Chirurgia (2010) 105: 419-422
Nr. 3, Mai - Iunie

Primary hydatid cyst with an unusual location - a case report

F. Săvulescu[1], I-I. Iordache[1], R. Hristea[2], C. Dumitru[3], A-M. Șandru[4], G. Bălașa[1], R. Pătrușoiu[1], M. Yazyi[1], C. Iordache[5]

[1]Second Surgical Department, Carol Davila Central Military Hospital, Bucharest, Romania
[2]Surgical Department, National Institute of Aeronautical and Space Medicine, Bucharest, Romania
[3]Surgical Department, Ştefan Milcu Medical Center for Diagnostic and Treatment, Bucharest, Romania
[4]Department of Internal Medicine, Nicolae Cajal Hospital, Bucharest, Romania
[5]University of Medicine and Pharmacy ,,Carol Davila", Bucharest, Romania

Rezumat

Chist hidatic primar cu localizare rară – prezentare de caz

Echinococoza chistică, o zoonoză provocată de infecția cu Echinococcus granulosus, reprezintă încă o problemă de sănătate publică în zonele endemice (China, Orientul Mijlociu, regiunea Mediteraneană, America de Sud, Federația Rusă). Echinococoza primară se poate dezvolta în aproape orice organ (ficat, plamâni, rinichi, splină, mediastin, inimă, creier, os, pancreas, sân, ovar, etc.). Ficatul şi plămânii sunt organele cel mai frecvent implicate. Chiar şi în zonele endemice localizarea hidatidozei primare la nivelul țesuturilor moi este extrem de rară. Scopul acestui articol este de a prezenta această patologie rară. O femeie în vârstă de 46 de ani s-a adresat serviciului nostru de chirurgie cu o formațiune rotundă, de 7/8 cm, nedureroasă, localizată subcutanat în regiunea anteromedială proximală a coapsei drepte. Pe baza investigațiilor clinice, de laborator şi imagistice s-a suspicionat diagnosticul de chist hidatic. S-a practicat intervenția chirurgicală, asociată cu administrarea de substanțe antihelmintice intraoperator şi Albendazole postoperator. După o recuperare lipsită de complicații pacienta a fost externată în cea de a-7-a zi postoperator. După o perioadă de un an de urmărire nu au fot decelate recurențe locale sau sistemice.

Cuvinte cheie: echinococoza chistică umană

Abstract

Human cystic echinococcosis, a zoonotic infection caused by Echinococcus granulosus, is still a largely extended public health problem in endemic regions (China, Middle East, Mediterranean region, South America, Russian Federation, etc.). Primary echinococcosis may develop in almost any organ (liver, lung, kidney, spleen, mediastinum, heart, brain, bones, pancreas, breast, ovaries, etc.). The liver and the lungs are the most frequently involved organs. Primary hydatid disease of the soft tissue is extremely rare, even in endemic areas. The paper will be focused on analyzing this rare disease. A 46-years old woman who came to our Department of Surgery with a 7/8 cm painless, round, palpable mass in the subcutaneous tissue of the proximal anteromedial side of the right thigh. Based on clinical and laboratory findings and imaging techniques we suspected a hydatid cyst. Conservative surgery associated with antihelminthic substances intraoperative and Albendazole postoperative was performed. After an uneventful recovery the patient was discharged 7 days after operation. No local or systemic recurrences were detected during 1 year follow up.

Key words: human cystic echinococcosis

Corresponding author: Săvulescu Alexandru Florin
Central Military Hospital
Second Surgical Department
Str. Mircea Vulcănescu, nr. 88, Sector 1,
010825, Bucharest, Romania
E-mail: savfl@yahoo.com

Introduction

Human echinococcosis is a zoonotic infection caused by the larval stage of Echinococcus granulosus, Echinococcus multilocularis, Echinococcus vogeli and Echinococcus oligarthrus.

