

## Case Report

# Saphenous neuropathy due to large hydatid cyst within long adductor muscle: case report and literature review

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### Abstract

An unusual case of saphenous neuropathy secondary to compression by a large hydatid cyst within the adductor longus muscle is reported. Solitary hydatid cyst(s) localized in the skeletal muscles occur rarely and often mimic soft tissue tumours. Presentation with signs of peripheral nerve compression by a hydatid cyst in an extremity is exceedingly rare. Diagnosis can be established by ultrasound, computerized tomography or magnetic resonance if clinically suspected. Clinical suspicion of hydatid origin of a solitary muscle cyst should be high especially in patients hailing from areas endemic for echinococcosis. Laboratory tests are usually unhelpful in such cases and needle biopsy carries the risk of anaphylactic shock and should therefore be avoided. Surgical removal of the unruptured cyst is the treatment of choice in cases of intramuscular hydatid cyst. In the present case, excision of the hydatid cyst was followed by complete clinical recovery. In the absence of systemic involvement, treatment with albendazole may be avoided.

**Key words:** Hydatid; cyst; muscle; peripheral; nerve compression

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### Introduction

Echinococcosis is an endemic parasitic disease in Albania as well as other countries in the Mediterranean. It is also common in other sheep rearing countries such as China, Central Asia, the Middle East, Eastern Africa, and parts of South America. It is caused by the larval form of *Echinococcus granulosus* [1]. The liver and lungs are the two most frequently affected organs in humans as the swallowed eggs hatch in the intestines and are transported by the portal system to the liver [1,2]. The embryos that escape the hepatic filter are transported by venous circulation to the right heart and subsequently to the lungs. If embryos leave the pulmonary circulation to reach the left heart chamber, the arterial circulation can diffuse them to any part of the body. Echinococcosis can also affect the brain, heart, kidney, ureter, spleen, uterus, fallopian tube, mesentery, pancreas, diaphragm and muscles [1,3,4]. Spinal echinococcosis has been documented in the literature with consequent neurological deficit from compression of the spinal cord and/or rootlets [5-7]. There are few reported cases of hydatid cyst in skeletal muscles [8-21]. Association of peripheral

neuropathy with adjacent hydatid cyst(s) is rare [4,9,22-25], and is mainly caused by compression of the nerve by the hydatid cyst.

### Case report

An 18-year-old boy sought medical attention in June 2008, for right thigh swelling, paresthesia and numbness along the medial part of the leg. He complained of progressive increase in size of the lump over the last three months. Six weeks prior to presentation, he developed numbness along the inner part of the right leg, radiating from the knee to the anterior aspect of the medial malleolus. He grew up in a town and during childhood had contact with small domestic animals such as dogs, cats, and sheep or cows in summer holidays in his grandparents' village. He was not a drug user. His parents reported that except for the common infectious diseases of childhood, he had led an otherwise healthy life. Their family used to regularly consume milk products, such as cheese and butter from unlicensed villagers. The physical examination revealed a large swelling in the medial aspect of the right thigh in the triangle of Scarpa. There was hypoesthesia along the medial

aspect of the knee up to the medial malleolus along the distribution of the saphenous nerve. No motor deficit was detected on formal motor examination of the quadriceps femoris. Adduction against resistance of the stretched right leg caused slight pain and discomfort over the swelling. Ultrasound documented a hypo-echogenic solitary cyst of 10 x 5.4cm, with clear-cut borders, without vascularisation within. Magnetic resonance of the thigh revealed a solitary cyst 5 x 6 x10 cm in the right adductor longus muscle with the characteristic features of a hydatid cyst with spiral scolex within the cyst. The cyst along its anterolateral aspect was seen compressing the femoral neuro-vascular bundle (Figures 1 and 2). Computerized tomography (CT) of the chest and ultrasound of the abdomen did not reveal any cysts. Routine hemogram (including eosinophil counts) was within normal limits and specific serodiagnostic assays (indirect hemagglutination test and the enzyme-linked immunosorbent assay (ELISA) for echinococcal antibodies were negative.

