



## A Rare Case of Large Hydatid Cyst at Atypical Location: Case Study

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### **Authors' contributions**

*This work was carried out in collaboration among all authors. Author AKD designed the study. Author URD wrote the protocol and wrote the first draft of the manuscript. Authors JVV and SD performed the statistical analysis. Author HD managed the analyses of the study. Author Arun managed the literature searches. All authors read and approved the final manuscript.*

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Case Study

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### **ABSTRACT**

Hydatid disease also known as Echinococcosis and caused by Echinococcus granulosus and Echinococcus multilocularis, which cause Cystic echinococcosis (CE) and alveolar echinococcosis (AE). Most common site for Hydatid cyst is liver and lung, but it can grow at unusual sites like bone, muscle, except hair and nail. Present case was a 65 years male who came with complaints of gradually increasing swelling of left thigh for last 9 months. There was recent onset of pain while changing posture from sitting to standing. USG (Ultrasonography) showed a hypoechoic lesion extending from groin to mid-thigh. MRI showed multiple round to oval cystic mass lesions in adductor muscle of left thigh with thin walled daughter cysts suggestive of Hydatid disease. Hydatid Serology was negative with no eosinophilia on hemogram. Albendazole was started 2 weeks preoperatively. Patient underwent en bloc excision and Histopathology report confirmed the diagnosis of hydatid disease. Patient was put on albendazole 400 mg for 14 weeks. Post-operative follow-up for one year showed no recurrence.

**Conclusion:** Hydatid cyst of a thigh muscle is a very rare condition and its diagnosis would require a high index of suspicion. The results of hydatid serology in cases of Musculo-skeletal organs could

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be negative but radiology imaging of the thigh including ultrasound scan, computed tomography scan and magnetic resonance imaging scan would tend to demonstrate features of the lesion that would be suggestive of Echinococcus disease of the thigh. Treatment of the disease does involve utilization of albendazole treatment for many weeks and complete excision of the lesion taking care to avoid spillage of the contents of the cyst.

**Keywords:** *Echinococcus granulosus, hydatid cyst; thigh; musculoskeletal; hydatid serology; USG; MRI; albendazole; surgical excision.*

## 1. INTRODUCTION

Hydatid disease is also known as Echinococcosis. This Zoonotic disease is caused by Echinococcus granulosus and Echinococcus multilocularis, adult or larval forms. It has one definitive host (Dogs, jackals and wolves) and second Intermediate host (sheep, cattle). Human is incidental intermediate host. Which cause Cystic echinococcosis (CE) and alveolar echinococcosis (AE) [1]. The disease is more common in most parts of the world, especially in cattle and sheep farming places of Asia, North and East Africa, South America, Australia and Middle East. It is transmitted by contaminated water or vegetables. Most common site for Hydatid cyst is liver (68.8-80%), lung (10-22.4%) or both [2]. But it can form cysts at unusual site like bone, muscle, sub cutaneous (2%) except hair, nail and teeth [3]. Primary Musculoskeletal hydatid cysts are rare even in endemic region. Hydatid cyst of muscle are usually secondary in nature, spontaneous from migration of larvae from primary site or trauma induced cyst rupture or iatrogenic [4]. Musculoskeletal hydatid cyst is like slow growing soft-tissue tumour which is painless without any inflammatory signs.

We report a rare case of 67 years male presenting with primary musculoskeletal hydatid cyst involving the adductor muscle of left thigh.

## 2. CASE PRESENTATION

A 67 year old male patient presented with a gradually increasing swelling of left thigh for last 9 months. He had history of trauma one month prior to the onset of swelling. Swelling was increasing gradually in size without any pain, difficulty in walking, fever and was not associated with loss of weight. Patient developed recent onset of pain at limb while changing posture from sitting to standing. Patient was a farmer belonging to a village in Haryana (North India). He had close contact with cattle. He was also a known case of coronary artery disease and he underwent stenting 2 years back and was

on medications. On examination he had a swelling present at anterior aspect of left thigh approximate size of 15\*10 cm with no redness, no rise of temperature, soft in consistency and deep to the fascia. USG showed 15\*5 cm hypoechoic lesion present in antero-medial aspect of left thigh extending from groin to mid-thigh suggestive of hydatid disease or old organised hematoma in view of previous history of trauma and blood thinner. MRI showed multiple round to oval shaped complex cystic masses of size 7.5\*6.8\*9.5cm involving adductor muscle of left upper thigh suggestive of hydatid disease (Figs. 1, 2).



**Fig. 1. T2 Weighted image shows multi-loculated cystic lesion in antero-medial aspect of adductor muscle compartment with multiple cystic lesions within it. Cyst shows hyperintense signals**

Echinococcal Serology was negative. Chest x-ray and USG abdomen for lung and liver was normal. Complete hemogram showed no eosinophilia.

Pre operatively patient was put on albendazole for two weeks and surgical en bloc excision of left thigh hydatid cyst was done without spillage and rupture of cyst under general anaesthesia (Figs. 3, 4). A medial longitudinal incision was made and cyst was identified and complete excision

was performed. Operative field was protected with 10% povidone iodine soaked sponges and wound closed over a negative suction drain. Post operatively patient was discharged on albendazole 400 mg BD for 14 weeks. Histopathology showed hydatid cyst. Patient was followed up for one year showing no recurrence.