Cystic echinococcosis is caused by the metacestode stage of E. granulosus. (1) E. granulosus are small tapeworms (2-7 mm), found on all continents, with high prevalence in China, Middle East, Mediterranean region, South America and the Russian Federation. (2) These tapeworms have intermediate (humans, sheep, pigs, etc.) and permanent hosts (dogs and other carnivores). The definitive hosts pass eggs in their feces. After the ingestion of eggs by the intermediate host, embryos penetrate the intestinal mucosa and migrate to the liver via the portal circulation. (2) Humans acquire the E. granulosus eggs through oral uptake. Primary echinococcosis may involve all anatomic sites, but the liver and the lungs are the most frequently infected organs. (3-5) Other organs that may be also affected are: the urinary tract, the spleen, the mediastinum, the heart, the brain, the bones, the pancreas, the breast, the ovaries, etc. (6-13)

The primary subcutaneous hydatid cyst is very rare. (14-31) Diagnosis in cystic echinococcosis is based on clinical grounds and confirmed either by imaging (US, CT, X-ray) or immunodiagnostic tests (ELISA, etc). (32,33)

Case Report

A 46-year-old woman came to our Department of Surgery with a history of a 3-month painless palpable mass in the upper anteromedial site of the right thigh which had slowly enlarged without any symptoms. The patient had no history of pain, trauma, fever, weigh loss or surgery for a hydatid cyst. During the physical examination the abdomen was soft, painless, mobile and no abdominal masses were palpated. A 7/8 cm fluctuant, slight, painless, loose mass adherent to the skin was palpated in the proximal anteromedial site of the right thigh. No local bruit was heard. The overlying layer of the skin and the lymph nodes were normal. No other abnormalities were noted on the physical examination.

A complete blood count was normal with a normal differential count, the erythrocyte sedimentation rate was normal; none of the routine laboratory findings reveal any abnormality. Both posteroanterior chest radiography and abdominal ultrasonography were normal. Computed tomography showed no involvement of the brain, lung or abdominal cavity. Ultrasonographic examination detected a 57/34 mm unilocular, round, well-circumscribed bordered, hypoechoic cystic structure with multiple daughter cysts, in the subcutaneous tissue of the proximal anteromedial aspect of the right thigh - a lesion resembling a hydatid cyst (Fig. 1A and B: Ultrasonographic view of primary subcutaneous hydatic disease – a round well-circumscribed, bordered, hypoechoic structure with multiple small daughter cysts)

In light of the physical examination, both laboratory findings and imaging studies suspected a hydatid cyst. Considering the risks and benefits we indicated the surgical treatment associated with antihelminthic drugs. A longitudinal incision had been made and a meticulous dissection was performed without injuring any vasculo-nervous structures or adjacent muscles. A cystic mass was discovered. The operative field was packed with swabs soaked with alcohol then the cyst was punctured with a needle and decompressed. The cyst content was aspirated with a cannula and replaced by alcohol for sterilization. After opening of the cyst, its content was removed. The cyst wall was excised along with the adjacent muscles. Thus the cystic mass was excised completely (Fig. 2A and B: The operative field, the hydatid cyst excised from thigh; lesion diameter approximately 6 cm. The surgical specimen containing germinative membrane and multiple small daughter cysts). After cystectomy, the cavity was covered with sponges soaked with Providone-jodine. After the wound was closed, a tube for drainage was kept for 3 postoperative days. Histopathologic examination of the specimen revealed a hydatid cyst. Treatment with oral Albendazole 10 mg/kg/day for 3 months was used to reduce the risk of recurrence. After an uneventful recovery the patient was discharged 7 days after operation. No local or systemic recurrences were detected during a 1 year follow-up.

