

RADIOLOGIC FINDINGS AND PERCUTANEOUS TREATMENT OF A RARE GIANT SOFT TISSUE HYDATID CYST

C. Yucesoy, E. Ozturk, B. Hekimoglu¹

The aim of this report is to present the radiologic findings and discuss the percutaneous treatment of an extremely rare giant soft tissue hydatid cyst located in the left thigh in a 44-year-old woman. Ultrasound showed a well circumscribed giant soft tissue cyst, and computed tomography revealed a well defined unilocular hypodense cystic mass originating from the adductor muscles and extending through the subcutaneous tissue without bony relations. On magnetic resonance imaging, the cystic mass was hypointense on T1 and hyperintense on T2-weighted images. Percutaneous treatment was performed successfully by catheterization technique with hypertonic saline and alcohol. No complications and recurrence were observed during the procedure, in the postprocedure or in the follow up period.

Key-word: Echinococcosis.

Hydatidosis is an infectious disease caused by the tapeworm, *Echinococcus granulosus*. Echinococcosis has a worldwide distribution and has its highest prevalence especially in the sheep-raising areas such as East Europe, Asia, Middle East, Australia, and South America (1-3). The liver is the most frequently (50-75%) involved organ, whereas the lungs are the second most common site of infection (15-25%) followed by the rest of the body (10%). Muscular involvement is extremely rare (0.5%-4.7%), and it has been accepted that muscle is an unfavourable site for the parasite because of the presence of high lactic acid levels. The most common muscular hydatidosis locations are the trunk muscles and the proximal limbs (4-7).

The traditional treatment of Echinococcosis is surgery. In addition, medical therapy such as Benzimidazole derivatives given either alone or as a supplementary treatment is currently used in cystic hydatid disease. Owing to the developments in percutaneous treatment, the percutaneous approach has also been reported as a safe and effective alternative treatment (2, 8-10).

Here we present the radiologic findings of an extremely rare giant soft tissue hydatid cyst, located in the left thigh originating from the adductor muscles and extending through the subcutaneous tissue treated with percutaneous approach. The aim of this case report is to emphasise and discuss the radiological features and percutaneous approach

to the hydatid cyst located in a soft tissue.

Case report

A 44-year-old female patient was admitted to our hospital with a slowly growing painless lump in the left thigh for the past one year and claimed that she had restricted range of motion and difficulty in walking as the lump rubbed the other leg. She had no history of trauma, surgery, or additional diseases. Physical examination revealed a non-tender fixed mass, measuring approximately 20 cm. There were no signs of inflammation or skin changes. The only significant haematological finding was positive hemagglutination test for *E. Granulosus* with a titer of 1/256 (Echinococcosis IHA, Fumouze Diagnostics, Levallois Perret, France: n.v. <1:160). Chest X-ray and abdominal sonography findings were normal. Ultrasound (US) examination demonstrated a well circumscribed giant unilocular cystic mass with a relatively thick wall without splitting membrane or daughter cysts (smaller cysts within a cyst) located in the subcutaneous fat layer. For further evaluation, computed tomography (CT) and magnetic resonance imaging (MRI) were planned. CT demonstrated a well-defined large hypodense cystic mass originating from adductor muscles with a narrow neck located in the medial part of the left thigh with intact bony structure (Fig. 1). The MRI showed a 20 x 18 x 10 cm giant pure cystic

mass hypointense on T1- and hyperintense on T2-weighted images with a low intensity peripheral rim on both sequences. The cystic mass was neighbouring and displacing gracilis and adductor muscles, and no daughter cysts, water lily sign, or splitting of the wall were diagnosed (Fig. 2). In the light of the imaging and laboratory findings, a hydatid cyst located in the soft tissue was strongly suspected, and percutaneous puncture and catheterization were planned for accurate diagnosis and treatment.

Before percutaneous treatment in order to prevent secondary dissemination, the patient received albendazole (Andazol; Biofarma, Istanbul, Turkey) in a 10 mg/kg dose per day for 15 days before and after the procedure. The patient was monitored for anaesthesia for possible risk of anaphylaxis, and as a premedication for hypersensitivity reactions, prednisolon 25 mg (Fako; Istanbul, Turkey) and pheniramine maleate 50 mg (Avil; Hoechst, Frankfurt, Turkey) were administered intravenously. Both drugs were given 20 min before the procedure, and the same dose of pheniramine maleate was repeated 12 hours later.

