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CASE REPORT

Subcutaneous hydatid cyst in the popliteal fossa at the site of a previous wasp sting

Istvan Battyany, Levai Andrea, Karoly Kalmar Nagy

ABSTRACT

We report an uncommon case of a primary Echinococcus cyst that developed in the subcutaneous tissue of the right popliteal fossa, at the spot of a previous wasp sting, suggesting the possibility of an unusual transmission of the eggs by insects. This unusual presentation was initially considered as a Baker cyst until parasytological results verified *Echinococcus hydatidosus*, the larval form of *Echinococcus granulosus*, as diagnosis. However, the most common path of *Echinococcus granulosus* infection is through contact with a definitive host or by ingestion of ova through contaminated water or food. Transmission by insects should also be reconsidered in endemic areas.

Key words: • *hydatid cyst* • *insect sting* • *ultrasonography*

Cystic hydatid disease is commonly caused by the larval stage of a parasite, *Echinococcus granulosus*. It still constitutes a serious public health problem in endemic regions. While liver and lung are the most commonly affected areas in adults, hydatid cysts may develop in almost any part of body (1). Cystic hydatid disease can occur in all viscera and soft tissues and has a high level of recurrence accounting for about 10% (2). Soft tissue hydatid cysts occur in 2.3% of cases reported from endemic areas; they are usually associated with involvement of other structures (3). Isolated involvement of soft tissues is relatively rare.

The development of primary Echinococcus cysts in humans is known to result from oral ingestion of *E. granulosus* eggs, released from the intestinal tract of carnivores. Humans are accidental intermediate hosts of this organism (3). Transmission by insects has not yet been published. We report an unusual case of a primary hydatid cyst in the subcutaneous tissue of the popliteal region which developed after a wasp sting.

Case report

A 63-year-old agriculture worker presented to our hospital with a mass located in the lower part of the popliteal region of the right lower limb. The patient reported that the swelling developed instantly at the spot where he was stung by a bee or wasp. At first it appeared as a small coffee bean shaped mass of 1–1.5 cm diameter and subsequently started to grow slowly, reaching 3–4 cm of size in a couple of days. He had no history of fever, weight loss, complaints of pain or claudication. On physical examination the non-tender, slightly mobile mass with a sharp contour was located in the lower and lateral aspect of the popliteal fossa. The skin above the swelling was somewhat hyperemic but not warm (Fig. 1). Posteroanterior chest radiograph and abdominal ultrasound revealed no abnormalities except for multiple small anechoic well-defined cysts in the bilateral renal sinuses. Laboratory investigation including liver function tests were unremarkable.

On ultrasonography of the mass located at the right popliteal region, a well-defined oval cystic lesion (40×17×18 mm) was visible in the subcutaneous tissue, with a double wall and multiple sized adjacent cyst-compartments with internal echoes of viscous content and solid particles (Fig. 2). Fine-needle aspiration cytology (FNAC) of the cysts was performed without complications, and 7 mL of fluid was aspirated and sent for cytological and parasitological examination. Cytological findings of the sample only revealed leukocytes among fragmented red blood cells, with activated and occasionally multinucleated macrophages and mast cells. The sonographic pattern of the cyst suggested the diagnosis of hydatid disease; according to the classification by Lewall and McCorkell, the cystic lesion represented Echinococcus cyst Type II (1). Since the cysts filled back up, surgical excision of the lesion was indicated, with

From the Departments of Radiology (I.B. ✉ *istvan.battyany@aok.pte.hu*, L.A.), and Surgery (K.K.N.), Medical University of Pécs, Pecs, Hungary.

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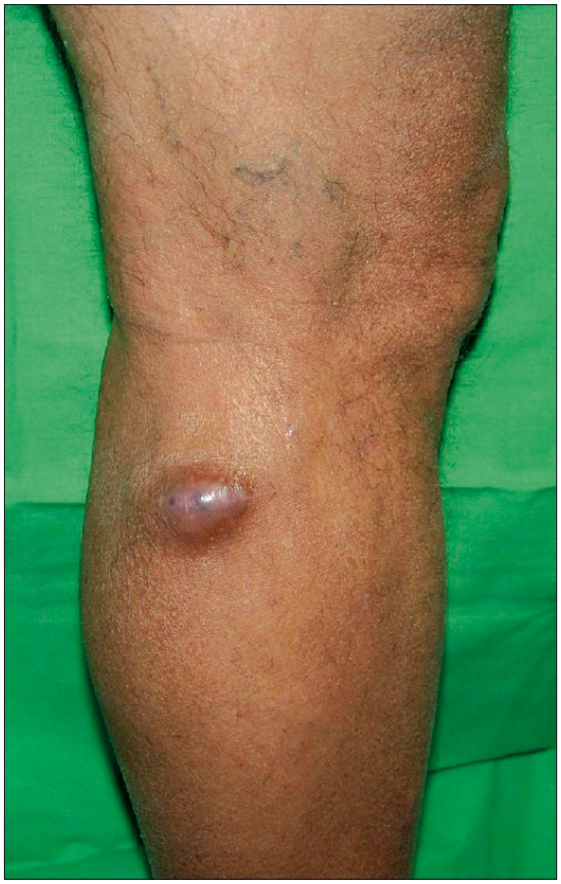

