

## Case Report

# Primary hydatid cyst of muscle-a rare cause of mass in the shoulder: a case report

Gaurav Chhaya<sup>1</sup>, Deval K. Shah<sup>2</sup>, Hiren Prajapati<sup>3</sup>, Mayank Rajawat<sup>3\*</sup>

<sup>1</sup>Shivam Medicare Clinic, Ahmedabad, Gujarat, India

<sup>2</sup>Swastik Surgical Hospital and Dental Clinic, Ahmedabad, Gujarat, India

<sup>3</sup>Medical Affairs, Eris Lifesciences Ltd. Ahmedabad, Gujarat, India

**Received:** 10 May 2023

**Accepted:** 08 June 2023

### \*Correspondence:

Dr. Mayank Rajawat,

E-mail: [mayank.medicalaffairs@gmail.com](mailto:mayank.medicalaffairs@gmail.com)

**Copyright:** © the author(s), publisher and licensee Medip Academy. This is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

## ABSTRACT

Hydatid cyst also called as cystic echinococcosis is a parasitic zoonotic infection caused by *Echinococcus granulosus*. Humans get infected by this parasite by consumption of contaminated food with dog faeces or by direct contact with animal and it particularly affecting liver and lung. However, cases of primary muscle hydatid are about 5%. In India, reported cases of primary muscle hydatids are very rare. In this case report, we presented an exceptional rare case of unusual primary hydatid cyst localized in infraspinatus muscle of scapular region.

**Keywords:** Infraspinatus muscle, Hydatid cyst, Echinococcosis

## INTRODUCTION

Hydatid cysts also called as cystic echinococcosis is a severe parasitic zoonotic infection caused by helminth named *Echinococcus granulosus* (*E. granulosus*). Dogs are the predominant host of echinococcosis. In humans, cystic echinococcosis is transferred by ingestion of food contaminated with dog faeces and/or by direct contact with animal. In human causative agent is larvae of this parasite.<sup>1-4</sup>

Apart from middle east and Mediterranean countries, infection is also endemic in India.<sup>4</sup> It may affect any part or organ of the body, but most common sites involved is liver (50–60%) and lungs (20–25%). Other sites involved are muscles (5%), bones (3%), kidney (2%), and spleen (1%). The reported cases of primary muscular hydatids are very rare in India with extremely rare involvement of scapular region (Table 1).<sup>5-11</sup>

In India, still there is no single case reported for primary hydatid cyst of infraspinatus muscle in scapular region.

**Table 1: Review of literatures on hydatid disease in India.**<sup>5-11</sup>

| S. no. | Author         | Year | Site of lesion                            |
|--------|----------------|------|-------------------------------------------|
| 1      | Arora et al    | 2006 | Muscle (thigh)                            |
| 2      | Babu et al     | 2008 | Intraabdominal wall                       |
| 3      | Rao et al      | 2011 | Anterior abdominal wall                   |
| 4      | Shanthi et al  | 2011 | Spleen                                    |
| 5      | Malik et al    | 2011 | Spleen                                    |
| 6      | Rao et al      | 2012 | Spectrum of hydatid disease in India      |
| 7      | Usharani et al | 2013 | Anterior abdominal wall (inguinal region) |

In this case report, we presented an exceptional rare case of an unusual primary hydatid cyst localized in the right infraspinatus muscle of scapular region.

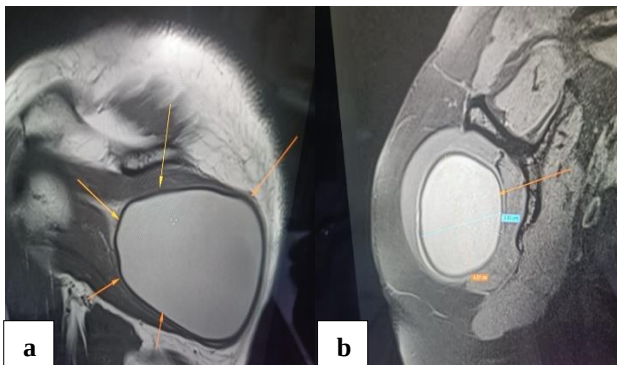
## CASE REPORT

A 50-year-old female presented a large swelling with pain in the right shoulder. She noted the pain about 2 months ago with disability in the movement. There was no history of fever, abdominal pain, chest pain, or trauma. Also, there was no history of dog bite or familiarity with dog. Physical examination revealed diffused swelling occupying the scapular region. No history of pre echinococcal indirect hemagglutination test (IHAT) or another test is done to confirm the same.