The procedure was performed under sonographic and fluoroscopic guidance. As the diameter of the cyst was over five cm, the patient was treated by catheterization technique with hypertonic saline and alcohol. Using the ellipsoid formula (volume = height x width x length x 0.523), the cyst volume was calculated as 1800 cc. After the cyst cavity was punctured with a 18-gauge needle, half of the estimated cyst fluid (900 cc) was aspirated gradually, and at an amount equal to 70% of aspirated fluid (630 cc), hypertonic saline (20-30 NaCl) mixed with contrast material was injected. After separation of the

From: 1. Department of Radiology, S.B. Ankara Diskapi Training and Research Hospital, Ankara, Turkey.

Address for correspondence: Dr C. Yucesoy, M.D., Mutluköy Sitesi 12. Sok. No. 17 Ümitköy Ankara, Türkiye. E-mail: yucecun2000@yahoo.com

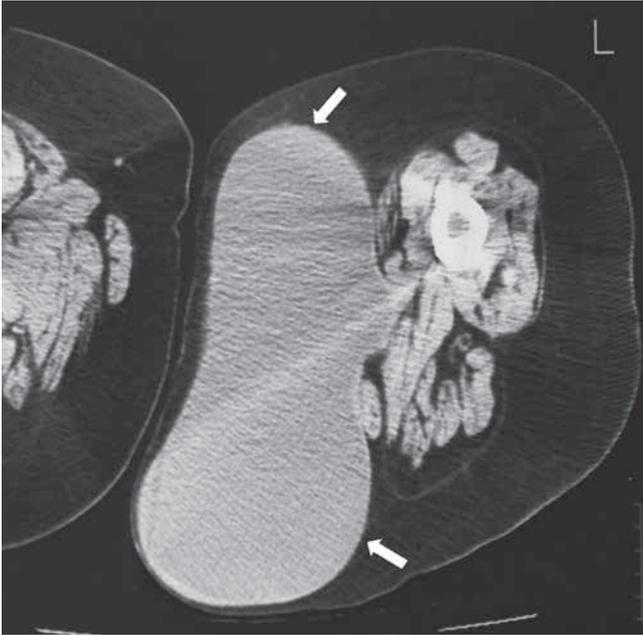


Fig. 1. — The CT scan of the left thigh in the axial plane showing a well-circumscribed unilocular giant cystic mass (arrows) originating from the adductor muscles and extending through the subcutaneous tissue. The cyst seems to have a narrow neck in the medial portion posterior to the vessels. Note that the bony structures are intact.

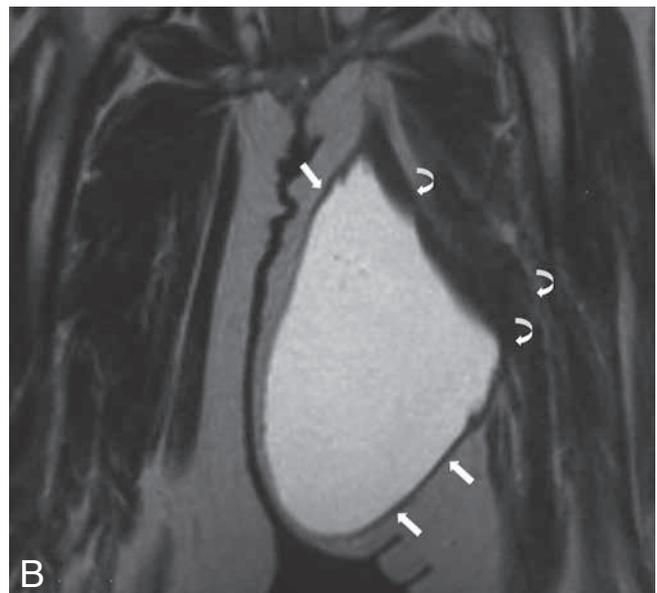


Fig. 2. — The MRI of the left thigh on the coronal plane showing a giant cystic mass hypointense on T1 (A) and hyperintense on T2 (B) weighted images with a hypointense rim on both sequences (arrows). Note that the gracilis and adductor muscles are displaced (curved arrows).

endocyst from the cyst wall, a 14 F pigtail catheter was placed (Fig. 3A) and all of the fluid in the cavity was evacuated. The cavity was left free for drainage.