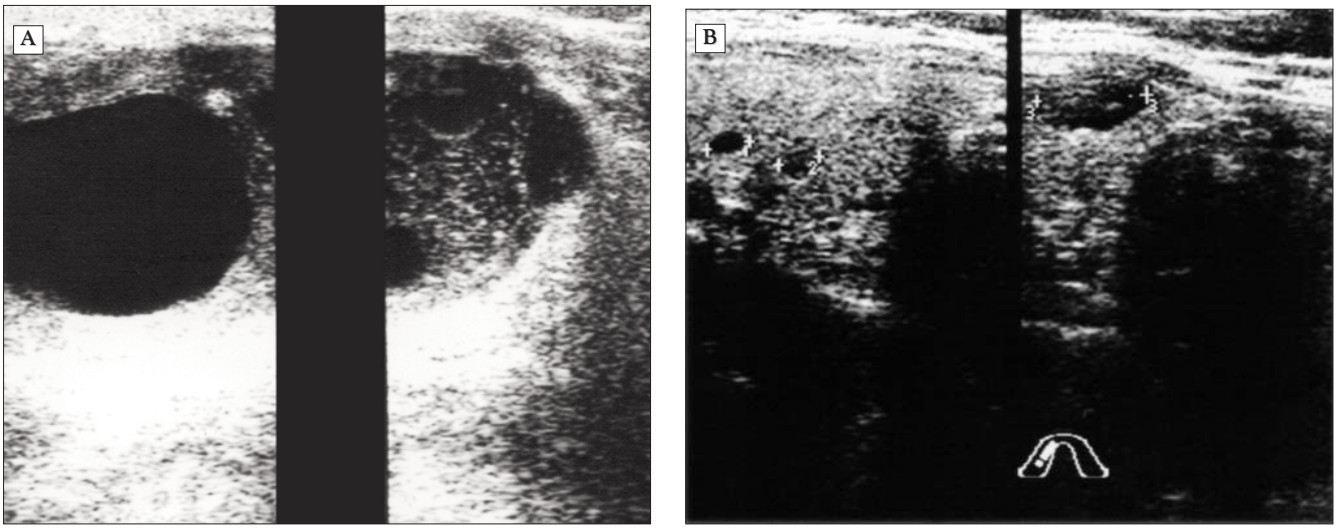

Figure 1 A, B. *Ultrasonographic view of primary subcutaneous hydatic disease – a round well-circumscribed, bordered, hypoechoic structure with multiple small daughter cysts*

Figure 2 A, B. *The operative field, the hydatid cyst excised from thigh; lesion diameter approximately 6 cm. The surgical specimen containing germinative membrane and multiple small daughter cysts*

Discussion

Human echinococcosis causes significant morbidity and mortality especially in endemic regions. Subcutaneous tissue is a very rare site for the primary hydatid cyst (the incidence is 0.2-2%). How the solitary hydatid cyst occurs in subcutaneous tissue-an unusual site-passing through liver and lung filter remains unclear, lymphatic route is probably involved. (1,14-31)

The process of diagnosis of human cystic echinococcosis can be sometimes very problematic as the clinical manifestations are highly variable and uncharacteristic. When present, clinical symptomatology depends on: the organ involved, the size of the cysts, local mass effect on adjacent structures or complications like rupture, local inflammation, bacterial infection, obstruction. (32,34) Imaging methods (US, CT, X-ray, MRI) are commonly the primary approaches. Routine laboratory blood work is nonspecific (eosinophilia, leukocytosis, hypogammaglobinemia, elevated bilirubin or alkaline phosphatase level). Indirect hemagglutination test and ELISA are the initial screening tests, immunodiffusion and immunoelectrophoresis are used to confirm the reactivity. Immunodiagnostic procedures are used for the etiological confirmation or for differential diagnosis. (32) The differential diagnosis in our case included benign and malign soft tissue tumors, cysts, abscesses, hematomas, aneurysms, etc. (35,36)

The therapy of choice is complete excision without opening the cyst. Sometimes localization of the cyst and the risk of invasion in adjacent structures contraindicate complete excision, in our case we first puncture the cyst, aspirate cyst fluid, inject protoscolicidal substance (alcohol), re-aspire after 20 minutes the new cyst-content, then we remove the germinative membrane. (37) To prevent recurrence we irrigated the surgical area with protoscolicidal agents. Intraoperative or postoperative associated morbidity (hemorrhage, infectious, mechanical damage of adjacent tissues, anaphylactic reactions) and recurrence were avoided using adequate surgical techniques and protoscolicidal substances intraoperative, and Albendazole for 3 months postoperative. Adverse reactions associated with Albendazole treatment (transaminase elevation, abdominal pain, headache, fever, nausea, etc) were not detected. (38)