Hematological examination showed normal haemoglobin and red blood cells (RBC) levels. Total white blood cells count was  $7300 \text{ mm}^{-3}$  with eosinophil count of 2%. Also, it showed normal erythrocyte sedimentation rate.

Ultrasonographic examination of the local part including right shoulder soft tissue revealed well-defined cystic lesion in infraspinatus muscle of scapular region without solid component or vascularity. Ultrasonography (USG) suggested possibility of huge cystic lesion with dimension of  $8 \times 5 \text{ cm}$ .

Further patient underwent for magnetic resonance imaging (MRI) examination, which revealed altered signal intensity lesion within the substance of infraspinatus muscle near its myotendinous junction measuring  $80 \times 56 \times 42 \text{ mm}$ . There was a hyperintense on T2W/STIR images and hypointense on T1W images. No evidence of solid component. Findings suggested possibility of benign cystic lesion.



**Figure 1: MRI aspects of right shoulder (a) PD-FS-TSE coronal image, and (b) PD-FS-TSE sagittal image show the lesion in infraspinatus region.**

PD-FS-TSE: Proton density-fat saturation-turbo spin echo

Surgically, the mass was excised under general anesthesia. Histological gross examination of excised specimen revealed mass was in two pieces consisted of multiple cystic soft tissue measuring  $1.5 \times 1.0$  to  $6.0 \times 5.0 \times 4.0 \text{ cm}$  with clear fluid content (Figure 2). Microscopic examination showed one cystic lesion wall which is composed of outer acellular laminated layer. Also, wall was composed of inner cellular layer with attached scattered protoscolices and few refractile hooklets. Other

cystic wall composed of dense fibrous tissue, with synovial cell lining and multinucleated foreign body type giant cells surrounded by muscle coat. Also, there was a presence of focal inflammation with no evidence of granuloma or malignancy. Histological examination altogether confirmed the hydatid cyst.



**Figure 2: Gross specimen of hydatid cyst.**

Post-operatively patient was put on the tablet albendazole 400 mg twice daily for 28 days. Serological test of IgM for recurrence on follow-up was negative.

## DISCUSSION

Cystic echinococcosis or hydatid disease is zoonotic infection caused by the tapeworm *E. granulosus*.<sup>1,2</sup> This species of tapeworm is endemic in middle east and Mediterranean countries, India, Australia, Eastern Europe, and Africa. Predominantly hydatid disease is a disease occurring in dogs.<sup>3,4</sup> However, they are easily transmitted in mammals by expelled larval eggs of *E. granulosus* in the faeces of dog.<sup>3</sup> In human infestation of hydatid cyst is more commonly occurs in liver because ingested eggs hatch in the intestines and are transported to the liver through hepatic portal system.<sup>1</sup> Apart from liver the other common site is lung, but they can be infested other organs or tissues. Primary muscle hydatid cysts are rare and uncommon. Evidently hydatid cysts are localized in peripheral muscles such as supraspinatus, deltoid, thigh, gluteus muscles, quadriceps, triceps brachii, gastrocnemius, biceps femoris and muscles of chest wall.<sup>1-3</sup> Hydatid disease of infraspinatus muscle is rare. Till now only one case of hydatid cyst in infraspinatus muscle was reported in Greece across the world.<sup>3</sup> In India, reported cases of extrahepatic hydatid cysts involve thighs, intraabdominal wall, spleen, inguinal region, and shoulder.<sup>5-11</sup> However, hydatid disease in the infraspinatus muscle is not reported.

Main symptom reported in muscular hydatid disease is palpable swelling.<sup>12</sup> History of contact with animals must be questioned during screening. Diagnosis with ultrasonogram, CT or MRI pre-operatively can be made for identification of location and size of the cysts.<sup>13</sup> Other

laboratories routine test should be considered like total white blood cell count and % haemoglobin. Occasionally eosinophilia may be present in case of cyst leakage or may be normal.<sup>11,13</sup> Serological tests involves detection of specific antibodies like IgG, IgA, and IgM to the parasite and are the most common method used to diagnose earlier and recent infection with *E. granulosus*. High titres of IgG antibodies indicate exposure to the parasite while IgM and IgA antibodies are observed in active infection.<sup>13</sup>

Generally complete diagnosis is made during surgical procedure and confirmed histologically. Histologic assessment of the cyst wall commonly shows an outer chitinous (or fibrous laminar) and an inner germinal layer, enclosed by either granulation tissue or a fibrous capsule. Protoscoleces can easily identified using microscopic examination in histological assessment.<sup>14,15</sup>

Various reports suggested surgical procedure as gold standard and effective treatment for complete eradication of hydatid disease.<sup>13</sup> In case of muscular hydatids, use of perioperative medical treatment as adjuvant with surgery is rationale to reduce the risk of dissemination and preventing the local recurrence.<sup>13,16</sup> Pre-operative medical treatments with albendazole should be considered for sterilization of cyst, reduction of tension in cyst, and chances of leakage and consequent anaphylaxis. Post-operative treatment with anthelmintic like albendazole reduces the recurrence rate.