Figure 1. Well-shaped and well-demarcated mass located in the lower and lateral aspect of the popliteal fossa.

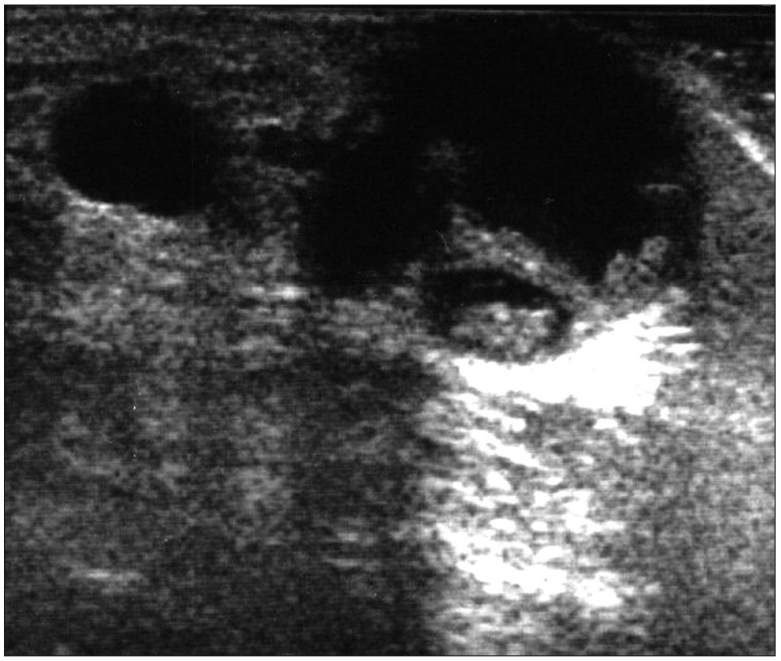

Figure 2. Ultrasonography revealed a multiloculated subcutaneous cystic lesion with internal echoes.

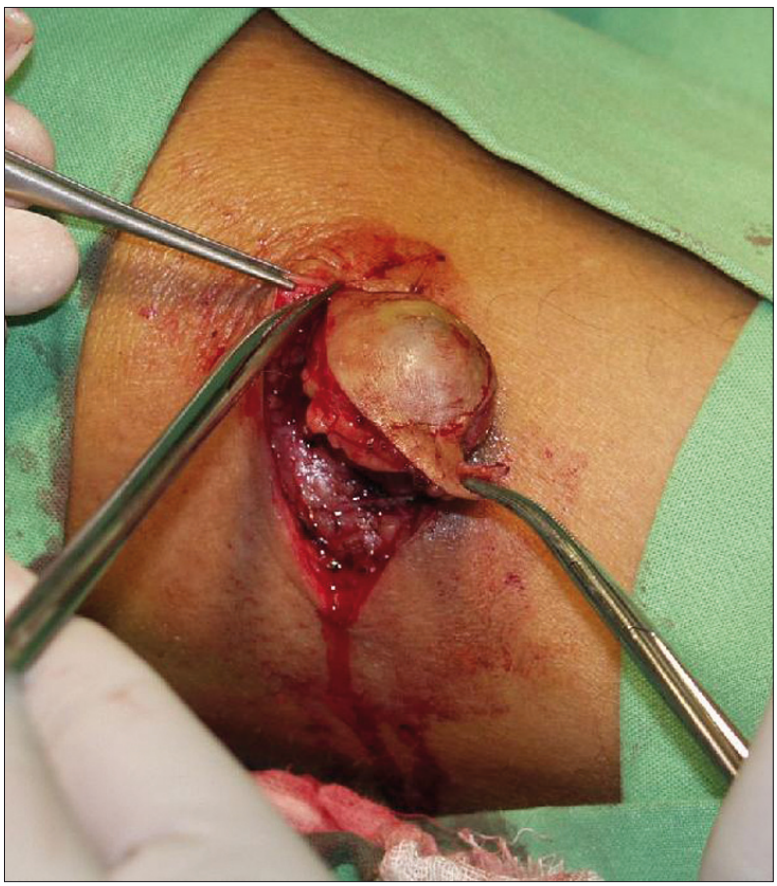

Figure 3. Surgical removal of the hydatid cyst.

preoperative and postoperative administration of mebendazol.

