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Unexpected Site of Hydatid Cyst, in Thigh- A Case Report

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Authors' contributions

This work was carried out in collaboration among all authors. Author CTD made the case report. Authors CTD and ST were involved in the management of the case. Authors CS and MM helped in corrections of the case reports. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Introduction: Hydatid cyst is a parasitic disease common in India. Usual locations of the cyst are the lungs and liver. The occurrence of the hydatid cyst in other locations is rare.
Presentation: The presenting concern is a 10-year-old girl with a painless swelling in lower 1/3rd of the right thigh on the posterior side. Clinical examination is suggestive of a palpable cystic swelling which is involving the muscle underlying it. USG and CT of thigh revealed a cystic swelling in the Biceps femoris muscle with high suspicion of hydatid cyst. Pre-operative albendazole was used followed by complete removal of the cyst and the use of albendazole postoperatively.
Discussion: Hydatid cyst most commonly utilizes oxygen for growth, but skeletal muscle contains Lactic acid. Hence occurrence of cyst in skeletal muscle is rare. Ultrasound has a sensitivity of 100% in typical cases and is diagnostic. Computed tomography provides better information about cyst. Though chemotherapy is controversial, Preoperative and postoperative chemotherapy shown to decrease the incidence of recurrent disease. Excision in one piece with total cystectomy is the ideal process. Early treatment is mandatory to avoid local and general complications that are directly related to duration of cyst.
Conclusion: This case report helps us to keep hydatid cyst a differential diagnosis of intramuscular

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cystic swelling and treat accordingly. The presence of this cyst in unusual locations makes diagnosis and treatment challenging and more interesting. Timely diagnosis, timely intervention and chemotherapy is required to prevent intra operative complications and recurrence. This case study helps in making a specific management protocol and adds up to the previous case reports.

Keywords: Hydatid cyst; hydatid cyst of thigh; Echinococcus granulosus; rare hydatid cyst.

ABBREVIATIONS

USG : Ultrasonography
CT : Computed Tomography
LFT : Liver Function Tests

1. INTRODUCTION

Hydatid cyst is a parasitic infestation caused by Echinococcus granulosus. Liver is the most common site (75% of cases), followed by lungs (15%), spleen (5%), and other organs (5%) [1]. It is are rarely detected in muscle tissue. Primary muscular hydatidosis is uncommon even in endemic regions (1% to 4%) [2].

Theoretically, the muscle is inhospitable for echinococcal infestations because of its contractility and high level of lactic acid [3]. The presence of lactic acid creates an unfavorable milieu for growth [4] Because of asymptomatic evolution, the diagnosis of musculoskeletal hydatidosis remains difficult and therapeutic management is often complicated [2].

Hydatid disease should be considered for every soft cystic mass in any anatomical location, especially in areas where the disease is endemic [1] and where patients have risk factors like exposure to dogs or rural community.

2. AIM

The main purpose of this case report is to know in detail about Hydatid cyst of the muscular tissue and its clinical presentation since it is usually misdiagnosed.

2.1 Case Presentation

A 10 yr old girl residing in an urban community of Raipur was presented with a swelling on the back of right thigh for 1 year and occasional dragging pain for 1 month. There was a history of exposure to dogs.

3. CLINICAL FINDINGS

3.1 Local Examination of the Right Thigh

A single palpable oval swelling of size 4 x 3 cm was noted in the lower 1/3 rd of the posterior aspect of right thigh. It was Soft/cystic in consistency with restricted mobility (Fig. 1) and became less prominent on contraction of hamstring muscle. No tenderness/local rise of temperature was elicited.

3.2 Investigations

1. Ultrasound of right thigh revealed a 3.5 x 3.5 cm round cystic swelling with thin septations raising a high suspicion of Hydatid cyst.
2. CT of right thigh reported a cystic swelling with thin septations arising within Biceps Femoris muscle (Fig. 2).
3. Routine blood investigations, Eosinophilic count, LFT, X-ray chest, and USG abdomen were normal which is suggestive of non involvement of Liver and Lungs.

Fig. 1. A single palpable oval swelling of size 4 x 3 cm in the lower 1/3 rd of posterior aspect of right thigh

3.3 Therapeutic Intervention

Pre-operatively albendazole (10 mg/kg/day) is given for 14 days followed by en bloc dissection of cyst without spillage of the content (Fig. 3).

3.4 Follow-up and Outcomes

Gross: Pearly white cystic swelling of size 4 x 3 cm with multiple daughter cysts and viscous hydatid fluid as content was observed.

Histology: Eosinophilic laminated hyaline membrane with scolex and hooks identified- HYDATID CYST.

Pericyst showed muscular tissue, fibrosis with lymphoid follicles.

Postoperatively albendazole (10mg/kg/day) was given for 1 month.

Patient was followed up after 1 month, 3 months, 6 months and 1 year. Patient did not show clinical or radiological signs of recurrence.

3.5 Timeline

Patient came to hospital (Day 1) → Radiological investigations (Day 2) → preoperative Albendazole (Day 2-15) → surgery on Day 16 → postoperative Albendazole for one month → followup at 3 months, 6 months and 1 year.

