Primary subcutaneous hydatid cyst of the leg: An unusual location and review of the literature

Sir,

Hydatid disease is an endemic zoonotic infestation caused by the larval form of *Echinococcus* species.[1,2] The most commonly afflicted organ is the liver but any tissue other than hair, nails and teeth may be involved.[3,4] Primary subcutaneous involvement is rare and in these cases, even with inflammatory signs, a painless mass is the usual presentation.[1,5] We describe a case of solitary subcutaneous hydatid cyst on the anterior aspect of the leg with clinical features of cellulitis.

A 34-year-old Iranian man presented with a painful erythematous swelling on the anterior aspect of his right leg for 5 months. At onset, he noticed purulent discharge from the swelling. He had taken multiple courses of antibiotics and had used 10% ichthyol ointment topically which led to a partial improvement in erythema but no change in the swelling. On examination, there was a tender, ill defined, skin-colored swelling of size 6 cm × 8 cm with two visible sinus tracts covered with hemorrhgic crusts [Figure 1]. Ultrasonography revealed a thick-walled collapsed cystic lesion in the superficial part of the anterior tibialis with a sinus tract extending into the underlying muscle. The wall thickness was 5 mm [Figure 2]. The lesion was excised in its entirety and macroscopic examination revealed a cystic mass. On histopathologic examination, the lesion showed a periodic acid–Schiff-positive laminated membrane with no germinal epithelium or protoscolex. Therefore, the diagnosis of a hydatid cyst was made [Figure 3]. The patient denied close contact with animals, prior history of trauma at the site or any previous surgery for hydatid cysts. Serum anti-*Echinococcus granulosus* immunoglobulin G (IgG) was positive. A thorough radiologic evaluation of other organs (including total bone scan and computed tomography of the chest, abdomen and pelvis) failed to detect a primary focus. Following excision, the patient received 400 mg of albendazole twice a day for 1 month and he remained free of recurrence at follow up 6 months later.

Echinococcus granulosus infection may remain asymptomatic for years or may manifest with dangerous complications such as infection, rupture, anaphylaxis and death.[5] We were able to find 55 reported cases of primary subcutaneous hydatid cysts [Table 1].[1,4,5]

Figure 1: Preoperative subcutaneous mass of the leg after antibiotic therapy

Figure 2: Ultrasonography of the cystic lesion

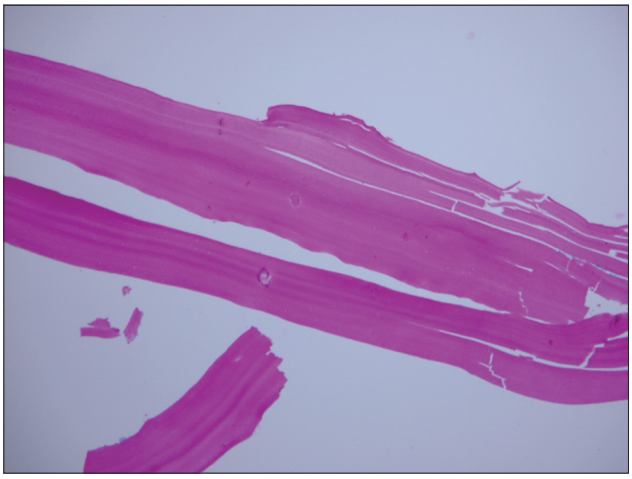

Figure 3: The periodic acid–Schiff-positive laminated wall, characteristic of Echinococcus cyst (periodic acid–Schiff stain, ×400)

However, we were unable to find any previous case of primary subcutaneous hydatid cyst located on the leg.

Subcutaneous hydatid cyst can be either secondary or primary. The secondary type is associated with an underlying primary focus of hydatid disease such as the liver.[2] On the other hand, the mechanism of primary subcutaneous localization is still not completely clear. There are two potential mechanisms: systemic dissemination via the lymphatic route or direct subcutaneous contamination through injured skin. In our patient, the first mechanism seems plausible as he had no history of skin injury.[6]

In a review of 22 cases of primary subcutaneous hydatid cysts, the age and sex distribution of patients was found to be similar to patients with liver hydatid cysts.[6] The most frequent subcutaneous location was

Table 1: Reported cases of primary subcutaneous hydatid disease with clinical details