**Fig. 2. MRI T1 image shows multi cystic lesion at anterior aspect of left thigh and cyst shows hypo intense signal**



**Fig. 3. Large Excised hydatid cyst specimen**



**Fig. 4. Cut section of excised specimen shows multiple small cystic lesions (daughter cysts) with clear fluid**

### 3. DISCUSSION

Soft tissue hydatid cysts are usually secondary cysts with primary elsewhere in the body [5]. Primary hydatid cyst of musculoskeletal system is very rare [3,6,7,8]. This is because of the following reasons: 1) Filtration barrier in lung and liver (Most of larvae and eggs are traps here so liver and lung are most common site for hydatid cyst). 2) Muscle contain high level of lactic acid and high level of lactic acid is not a favourable site for development of hydatid cyst. 3) contractile nature of muscle fibres prevents fixation of larvae to the tissue. So hydatidcyst of musculoskeletal system are usually secondary. Primary hydatid cyst of musculoskeletal system shows no liver or lung hydatid and with no previous history of hydatid surgery. The investigations also show no hydatid cyst at other places of body (liver, lung, brain, spleen).

Diagnosis of hydatid cyst is made by clinical, radiological and histopathological examination.

Any patient coming from endemic area with a painless cystic soft tissue swelling with gradually increasing in size. Other differential diagnosis [3] for such cystic swellings of thigh are cystic tuberculosis, soft tissue tumour, sciatic nerve hematoma and cystic lymphangioma [3].

Hemogram of hydatid disease shows eosinophilia and positive serology. Serology is positive only in active and most of the primary cases (sensitivity 90% in hepatic hydatid cyst). Negative serology doesn't exclude the diagnosis of hydatid cyst [3,4,6,8,9]. 30-40% patients with hydatid disease are antibody negative and half of the intra muscular hydatid cyst shows false negative serology. In our case there was no eosinophilia and hydatidserology was also negative.

USG is the first radiological tool to diagnose the hydatid cyst. The sensitivity of USG in detecting the hydatid cyst is 95% and sensitivity increases to 100% if the cyst shows vesicular fibres [8]. Our case did not show any features of hydatid cyst on USG.

MRI is used as a diagnostic tool [9] and it shows the lesion its extent and margins and involvement of surrounding structures. MRI can differentiate cyst from malignancy [10] and help in planning for surgical procedure [6]. In our case MRI T1 images showed cystic lesion with hypoechoic lesion and T2 images showed hyperechoic lesions with double wall, and daughter cysts suggestive of hydatid cyst.

Best treatment for hydatid cyst at atypical places is complete surgical excision without opening the cyst, if necessary the cyst can be managed by PAIR(puncture aspiration injection and re-aspiration) of Ethanol or protoscolicidal agents [3,10]. It prevents dissemination of hydatid material and prevent anaphylactic reaction.

Histopathological report of specimen showed it to be hydatid cyst. Patient was continued on treatment with albendazole 400mg BD for 14 weeks to reduce the local recurrence of the disease [7]. No recurrence was there during one year follow up.

Diagnosis of Hydatid disease at unusual location may be challenging even in endemic areas and pose diagnostic dilemma because primary extra hepatic and extra pulmonary hydatid disease are rare [11]. In patients presenting with cystic swelling at unusual location, we should try to rule out hydatid disease before planning any intervention on it.

#### 4. CONCLUSION

Hydatid cyst of atypical places are slowly growing painless masses. It is necessary to rule out hydatid cyst before performing any procedures on it by clinical, radiological. Otherwise cyst rupture cause dissemination of hydatid material and anaphylactic reaction can happen.

#### CONSENT

As per international standard or university standard, patient's consent has been collected and preserved by the authors.

#### ETHICAL APPROVAL

It is not applicable.

#### COMPETING INTERESTS

Authors have declared that no competing interests exist.

#### REFERENCES

1. Zhang W, Li J, McManus DP. Concepts in immunology and diagnosis of hydatid disease. Clin. Microbiol. Rev. 2003; 16 (1):18–36.
2. Al-Hakkak SMM. Adductor magnus muscle primary hydatid cyst rare unusual site: A case report. Int J Surg Case Rep. 2018; 51:379-84.
3. Samiee-Rad F, Emami A. An Iranian man with increased thigh mass due to a hydatid cyst. GMS Hyg Infect Control. 2020;15: Doc20.
4. Acu L, Acu B, Acu R, Sahin SA. Hydatid cyst presented as a subcutaneous thigh mass. J Ultrasound. 2016;20(4):321-24.
5. Dirican A, Unal B, Kayaalp C, Kirimlioglu V. Subcutaneous hydatid cysts occurring in the palm and the thigh: two case reports. J Med Case Reports. 2008;2:273.
6. Durakbasa MO, Kose O, Islam NC, Kilicoglu G. A primary hydatid cyst of the gracilis: A case report. J. Orthop. Surg. 2007;15:118–120.
7. Zazo A, Zazo R, et al. Uncommon Musculoskeletal femoral hydatid cyst disturbs a female for a year: Case report. Annals of Medicine and Surgery. 2020; 55:30-32.
8. Singal R, Dalal U, Dalal AK, Singh P, Gupta R. Subcutaneous hydatid cyst of the thigh. Southern Medical J. 2010;103(9): 965-66.
9. Salih AM, Kakamad FH, Salih RQ, et al. Hydatid cyst of the thigh: A case report with literature review. Int J Surg Case Rep. 2018;51:8-10.
10. Gigeikli O, Akgul T, An unusual case of mass localized on Vastuslateralis muscle in childhood: Hydatid cyst. International Journal of Surgery Case Reports. 2015;6: 179-181.
11. Dalal AK, Dalal UR, Wani AJ. Pelvic hydatid mimicking inguinal hernia. J. Evid. Based Med. Healthc. 2017;4(91):5523-25.

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