Case Report:

Primary musculoskeletal hydatid cyst in Thigh: Diagnostic challenge for an unusual localization

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Abstract

Hydatid diseases are cystic parasitic infestation caused by Echinococcus. The most frequently affected organs are liver and lung. The nonspecific clinical presentation and possible post-therapeutic complications lead to problems for the diagnosis of this infection and the support of the patient. A 42 year old female patient presented with complaint of swelling and pain in the left thigh since 5 years. She was initially diagnosed as lipoma of thigh, but ultrasonography revealed to be a cystic swelling suggestive of hydatid disease. MRI further reinforced the diagnosis. Patient was given albendazole preoperatively. The swelling was removed en bloc and advised for adjunctive albendazole chemotherapy (15 mg/kg/day) for three months.

Keywords: Hydatid cyst, Primary, Thigh

Introduction

Hydatid diseases (HDs) are cystic parasitic infestation caused by Echinococcus which is the cestode of the Taeniidae family [1,2]. Hydatid disease is a cosmopolitan anthropozoonosis more prevalent in livestock-rearing countries (3). Hydatid cysts (HC) are most frequently found in the liver (75%) and lungs (15%) (4). Primary muscular hydatidosis is uncommon even in endemic regions (1% to 4%) (4,5). Other rarely affected organs are bone, skeletal and smooth muscles, viscera and mediastinum [7,8,9]. The diagnosis of musculoskeletal hydatidosis remains difficult due to asymptomatic evolution.

Case Report

A 42 year old female patient presented with complaint of swelling and pain in the left thigh since 5 years. There was no history of abdominal pain, cough, fever, weight loss or urticaria. Patient was a known case of Type 2 diabetes mellitus and hypertension on medications. On local examination, swelling was approximately 12x7 cm, non tender and soft. There was no overlying skin change and temperature was normal. She was initially diagnosed as lipoma of thigh, but ultrasonography revealed of well defined lesion with multiple cysts in the intramuscular plane measuring 13.6 X 5.8 cms, with no internal vascularity (Fig. 1). MRI was advised which revealed well- defined multiloculated cystic lesion with multiple daughter's cysts within noted measuring 7.0 x 5.6 x 12 cms in the intramuscular plane of left thigh involving vastus medialis muscle, subtle rim enhancement on post contrast study. (Fig. 2,3,4,5) Serologic test, ELISA was negative. CT head, ultrasound abdomen and thorax and X-ray chest were normal. Longitudinal elliptical incision was performed in the anterolateral aspect of the left thigh, the Cyst was en bloc enucleated (Fig. 7)., which was 20 cm × 15 cm in dimensions (Fig.7). Cut section showed multiple daughter cysts (Fig.8). Cyst cavity was irrigated with 10% betadine and wound closed with closed suction drain. Histopathological diagnosis confirmed the diagnosis.



Fig 1. Ultrasound of left thigh

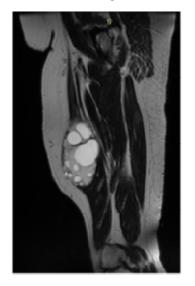




Fig 2. Coronal and saggital T2WI.(Hyperintense cystic lesion)

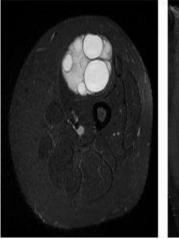




Fig 3. Axial and coronal STIR (hyper intense cystic lesions)



Fig4. Coronal T1 (hypo to isointense cystic lesion)



Fig5. Coronal image (no areas of blooming noted within the cystic lesion)



Fig6. Pre operative(swelling of left thigh)

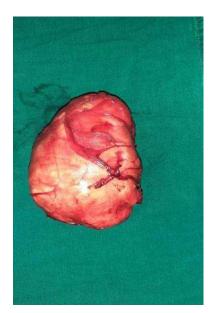


Fig7. Specimen intoto



Fig8. Daughter cysts.