Author, year	Sex/age	Region	Location	Physical examination	Treatment	Follow-up	Recurrence
Chevalier, 1994	Male/40	Créteil	Thigh	Slowly growing, painful mass	Surgery, postoperative albendazole treatment for 2 months	Not available	Not available
Voucharas, 1997	Female/50	Greece	Thigh	Painless, mobile mass	Surgery	Not available	Not available
Memis, 1999	Female/41	Turkey	Popliteal	Painless, slowly growing mass	Surgery	Not available	Not available
Ok, 2000	Female/12	Turkey	Submandibular	Fluctuant, painless, mobile mass	Surgery	4 years	No
Ozturk, 2001	Male/20	Turkey	Malar	Painless, firm, mobile mass	Surgery	14 months	No
Acar, 2001	Female/42	Turkey	Thigh	Painful, mobile mass	Surgery, preoperative albendazole treatment for 4 weeks	6 months	No
Baldi, 2002	Female/54	Italy	Scapula	Subcutaneous mass	Surgery	60 months	No
Arinc, 2003	Male/39	Turkey	Sternum	Painless mass	Surgery with adjuvant therapy	12 months	No
Orhan, 2003	Female/43	Turkey	Thigh	Painful, erythematous mass	Surgery, postoperative albendazole treatment for 1 month	12 months	No
Losanoff, 2004	Male/38	Columbia	Axillary region	Painless, palpable mass	Surgery	Not available	Not available
Guiral, 2004	Female/34	Segovia	Knee	Asymptomatic, mobile mass	Surgery, pre- and post-operative albendazole treatment for 1 and 8 weeks respectively	12 months	No
Koybasioglu, 2004	Female/22	Turkey	Infraumblical	Slowly growing mass	Surgery	Not available	Not available
Alouini, 2005	Female/41	Tunisia	Thigh	Slowly growing mass	Surgery	Not available	Not available
Kiyak, 2006	Female/62	Turkey	Inguinal	Slowly growing painful mass	Surgery	Not available	Not available
Gurbuz, 2006	Female/10	Eskişehir	Retroauricular	Fixed, slowly enlarging painless mass	Surgery, postoperative mebendazole chemotherapy for 2 months	12 months	No
Bedioui, 2007	Female/70	Tunisia	Hypogastric	Slowly growing mass	Surgery	Not available	Not available
Demirel, 2007	Female/73	Turkey	Subclavicular	Slowly growing mass	Surgery	6 months	No
Dogmus, 2007	Female/21	Turkey	Lumbar	Slowly growing, mobile mass	Surgery with adjuvant therapy	24 months	No
Daoudi, 2008	Female/21	Morocco	Gluteal	Slowly growing, semi-mobile mass	Surgery	36 months	No
Safioleas, 2008	Male/73	Greece	Gluteal	Mobile, slowly growing painless mass	Surgery, postoperative albendazole treatment for 4 months	3 years	No

Contd...

Table 1: Contd...

Author, year	Sex/age	Region	Location	Physical examination	Treatment	Follow-up	Recurrence
Parsak, 2008	Female/29	Turkey	Thigh	Painful, fixed, mass	Surgery, postoperative albendazole treatment for 6 months	12 months	No
Dirican, 2008	Male/64	Turkey	Thigh	Slowly growing mobile, painless mass	Surgery, postoperative albendazole treatment for 3 months	3 years	No
Dirican, 2008	Male/67	Turkey	Palm	Fixed and swelling mass	Surgery, postoperative albendazole treatment for 3 months	3 years	No
Gupta, 2008	Female/12	India	Shoulder	Painless, mobile, firm mass	Surgery	Not available	Not available
Gupta, 2008	Male/20	India	Back	Painless, mobile, firm mass	Surgery	Not available	Not available
Steurer, 2008	Female/57	Austria	Gluteal	Painless, fixed mass	Surgery, postoperative albendazole treatment for 4 weeks	Not available	No
Singal, 2010	Female/26	India	Thigh	Painless, mobile mass	Surgery, postoperative albendazole treatment for 3 months	12 months	No
Savulescu, 2010	Female/46	Romania	Thigh	Painless mass	Surgery, postoperative albendazole treatment for 3 months	12 months	No
Ozkan, 2010	Female/84	Turkey	Thigh	Painless mass	Died before surgery due to congestive heart failure	Not available	Not available
Iynen, 2011	Female/21	Turkey	Supraclavicular	Painless, mobile mass	Surgery, postoperative albendazole treatment for 4 weeks	Not available	Not available
Battyany, 2011	Male/63	Hungary	Popliteal	Painless, hyperemic, mobile mass	Surgery, pre- and post-operative mebendazole treatment	5 years	3 times
Sallami, 2011	Male/42	Tunisia	Lumbar	Painless, semi-mobile mass	Surgery	6 years	No
Ousadden, 2011	Female/70	Morocco	Abdominal wall	Painless, mobile mass	Surgery	2 years	No
Bansal, 2011	Male/42	India	Face	Painless, slowly growing mass	Surgery, postoperative albendazole treatment for 6 weeks	22 months	No
Pathak, 2011	Female/30	India	Thigh	Slowly growing mass	Surgery, pre- and post-operative albendazole treatment for 4 weeks	12 months	No
Mushtaque, 2012	Not available	India	Gluteal	Palpable lump	Surgery, postoperative albendazole treatment for 3 cycles (21 days each cycle)	Not available	Not available
Mushtaque, 2012	Not available	India	Not available	Palpable lump	Surgery, postoperative albendazole treatment for 3 cycles (21 days each cycle)	Not available	Not available
Rais, 2012	Female/58	Morocco	Scalp	Palpable mass	Surgery	Not available	Not available
Mahmoudi, 2012	Female/14	Morocco	Thigh	Painless, mobile mass	Surgery	24 months	No
Abhishek, 2012	Female/60	India	Abdomen	Painless swelling with dilated veins	Preoperative albendazole, surgery, postoperative albendazole+praziquantel for 3 months	6 months	No
Jarboui, 2012	Female/53	Tunisia	Supraclavicular	Erythematous, painful mass	Surgery, postoperative albendazole treatment for 8 weeks	4 months	No