The cyst fluid at the initial puncture was sent to microbiology for cytologic examination

Twelve days after the catheterization a cystography was performed to confirm no leakage out of the cavity (Fig. 3B). No leakage was observed, and equal to half the volume of the aspirated material (450 cc), 95% alcohol was infused as a sclerosant agent and kept in the cavity for 20 minutes. Meanwhile, the patient was placed in supine, prone, right, and left decubitus positions at every 5 minutes. Afterwards, the alcohol was drained; the cavity was irrigated with normal saline, and the catheter was removed. The patient was discharged and asked to visit six months later for follow up. In the follow up period as the patient was living in the rural area, US follow up was achieved ten months later at the local hospital. The local radiology specialist claimed that the patient had no difficulty in walking and the mass volume was reduced and measured 10 x 8 x 5 cm in size on ultrasound.

Discussion

Hydatid disease is a worldwide problem and can develop anywhere in the body. However, the liver and the lungs are the most commonly involved organs, whereas muscles are considered an unfavourable site for the disease because of high lactic acid levels (4-7). This might be the real basis for the rarity of the primary disease located within the muscles. In hydatid disease, the unusual loca-

tions are generally involved as a result of spread from hepatic or pulmonary foci. In the present case, the cyst was primary and solitary and was in a giant form of skeletal hydatidosis without any primary foci.

US, CT and MRI can be used as a diagnostic tool for soft tissue hydatid cyst. US is a non-invasive, cheap, repeatable imaging modality; it is a widely used and accepted modality in the diagnosis of the disease. The classification of the cyst can be done