Discussion

The prevalence of subcutaneous HDs is not well determined; it has been postulated that it is around 2%(²). Hydatid cyst can occur in any part of the body as worm can remain dormant for months to decades without symptom. Either it is diagnosed incidentally or causing pressure symptoms making the victim seek medical advice(¹⁴). 17 cases of HD of the thigh have been reported in the literature [², 8,9,10,11,12,13,14,15,16,17,18,19,20,21,22,23,2⁴]. The age ranged between 8 to 61 years [¹²,²¹]. Ten patients (58%) were female. Left side was affected more than the right one (ten in left side, 7 in right side) [2,[8], [9], [10], [11], [12], [13], [14], [15], [16], [17], [18], [19], [20], [21], [22], [23]]. Before we plan for surgical excision or biopsy or FNAC of slowly growing soft tissue is present in a patient from endemic area, diagnosis of hydatid disease should be excluded to avoid leakage of cyst contents, risk of anaphylaxis and secondary hydatidosis. ELISA is 80–100% sensitive and 88–96% specific for hydatid liver disease but less sensitive for lung (50–56%) or other organ involvement (25–26%).(²⁵). Ultrasonography (US) should be the first investigation used for detection of hydatid disease of soft tissue [²²]. MRI is the diagnostic tool of choice for intramuscular or subcutaneous hydatidosis because of detailed information about soft tissue structure and relationship.(¹¹). Surgical resection in toto is the definitive curative treatment of choice(¹²,²²6)

References

- 1. Salih A.M., Kakamad F.H., Hammood Z.D., Yasin B., Ahmed D.M. Abdominal wall hydatid cyst: a review a literature with a case report. Int. J. Surg. Case Rep. 2017;37:154–156. [PMC free article] [PubMed] [Google Scholar]
- 2. Ahmed A.E. A solitary primary subcutaneous hydatid cyst in the thigh: a case report. Int. Surg.
- J. 2016;3(1):411–414. [Google Scholar]
- 3. Gougoulias NE, Varitimidis SE, Bargiotas KA, Dovas TN, Karydakis G, Dailiana ZH. Skeletal muscle hydatid cysts presenting as soft tissue masses. Hippokratia. 2010;14:126–30. [PMC free article] [PubMed] [Google Scholar]
- 4. Hamdi MF, Touati B, Abid A. Primary hydatid cyst of the biceps femoris. Musculoskelet Surg. 2010;94:59-
- 61. [PubMed] [Google Scholar]
- 5. Vicidomini S, Cancrini G, Gabrielli S, Naspetti R, Bartoloni A. Muscular cystic hydatidosis: Case report. BMC Infect Dis. 2007;7:23. [PMC free article] [PubMed] [Google Scholar]
- 6. Salih A.M., Kakamad F.H., Rauf G.M. Isolated hydatid cyst of the diaphragm, a case report. Int. J. Surg. Case Rep. 2016;29:130–132. [PMC free article] [PubMed] [Google Scholar]
- 7. Mustafa H.M., Muhammad N.H., Kakamad F.H., Essa R.A., Rahem H.M. Hydatid disease causing paraplegia: a case report with literature review. Edorium J. Infect. Dis. 2017;3(1):17–20. [Google Scholar]
- 8. Baram A., Kakamad F.H., Alwan A.A. Primary posterior mediastinal hydatid cyst mimicking malignant mediastinal neurogenic tumor. Int. J. Case Rep. Imag. 2014;5(1):54–57. [Google Scholar]
- 9. Abebe A.A. Hydatid cyst of the left thigh: a case report. East Cent. Afr. J. Surg. 2010;15(1):139-
- 140. [Google Scholar]
- 10. Argy N., AbouBacar A., Boeri C., Lohmann C., Pfaff A.W., Hansmann Y. Primary musculoskeletal hydatid cyst of the thigh: diagnostic and curative challenge for an unusual localization. Can. J. Infect. Dis. Med. Microbiol. 2013;24(3):99–101. [PMC free article] [PubMed] [Google Scholar]
- 11. Bansiwal R.K., Sharma R., Attri A.K. A large primary hydatid cyst of thigh: a case report. Indian J. Surg. 2011;73(²):158–160. [PMC free article] [PubMed] [Google Scholar]