Timely diagnosis, timely intervention and chemotherapy is required to prevent intra operative complications and recurrence.

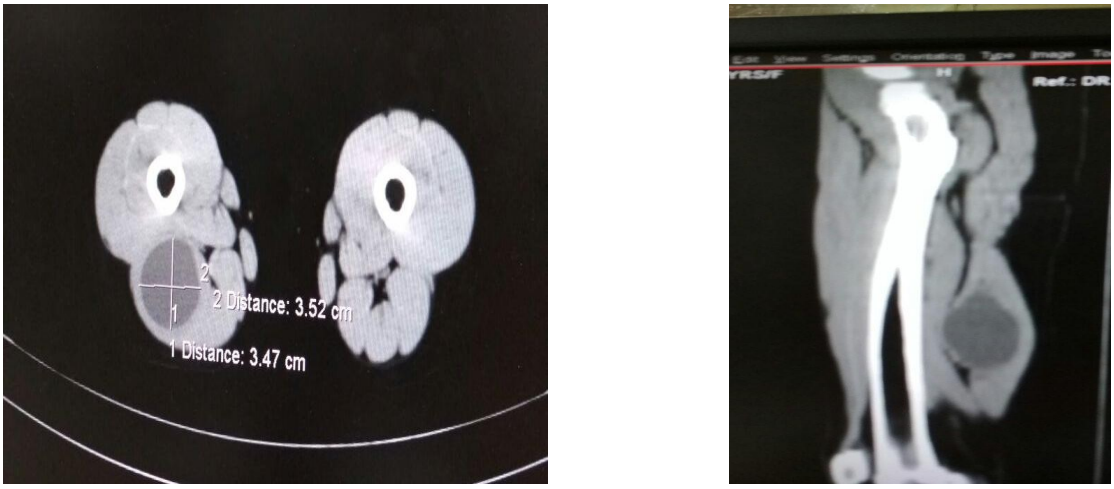
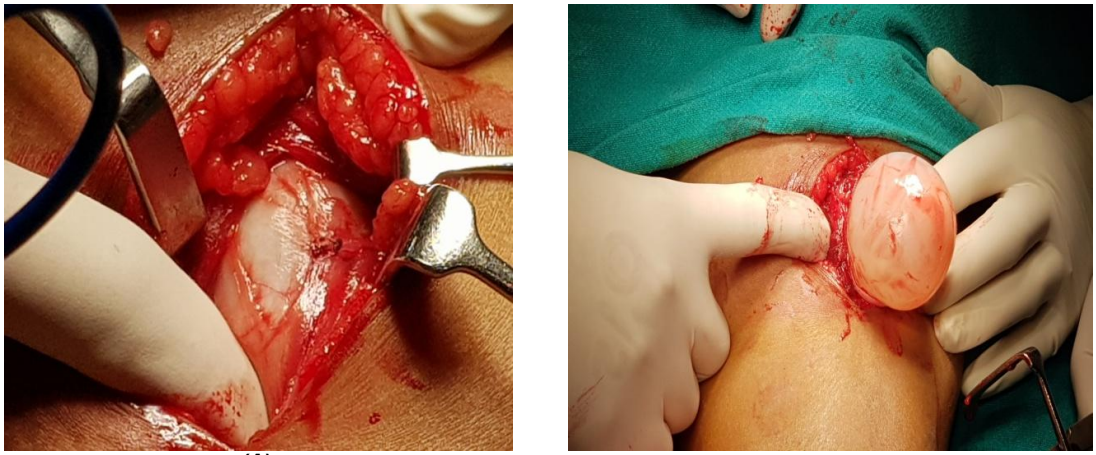
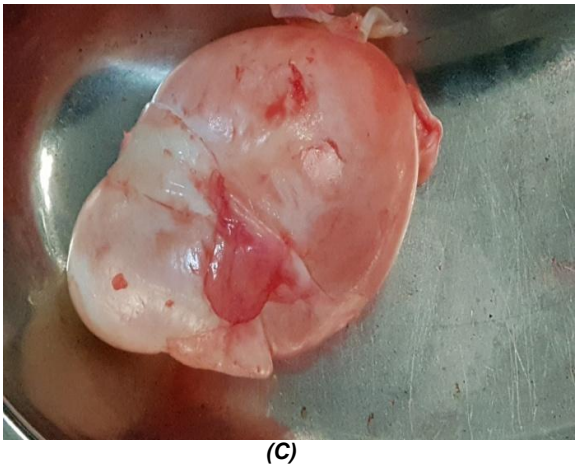


Fig. 2. CT images showing cystic swelling with thin septations arising within Biceps Femoris muscle



(A) ***(B)***



(C)

(D)

Fig. 3. Intra operative pictures A) Vertical incision of 5cm in the posterior aspect of right thigh with muscle splitting B) Cyst popped up C) Entire cyst is removed en bloc without spillage D) Incision closed with interrupted sutures

4. DISCUSSION

Primary hydatid cyst of skeletal muscle is rare, with a reported prevalence of 0.5% to 4.7%, because the cyst uses oxygen for growth and muscle usually contains lactic acids [5] with predilection for the lower limbs [4]. The hydatid cyst of the striated muscle preferentially affects the larger muscles, with a predilection for the proximal muscles [6]. Theoretically, the muscle is inhospitable for echinococcal infestations because of its contractility and high level of lactic acid [3]. The cyst grows with difficulty in muscles because of their contractility and the presence of lactic acid [3].

