can We Really Have Coexistence of Hydatid Cyst and Tapeworm in Humans? Am J Roentgenol 2001;176:252-3
[10]. Richter J, Hatz C, Haussinger D. Ultrasound in tropical and parasitic diseases. Lancet 2003;362:900-02.
[11]. WHO-IGWE (WHO informal working group on Echinococcosis). Acta Trop 2003;85:253-61.
[12]. Koul PA,Koul AN,Wahid A et al.CT in pulmonary hydatid disease: unusual appearances. Chest 2000;118:1645-7
[13]. Durakbasa MO, Kose O, Islam NC, Kilicoglu G. A primary hydatid cyst of the gracilis: a case report. J Orthop Surg (Hong Kong) 2007;15(1):118-20.
[14]. Duvqulu F, Karaoqlu S, Erdoqan N, Yildiz O. Primary hydatid cyst of the thigh: a case report of unusual localization. Turk J Pediatr 2006; 48: 256-9.
[15]. Taşpınar V, Erdem D, Erk G, Baydar M, Göğüş N. [Intraoperative anaphylaxis caused by a hydatid cyst (case report)]. TurkiyeKlinikleri J Anest Reanim 2004;2(1):36-40.