- 12. Barkati S., Butler-Laporte G., Ndao M., Ajise O.K., Semret M., Yansouni C.P. A recurrent hydatid cyst of the thigh diagnosed 13 years after initial presentation. IDCases. 2018;11:12–15. [PMC free article] [PubMed] [Google Scholar]
- 13. Duygulu F., Karaoglu S., Erdogan N., Yildiz O. Primary hydatid cyst of the thigh: a case report of an unusual localization. Turk. J. Pediatr. 2006;48(3):256. [PubMed] [Google Scholar]
- 14. Garagnani L., Sudanese A., Rimondi E., Bacchini P. Primary hydatid cyst of the root of the thigh: a case report and review of the literature. La Chirurgiadegliorgani di movimento. 2008;92(²):113
- 118. [PubMed] [Google Scholar]
- 15. Goel A., Dewanda N.K. Hydatid cyst of thigh: a rare presentation. J. Evol. Med. Dent. Sci. 2013;2(40):7761–7764. [Google Scholar]
- 16. Gupta A., Singal R.P., Gupta S., Singal R. Hydatid cyst of thigh diagnosed on ultrasonography—a rare case report. J. Med. Life. 2012;5(²):196. [PMC free article] [PubMed] [Google Scholar]
- 17. Kazmi Z., Qureishi S., Quraishy M.S., Mallick F.A., Rizvi S. Atypical presentation of hydatid cyst in the thigh. J. Coll. Phys. Surg. 2017;27(1):51–52. [PubMed] [Google Scholar]
- 18. Pathak T.K., Roy S., Das S., Achar A., Biswas A.K. Solitary hydatid cyst in thigh without any detectable primary site. JPMA—J. Pak. Med. Assoc. 2011;61(12):1244. [PubMed] [Google Scholar]
- 19. Landolsi M., Kouki S., Abdennadher A. Hydatid cyst of the thigh: a challenging diagnosis. BMJ Case Rep. 2017;1(1):1–2. [Google Scholar]
- 20. Madhar M., Aitsoultana A., Chafik R., Elhaoury H., Saidi H., Fikry T. Primary hydatid cyst of the thigh: on seven cases. Musculoskelet. Surg. 2013;97(1):77–79. [PubMed] [Google Scholar]
- 21. Mourafiq O., Echchaoui A., Chafri B., Bouabid A.S., Benchebba D., Boussouga M. Primary hydatid cyst: an unusual cause of a mass in the thigh. BMH Med. J. 2017;4(4):139–141. [Google Scholar]
- 22. Perelman H., Gottlieb L. Hydatid cyst in muscle of the thigh. Calif. Med. 1961;94(2):101. [PMC free article] [PubMed] [Google Scholar]
- 23. Sarda D.K., Lohiya S.J., Jawandiya V.G., Kamble A.T. Migration of retroperitoneal hydatid cyst into the thigh: a rare presentation. Indian J. Surg. 2006;68(5) [Google Scholar]
- 24. Shaikh R. Primary musculoskeletal hydatid cyst of thigh. J. Islamabad Med. Dent. Coll. 2017;6(2):113-
- 115. [Google Scholar]
- 25. Charles King H. Cestodes. In: Mandell GL, Bennett JE, Dolin R, editors. Principles and practice of infections diseases. 4. NY: Churchill Livingstone Inc; 1995. pp. 2543–2553. [Google Scholar]
- 26. Garcia-Alvarez F, Torcal J, Salinas JC, Navarro A, Garcia-Alvarez I, et al. Musculoskletal hydatid disease: a report of 13 cases. Acta Orthop Scand. 2002;73:227–231. doi: 10.1080/000164702753671858. [PubMed] [CrossRef] [Google Scholar]
- 27. Memis A, Arkun R, Bilgen I, Ustun EE. Primary soft tissue hydatid disease: report of two cases with MRI characteristics. Eur Radiol. 1999;9:1101–1103. doi: 10.1007/s003300050798. [PubMed] [CrossRef] [Google Scholar]