Hydatid cyst should be considered when evaluating cystic masses, especially in the endemic areas [4]. The pathogenesis of hydatid cyst occurring at rare sites are not well demonstrated. Possible pathogenesis is dissemination through lymphatic channels [7].

Hydatid cysts are best treated by complete excision of the cyst, so preoperative diagnosis and localization is a must [8].

Ultrasonography should be the first diagnostic tool used for detection of hydatid disease of soft tissue [9]. Ultrasound can guide the diagnosis with a sensitivity of 100% in typical cases. Computed tomography provides better information about number, size, site, and structure of cysts, as well as their relationship with the surrounding tissues [2]. In my case, Ultrasonography revealed a cystic mass with high suspicion of hydatid cyst with septations. CT revealed a single cystic mass in the biceps femoris muscle.

Confirmation through imaging studies helps to prevent unnecessary cystic puncture and its complications such as local dissemination and anaphylactic shock.

Serologic tests are valuable when they are positive, but half of the primary intramuscular hydatidosis cases give a false negative. The IHA sensitivity rate has been reported as 67%. Although serology tests like IHA can help make the diagnosis, complete reliance on them is not recommended [9]. Intradermal Casoni test, the human basophil degradation test, and the complement fixation test have only historical relevance [1] which were not done in my case.

Pulmonary and hepatic location of Hydatid are the most common, a chest X-ray and an upper abdomen USG are always necessary to define if we are dealing with primary or secondary hydatid disease [10] which were negative in my case which was suggestive of primary muscular hydatosis without involvement of Liver or lung.

Chemotherapy is controversial and there is no consensus supporting its use in musculoskeletal echinococcosis [2] In fact, starting the medical treatment before excision yields a high serum level which reduces intracystic pressure, while the postoperative treatment is aimed at stopping parasitic dissemination [2]. Among the Anti Helmenthic drugs, albendazole alone is ovicidal, larvicidal, and vermicidal [1].

Once diagnosis has been made, pericystectomy need to be performed and anthelmintic drugs should be administered as prophylaxis against possible recurrences [10] Recurrent disease has been reported in 10 % of patients. Perioperative chemotherapy using albendazole is shown to decrease the incidence of recurrent disease [7].

Combination adjunctive chemotherapy with anthelmintics is recommended to cover the risk of dissemination during exploration. Many authors recommend preoperative use of anthelmintics to sterilize the cyst, and reduce the consequences of spillage, and the chances of anaphylaxis and dissemination, at surgery [11]. Hence in this case preoperative albendazole for 14 days, post-operative albendazole for 1 month was given.

Some studies state that, incomplete excision of the cyst requires additional chemotherapy [12].

Incisional biopsy and marginal excision are contraindicated if the list of differential diagnoses includes hydatid cyst [4].

The treatment of a muscular hydatid cyst is surgical. Surgery should be prudent and should avoid the opening of the cyst during the dissection. One must protect the operative field with a solution of hypertonic saline and/or hydrogen peroxide [6]. The fluid of the Echinococcus granulosus cyst contains significant amounts of foreign protein and is extremely toxic to the host [4]. Excision in one piece with total cystectomy is the ideal process [6] and the same procedure was done with this case.

Early treatment is mandatory to avoid local general complications that are directly related to the duration of cyst. Aim of treatment is the complete removal of the parasite without any spillage during operation and unnecessary damage to host tissue [1]. Timely diagnosis is necessary to avoid rupture, immunological reactions, and secondary infection.

This diagnosis requires a multidisciplinary approach including clinical, radiological, and biological examinations. Surgical procedure combined with albendazole appears to be the optimal curative course of action [2].

5. CONCLUSION

Hydatid cyst of thigh is a very rare manifestation which requires a combined approach of clinical and radiological investigations for the diagnosis. This rare entity has to be considered as a differential diagnosis for any cystic swelling in the musculoskeleton of extremities.

Pre-operative Albendazole (10 mg/kg) given for 14 days and postoperative albendazole for 1 month helps in preventing recurrence. However preoperative and postoperative treatment duration may vary from institution to institution and place to place.

Timely diagnosis, timely intervention, chemotherapy is required to prevent intra operative complications and recurrence.

This study adds up to the other such case reports which helps in the formation of a specific treatment protocol for an approach to this rare entity.

CONSENT

All authors declare that written informed consent was obtained from the patient for publication of this case report and accompanying image.

ETHICAL APPROVAL

As per international standard or university standard ethical approval has been collected and preserved by the authors.

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COMPETING INTERESTS

Authors have declared that no competing interests exist.

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