## CONCLUSION

Hydatid cysts are commonly found in liver and lungs, but muscular hydatid disease is rare. In this case we reported hydatid cysts of very rare site which was infraspinal muscles. In endemic regions, differential diagnosis of muscular hydatid cysts especially in the rare sites must be considered. Multiple diagnostic modalities are used for diagnosis of hydatid cysts. Gold standard treatment is complete removal of hydatid cyst through surgical procedure. Post-operative treatment with albendazole reduces the risk of recurrence of cyst.

*Funding: No funding sources*

*Conflict of interest: None declared*

*Ethical approval: Not required*

## REFERENCES

1. Tekin R, Avci A, Tekin RC, Gem M, Cevik R. Hydatid cysts in muscles: clinical manifestations, diagnosis, and management of this atypical presentation. *Rev Soc Bras Med Trop*. 2015;48(5):594-8.
2. Yilmaz S, Gulcek M, Demirtas Y, Turan S. An unusual cause of mass in the shoulder: A primary hydatid cyst. *Int J Shoulder Surg*. 2014;8(2):58-60.
3. Gougoulas NE, Varitimidis SE, Bargiotas KA, Dovas TN, Karydakis G, Dailiana ZH. Skeletal muscle hydatid cysts presenting as soft tissue masses. *Hippokratia*. 2010;14(2):126-30.
4. Deplazes P, Rinaldi L, Alvarez Rojas CA, Torgerson PR, Harandi MF, Romig T, et al. Global Distribution of Alveolar and Cystic Echinococcosis. *Adv Parasitol*. 2017;95:315-493.
5. Arora V, Nijjar IS, Gill KS, Singh G. Case report: primary hydatid cyst of muscle – a rare site. *Indian J Radiol Imaging*. 2006;16(2):239-41.
6. Babu KS, Goel D, Prayaga A, Rao IS, Kumar A. Intraabdominal hydatid cyst: a rare case report. *Acta Cytol*. 2008;52(4):464-6.
7. Rao NM, Shanthi V, Ramakrishna BA, Chaitanya B, Sekhar BH. Anterior abdominal wall hydatid: a rare case report. *J Biosci Technol*. 2011;2(4):349-52.
8. Shanthi V, Rao NM, Lavanya G, Krishna BR, Muralimohan KV. Splenic hydatid cyst: a rare site of presentation. *J Biosci Tech*. 2011;2(6):436-8.
9. Malik AA, Bari S, Younis M, Wani KA, Rather AA. Primary splenic hydatidosis. *Indian J Gastroenterol*. 2011;30(4):175-7.
10. Rao SS, Mehta B, Narang R. The spectrum of hydatid disease in rural central India: an 11-year experience. *Ann Trop Med Public Health*. 2012;5(3):225-30.
11. Usharani A, Deepica G, Aruna S, Kulkarni S, Sai Kamal Kumar G, Balamuralikrishna P. Case reports of hydatid disease. *J Epidemiol Glob Health*. 2013;3(2):63-6.
12. Di Gesu G, Picone A, La Bianca A, Massaro M, Vetri G. Muscular and subcutaneous hydatidosis. *Minerva Medica*. 1987;78(12):835-40.
13. Derbel F, Ben M, Hadj Hamida MB, Mazhoud J, Youssef S, Ben A, et al. Hydatid Cysts of the Liver - Diagnosis, Complications and Treatment. *Abdominal Surgery*. 2012.
14. Toğral G, Arıkan ŞM, Ekiz T, Kekeç AF, Ekşioğlu MF. Musculoskeletal Hydatid Cysts Resembling Tumors: A Report of Five Cases. *Orthop Surg*. 2016;8(2):246-52.
15. Torgerson PR, Deplazes P. Echinococcosis: diagnosis and diagnostic interpretation in population studies. *Trends Parasitol*. 2009;25(4):164-70.
16. Arazı M, Erikoglu M, Odev K, Memik R, Ozdemir M. Primary echinococcus infestation of the bone and muscles. *Clin Orthop Relat Res*. 2005;432:234-41.

**Cite this article as:** Chhaya G, Shah DK, Prajapati H, Rajawat M. Primary hydatid cyst of muscle-a rare cause of mass in the shoulder: a case report. *Int J Adv Med* 2023;10:578-80.