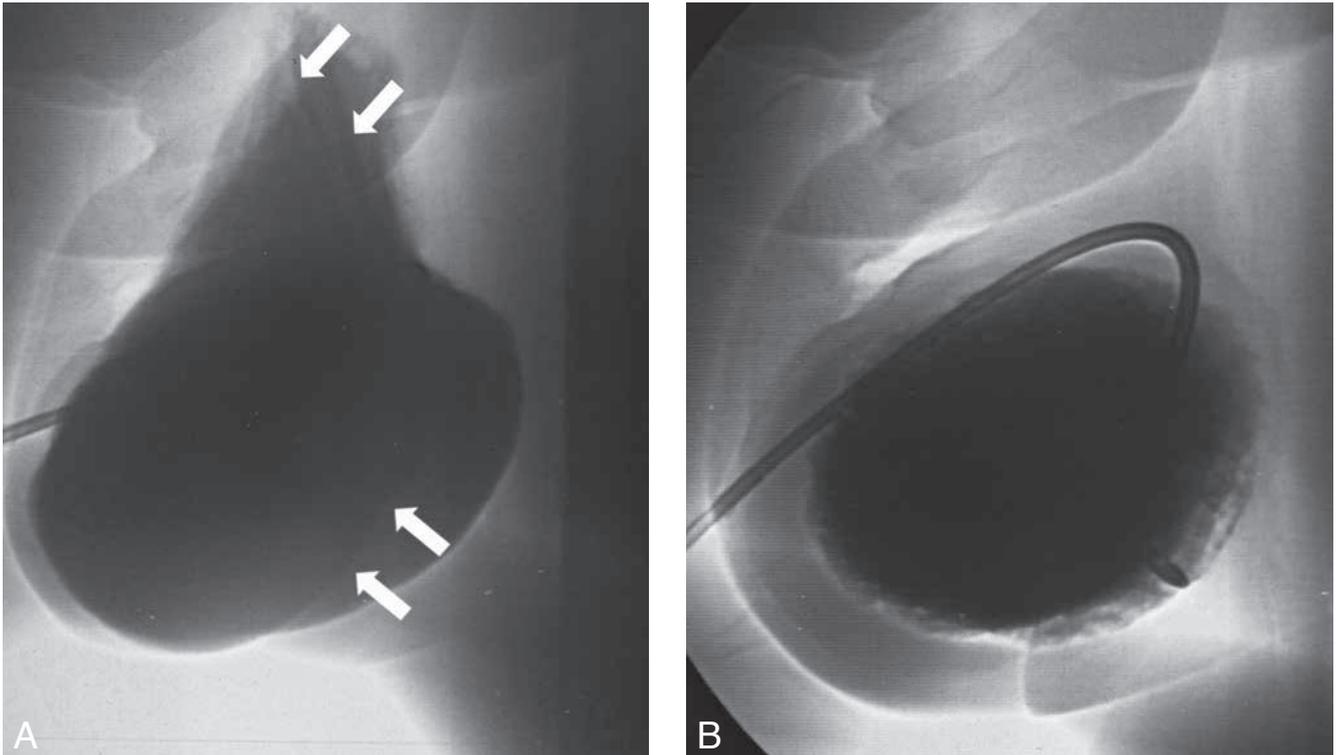


Fig. 3. — The cystogram during the procedure presenting detachment of the membrane (arrows) and 14 F pigtail catheter in the cavity (A). The cystogram before the sclerosant therapy (B): note that the cavity is intact and there is no leakage from the cavity.

according to the US criteria of Gharbi (11). Besides, US is a practical guide during interventional procedures. Daughter cysts, detached membranes, or double line sign are the most characteristic features of hydatid cyst on US (3-5, 7, 9-12). Although CT is superior in detecting wall calcifications and bony relations, the appearance of the cyst is rarely typical. On the other hand, presence of daughter cysts and detached membranes on CT can also help the diagnosis (3, 4, 7). In addition bony invasion is critical in soft tissue located cysts, and CT is advised to be performed as a part of screening and treatment protocols. MRI is the most important imaging modality especially in the detection and characterisation of soft tissue masses. Recent studies have defined the MR imaging findings of hepatic and extrahepatic hydatid cyst. MRI not only shows the cystic mass but also can reveal the typical features of hydatid disease such as the collapsed cyst membrane in the inferior portion of the cyst called 'water lily sign.' or daughter cysts. The cyst wall and septa may not always enhance on post-gadolinium T1-weighted images (13-17) but in the present case, as percutaneous treatment was planned for accurate diag-

nosis and treatment, contrast administration was not performed. Nevertheless, the cyst wall showed a surrounding peripheral hypoechoic rim both on T1 and T2 weighted images, which is considered a characteristic sign and is critical in the differential diagnosis, especially from nonparasitic epithelial cysts.

Although the traditional treatment of hydatid cyst is surgery, percutaneous treatment has become a serious alternative method in recent years. Percutaneous treatment used in many centres during the past decade has proven to be successful management of the disease. Furthermore, the morbidity and mortality rates, hospitalization times, and recurrence times are well-documented in the literature. One of the most important potential risks of percutaneous treatment is anaphylaxis. The anaphylactic reactions cannot be predicted before the procedure, and one should be prepared (monitoring the patient by anaesthesia and premedication) for this condition as it may occur during or after the procedure. The other unfavourable risk in percutaneous treatment is dissemination. However, percutaneous treatment produces a much smaller exposure surface for the cyst fluid than open surgery does. Besides, administra-

tion of benzimidazole derivatives before and after the procedure are known to be helpful in preventing dissemination. These factors may explain much lower recurrence rates and the absence of secondary dissemination with percutaneous treatment (9, 10, 12).

There are some critical points that must be taken into consideration during percutaneous treatment. For example, the catheterization must be done after the separation of the endocyst (the evidence of inactivation) in order to minimize the complications and to avoid the spillage of viable scolices. The major aim of the opacified hypertonic saline used with an amount equal to 70% of the evacuated fluid is to visualize the cyst cavity under fluoroscopy and to accomplish the guidance as well as US. Besides, it is important that the amount of the injected opacified hypertonic solution must be much more under the estimated volume in order not to cause dissemination by elevation of the intracystic pressure.