References

1. Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev. 2004;17(1):107-35.
2. White C, Weller P. Cestodes. In: Fauci A, et al editors. Harrison's principles of internal medicine, 17th ed. McGraw-Hill; 2008. p. 1213-1336.
3. Safioleas MC, Misiakos EP, Kouvaraki M, Stamatakos MK, Manti CP, Felekouras ES. Hydatid disease of the liver a continuing surgical problem. Arch Surg. 2006;141(11):1101-8. Comment in: Arch Surg. 2007;142(7):690; author reply 690-1.
4. Karaoglanoglu N, Kurkcuoglu IC, Gorguner M, Eroglu A, Turkyilmaz A. Giant hydatid lung cysts. Eur J Cardiothorac Surg. 2001;19(6):914-7.
5. Rădulescu S. Cestode. Echinococcus granulosus. În: Rădulescu S, editor. Parazitologie Medicală. Ed. All Educational; 2000. p. 282-295.
6. Perimenis P, Athanasopoulos A, Gyftopoulos K, Barbalias G. Primary echinococcal disease of the kidney: the case for a more conservative approach. Int Urol Nephrol. 2001;32(4): 609-13.
7. Franquet T, Montes M, Lecumberri FJ, Esparza J, Bescos JM. Hydatid disease of the spleen: imaging findings in nine patients. AJR Am J Roentgenol. 1990;154(3):525-8.
8. Eroğlu A, Kürkçüoğlu C, Karaoğlanoğlu N, Tekinbaş C, Kaynar H, Onbaş O. Primary hydatid cysts of the mediastinum. Eur J Cardiothorac Surg. 2002;22(4):599-601. Comment in: Eur J Cardiothorac Surg. 2003;23(2):257-8. author reply 258.