On control examination ten months after the excision, ultrasonography revealed two novel serous cysts (15 mm and 7 mm in diameter) in the subcutaneous tissue at the scar of previous surgery. Another FNAC was carried out followed by application of compression bandage. Despite multiple aspirations the cystic lesions gradually grew into the subcutaneous tissues and reached the original size in almost two years following the first excision. On ultrasonography the penetration into deep soft tissues of the hydatid cyst measuring 42×17×67 mm was observed. Definition of the exact extent of the cystic mass was made possible with magnetic resonance imaging (MRI). Curative cystectomy was subsequently indicated and performed for the second time (Fig. 3). Histological and immunohistochemical results showed cytokeratine positive and CEA negative polygonal epithelial cell proliferation with cystic compartments in the dermis. EMA reaction confirmed luminary membrane positivity. Malignant cells were not found in the specimen. Parasitological results verified *Echinococcus hydatidosus,* the larval form of *Echinococcus granulosus*. Parasitostatic medication was administered; a three-month course of mebendazole therapy was completed.

Nevertheless, after the second surgery further two consecutive cystectomies were needed within a three-year period due to recurring cysts. On the last operation the excision of an approximately 1-cm firm cyst was performed. By gross pathological examination the wall of the cyst was focally thickened, white and capsular, having inner trabeculae with fluid content. The macroscopic pattern of the excised mass suggested hydatid cyst as a diagnosis, the parasitological examination verified *Echinococcus hydatidosus* as well.

Discussion

Hydatid disease, caused by the larval form of *Echinococcus granulosus,* is endemic in cattle- and sheep-raising regions, such as Central Europe, the Mediterranean, the Middle East, South America, Australia, New Zealand, and South Africa (2). The adult worms live in the proximal small bowel of carnivores as the definitive host, attached by hooklets to the mucosa, and their eggs are excreted in the feces. Humans may become intermediate hosts through contact with a definitive host or by ingestion of ova through contaminated water or food. In our case, primary Echinococcus cyst developed in the soft tissue in the lower part of the popliteal region of the right lower limb, at the spot of a previous wasp sting, suggesting the possibility of an unusual transmission of the eggs by insects. Theoretically, wasps or other insects may inoculate Echinococcus eggs by getting in contact with contaminated feces, and spread them by stinging or biting the intermediate host. Lactic acid produced by the underlying muscles may assist in hatching of the ova, allowing the parasite embryos to form hydatid cysts.

Hydatid cystic disease of the soft tissues is uncommon, accounting for only 2.3%, according to the largest published series that consists of 24 cases in 1056 patients (4). Despite its rarity, soft tissue Echinococcus cyst might constitute an important differential diagnosis of any cystic mass in any anatomical location in endemic areas. Clinically, a hydatid cyst in the soft tissues might mimic teratomas, abscesses, or fibromatosis. Subcutaneous Echinococcus cysts are usually less than 5 cm (5); in the presented case, due to its penetration to deeper layers, the hydatid cyst reached 6.7 cm axial diameter.

As in the case reported here, imaging modalities and serologic investigations can reveal the correct diagnosis, especially in the presence of a competent pathologic examination. Ultrasound is an important imaging modality for hydatid disease and may clearly demonstrate the floating membranes and daughter cysts characteristically seen in purely cystic lesions (1). Preoperative diagnosis of subcutaneous hydatid disease is also possible by FNAC; the presence of diagnostic hooklets or laminated membrane ensures correct identification. No urticaria or anaphylactic reactions have been reported as a complication of this procedure (6), which encourages broader adoption of this method. In uncertain cases, MRI may be performed to confirm the hypothesis and visualize the lesion in various planes. The diagnosis is frequently delayed until the time of surgery, where observation of the laminated membrane possibly facilitates the identification. Postoperative positive indirect hemagglutination serology for *Echinococcus granulosus* can also confirm the missing diagnosis; however, sensitivity and specificity of serologic tests are unknown.

In a recent study by Akhan et al. percutaneous treatment was carried out by either a ''catheterization technique with hypertonic saline and alcohol'' or a''modified catheterization technique'', according to the type of the cyst, and an average of 96.1% volume reduction was obtained in six cysts of four patients (7). Cavity infection and cellulitis were observed as complications, which resolved after medical therapy (7). Percutaneous treatment is a safe and effective procedure in patients with soft tissue hydatid cysts and should be considered as a serious alternative to surgery (7).

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