Literature reveals limited number of studies on percutaneous treatment of giant hydatid cysts or cysts located in the soft tissue. In a study on percutaneous treatment of giant hydatid cysts in the abdomen, Men et al treated 15 giant cysts and

despite prolonged catheterization and hospitalization, they did not encounter any recurrences (12). Likewise, in their limited number of studies, Akhan et al. treated six cysts in the soft tissue and reported no major complications or recurrences in 34.8-month follow-up period (5).

Although long catheterisation time is unfavourable in giant cysts, percutaneous treatment seems to be useful choice for the treatment of giant and hydatid cysts located in the soft tissue. In conclusion, despite some limitations in the present case, such as the giant volume leading prolonged catheterization, we believe that percutaneous treatment was a safe and an effective treatment option in this patient because no major complications were observed and the ambulation of the patient improved.

References

1. Lewall D.B., McCorkell S.S.: Hepatic echinococcal cysts: Sonographic appearance and classification. *Radiology*, 1985, 155:773-775.
2. Saidi F.: Surgery of Hydatid Disease. Philadelphia, Saunders, 1976, 3: 31-155.
3. Beggs I.: The radiology of hydatid disease. *Am J Roentgenol*, 1985, 145: 639-648.
4. Czermak B.V., Unsinn K.M., Gotwald T., Niehoff A.A., Freund M.C., Waldenberger P., et al.: Echinococcus granulosus revisited: Radiologic patterns seen in pediatric and adult patients. *Am J Roentgenol*, 2001, 177: 1051-1506.
5. Akhan O., Gumus B., Akinci D., Karcaaltincaba M., Ozmen M.: Diagnosis and percutaneous treatment of soft-tissue hydatid cysts. *Cardiovasc Intervent Radiol*, 2007, 30: 419-425.
6. Ammari F.F., Khasawneh Z., Salem M.K., Mesmar M., Omari H., Masa'deh R.: Hydatid disease of the musculoskeletal system. *Surgery*, 1998, 124: 934-937.
7. Merkle E.M., Schulte M., Vogel J., Tomczak R., Rieber A., Kern P., et al.: Musculoskeletal involvement in cystic echinococcosis: Report of eight cases and review of the literature. *Am J Roentgenol*, 1997, 168: 1531-1534.
8. El-On J.: Benzimidazole treatment of cystic echinococcosis. *Acta Trop*, 2003, 85: 243-52.
9. Akhan O., Ozmen M.N.: Percutaneous treatment of liver hydatid cysts. *Eur J Radiol*, 1999, 32: 76-85.
10. Men S., Hekimoğlu B., Yücesoy C., Arda I.S., Baran I.: Percutaneous treatment of hepatic hydatid cysts: an alternative to surgery. *AJR*, 1999, 172: 83-89.
11. Gharbi H.A., Hassine W., Brauner M.W., Dupuch K.: Ultrasound examination of hydatid liver. *Radiology*, 1981, 139: 459-463.
12. Men S., Yücesoy C., Edgüer T.R., Hekimoğlu B.: Percutaneous treatment of giant abdominal hydatid cysts: long-term results. *Surg Endosc*, 2006, 20: 1600-1606.
13. Marani S.A., Canossi G.C., Nicoli F.A., Alberti G.P., Monni S.G., Casolo P.M.: Hydatid disease: MR imaging study. *Radiology*, 1990, 175: 701-706.
14. von Sinner W., te Strake L., Clark D., Sharif H.: MR imaging in hydatid disease. *AJR*, 1991, 157: 741-745.
15. Chevalier X., Rhamouni A., Bretagne S., Martigny J., Larget-Piet B.: Hydatid cysts of the subcutaneous tissue without other involvement: MR imaging features. *Am J Roentgenol*, 1994, 163: 645-646.
16. Comert RB., Aydingoz U., Ucaner A., Arikan M.: Water-lily sign on MR imaging of primary intramuscular hydatidosis of sartorius muscle. *Skeletal Radiol*, 2003, 32: 420-423.
17. Memis A., Arkun R., Bilgen I., Ustun E.E.: Primary soft tissue hydatid disease: report of two cases with MRI characteristics. *Eur Radiol*, 1999, 9: 1101-1103.