9. Rezaian GR, Aslani A. Endocardial hydatid cyst: a rare presentation of echinococcal infection. Eur J Echocardiogr. 2008;9(2): 342-3.
10. Guzel A, Tatli M, Maciaczyk J, Altinors N. Primary cerebral intraventricular hydatid cyst: a case report and review of the literature. J Child Neurol. 2008;23(5):585-8. Epub 2008 Jan 11.
11. Song XH, Ding LW, Wen H. Bone hydatid disease. Postgrad Med J. 2007;83(982):536-42.
12. Ilica AT, Kocaoglu M, Zeybek N, Guven S, Adaletli I, Basgul A, et al. Extrahepatic abdominal hydatid disease caused by Echinococcus granulosus: imaging findings. AJR Am J Roentgenol. 2007;189(2):337-43.
13. Yuksel BC, Ozel H, Akin T, Avsar FM, Hengirmen S. Primary hydatid cyst of the breast with elevated CA 19-9 level. Am J Trop Med Hyg. 2005;73(2):368-70.
14. Orhan Z, Kara H, Tuzuner T, Sencan I, Alper M. Primary subcutaneous cyst hydatic disease in proximal thigh: an unusual localisation: a case report. BMC Musculoskelet Disord. 2003; 4:25.
15. Atmatzidis K, Koutelidakis I, Papaziogas B, Alexandrakis A, Chatzimavroudis G, Grigoriou M, et al. Primary hydatid cyst of the thigh. Chirurgia (Bucur). 2006;101(4):419-21.
16. Sarisoy HT, Memisoglu K, Tamer GS, Sarlak AY. Primary hydatid disease in adductor muscles. Clin Invest Med. 2008; 31(5):E296-9.
17. Vasilevska V, Zafirovski G, Kirjas N, Janevska V, Samardziski M, Kostadinova-Kunovska S, et al. Imaging diagnosis of musculoskeletal hydatid disease. Prilozi. 2007;28(2):199-209.
18. Cankorkmaz L, Ozturk H, Koyluoglu G, Atalar MH, Arslan MS. Intermuscular hydatid cyst in a 4-year-old child: a case report. J Pediatr Surg. 2007;42(11):1946-8.
19. 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.
20. 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.
21. Marwah S, Subramanian P, Marwah N, Rattan KN, Karwasra RK. Infected primary intramuscular echinococcosis of thigh. Indian J Pediatr. 2005; 72(9):799-800.
22. Kocakusak A, Koyuncu A, Arikan S, Senturk O. Primary hydatid cyst of vastus lateralis muscle. Acta Chir Belg. 2004; 104(4):471-2.
23. Koc Z, Ağildere AM, Yalcin O, Pourbagher A, Pourbagher M. Primary hydatid cyst in the anterior thigh: Sonographic findings. J Clin Ultrasound. 2004; 32(7):358-60.
24. Keskin D, Ezirmik N, Karsan O, Gürsan N. Primary hydatidosis of the gracilis muscle in a girl. J Int Med Res. 2002; 30(4):449-51.
25. Alexiadis G, Lambropoulou M, Deftereos S, Papadopoulos N, Manavis J. Primary muscular hydatidosis. US, CT and MR findings. Acta Radiol. 2002; 43(4):428-30.
26. Merkle EM, Schulte M, Vogel J, Tomczak R, Rieber A, Kern P, Goerich J, et al. Musculoskeletal involvement in cystic echinococcosis: report of eight cases and review of the literature. AJR Am J Roentgenol. 1997;168(6):1531-4.
27. 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.
28. Chevalier X, Rhamouni A, Bretagne S, Martigny J, Larget-Piet B. Hydatid cyst of the subcutaneous tissue without other involvement: MR imaging features. AJR Am J Roentgenol. 1994;163(3):645-6.
29. Ok E, Sözüer EM. Solitary subcutaneous hydatid cyst: a case report. Am J Trop Med Hyg. 2000;62(5):583-4.
30. Voucharas C, Papaioannidis D, Papamichael K. Subcutaneous mass of the right thigh. Postgrad Med J. 1998;74(871):287-8.
31. Vicidomini S, Cancrini G, Gabrielli S, Naspetti R, Bartoloni A. Muscular cystic hydatidosis: case report. BMC Infect Dis. 2007;7:23.
32. Pawlowski ZS, et al. Echinococcosis in humans: clinical aspects, diagnosis and treatment. In: Eckert J, editor. WHO/OIE Manual on Echinococcosis in Humans and Animals: a Public Health Problem of Global Concern. Paris: World Organisation for Animal Health. 2001. p. 2-20.
33. Lewall DB. Hydatid disease: biology, pathology, imaging and classification. Clin Radiol. 1998;53(12):863-74.
34. King C. Cestode infections. In: Goldman L, editor. Cecil Medicine, 23rd ed. 2008; p. 375-2413
35. Christopher DM, Fletcher K, Krishnan U, Fredrik M. Pathology and genetics of tumours of soft tissue and bone. Lyon: IARC Press; 2002.
36. Hălălău F, Ardeleanu C. Morfopatologia tumorilor benigne şi maligne. În: Anatomia patologică, vol I. Bucuresti: Ed. Medicală; 2003. p. 9-157.
37. WHO Informal Working Group on Echinococcosis. Guidelines for treatment of cystic and alveolar echinococcosis in humans. Bull World Health Organ. 1996;74(3):231-42. [Article in English, French]
38. Opatrny L, Prichard R, Snell L, Maclean JD. Death related to albendazole-induced pancytopenia: case report and review. Am J Trop Med Hyg. 2005;72(3):291-4.
39. Peláez V, Kugler C, Correa D, Del Carpio M, Guangiroli M, Molina J, et al. PAIR as percutaneous treatment of hydatid liver cysts. Acta Trop. 2000;75(2):197-202.
40. Smego RA Jr, Sebanego P. Treatment options for hepatic cystic echinococcosis. Int J Infect Dis. 2005;9(2):69-76.