Contd...

Table 1: Contd...

Author, year	Sex/age	Region	Location	Physical examination	Treatment	Follow-up	Recurrence
Ozdemir, 2012	Female/29	Turkey	Shoulder	Painful swelling	Surgery, postoperative albendazole for 3 cycles (28 days each cycle)	Not available	Not available
Gupta, 2012	Male/38	India	Thigh	Painful, mobile mass	Surgery, postoperative albendazole	8 months	No
Mirzaei, 2012	Male/54	Iran	Scapula	Slowly growing, slightly tender mass	Surgery	Not available	Not available
Burgazli, 2013	Male/63	Turkey	Abdomen	Slowly growing mass	Surgery, postoperative albendazole for 3 months	Not available	Not available
Ay, 2013	Female/53	Turkey	Temporomandible	Painful, mobile mass	Surgery, postoperative albendazole	6 months	No
Ay, 2013	Female/37	Turkey	Scapula	Slightly painful, mobile mass	Surgery, postoperative albendazole	6 months	No
Almadani, 2013	Male/53	Saudi Arabia	Thigh	Slowly growing, firm mass	Surgery	Not available	Not available
Okus, 2013	Not available	Turkey	Back	Not available	Surgery	Not available	Not available
Okus, 2013	Not available	Turkey	Face	Not available	Surgery	Not available	Not available
Yucesoy, 2013	Female/44	Turkey	Thigh	Giant, soft mass	Percutaneous treatment	Not available	Not available
Vecchio, 2013	Male/68	Catania	Shoulder	Slowly growing, mobile mass with occasional pain	Surgery, postoperative albendazole for 28 days	6 months	No
Haslak, 2014	Female/37	Turkey	Lumbar	Palpable mass	Surgery, postoperative albendazole for 3 months	1 year	No
Ekşi, 2014	Female/62	Turkey	Thoracic	Painful, palpable mass	Surgery, postoperative albendazole for 3 months	1 year	No
Demir, 2014	Male/7	Turkey	Chest wall	Painful mass	Surgery	Not available	Not available
Present case 2015	Male/34	Iran	Leg	Painful, erythematous slowly growing mass with clinical features of cellulitis	Surgery, postoperative albendazole for 28 days	6 months	No

the thigh followed by the gluteal region whereas the upper extremities were least commonly involved.[1,6] The usual presentation of subcutaneous hydatid cyst is in the form of a painless, non-inflammatory and slowly growing mass without any deterioration of the patient's general condition; but our patient initially presented with clinical signs of cellulitis.[3,7] Diagnosis of hydatid cysts, especially the subcutaneous form is very difficult in patients from non-endemic areas. Preoperative diagnosis is important because of the risk of anaphylactic reaction or local recurrence due to accidental leakage of contents of the cyst during surgery undertaken without a diagnosis; but these have not been reported in the subcutaneous type.[2,6] Radiologic investigations such as ultrasonography, magnetic resonance imaging and computed tomography are useful in the diagnosis of soft tissue masses and also to delineate the relationship of the cyst to adjacent structures.[6] Although serology is helpful particularly for liver hydatid cysts, it is negative in 79% of subcutaneous cysts; but our patient had a positive test. It is recommended that all patients with subcutaneous hydatid cyst must have a whole body screening to detect any additional foci.[4,6]

The main treatment of subcutaneous hydatid cyst is surgical excision. Subcutaneous cysts need less extensive surgery than visceral cysts and contrary to liver hydatidosis, complete excision of subcutaneous cysts is possible but they are more susceptible to rupture because of the difficulty of preoperative diagnosis.[2,6] In cases of rupture, the cyst pouch should be irrigated with protoscolocidal solutions and the patient must receive a cover of antihelminthic drug like albendazole.[1,6] In general, the prognosis of primary subcutaneous hydatid cyst is quite good and relapse is very uncommon if the lesion is excised completely.[1]

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Conflicts of interest

There are no conflicts of interest.

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