The patient underwent surgery under general anesthesia in a recumbent position. A longitudinal incision over the swelling revealed a large mass under the fibers of the femoral adductor femoris muscle. The adductor muscle fascia was incised along its longest diameter. The cyst was carefully dissected and freed from the covering muscle fibers. The femoral vasculo-nervous bundle was compressed by the cyst and displaced laterally. The saphenous nerve was compressed, resulting in paresthesia and hypoesthesia along the medial aspect of the leg. To avoid inadvertent rupture of the cyst, the dissection of the cyst from the adjacent muscle was done with thick cotton gauze. Complete removal of the unruptured cyst was achieved with pericystectomy. Histological examination of the cyst wall with hematoxylin eosin dye demonstrated the characteristic ectocyst of the hydatid with homogenous eosinophilic chitinous membrane.

The patient was discharged the following day after surgery and able to walk independently. Paresthesia disappeared within one week following surgery. Sensation in the right leg returned one month later. In view of normal serological tests, complete removal of unruptured solitary cyst, and no evidence of systemic involvement, albendazole was not given postoperatively [13,21]. At two and a half years' follow-up, blood tests, repeat MRI of the thigh, ultrasound of abdomen, and CT of the chest were performed, and the patient remained disease-free.

## Discussion

Echinococcosis most frequently affects the liver (65% of cases), followed by the lungs (15–25%) and kidneys (3%) [1]. Other less common locations include the spleen (0.8–8%), bone (0.5–4%), and brain (1%) [1,9]. Liver and lungs are mostly affected because they are two primary stations for the larvae following systemic spread due to their dense capillary network. After exiting these filters, larvae may reach any organ in the body.

Isolated involvement of skeletal muscle is rare and represents 0.2% - 2.2% of all cases of echinococcosis [2,9-14,16,17,22]. The relative rarity of muscular involvement may be explained by lactic acid toxicity and muscular contractions, which may prevent fixation of the larva [26]. The lower extremities are affected more often than the upper extremities [27] and the proximal muscles of the limbs are involved predominantly [13].

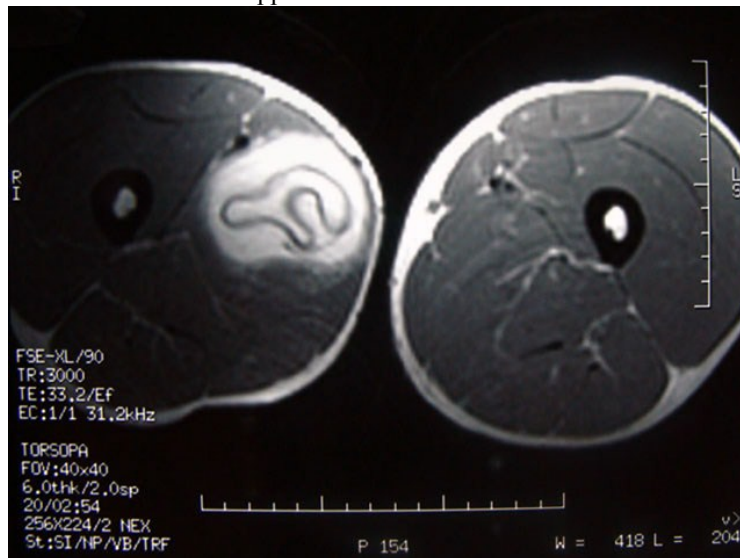
Localization of the hydatid cyst in the thigh is rare and constitutes 0.35% to 15% of in large reported series of skeletal muscle echinococcosis [28,29]. Rada reported a case of hydatid cyst in the wall of the femoral artery and another in the adductor muscles that resulted in an ischemic syndrome in the affected limb [25]. Neurological deficit associated with spinal and intracranial localization of echinococcosis is well documented [2,5-7]. Peripheral nerve involvement is very rare. We could find only isolated cases reported in the literature of localization of the hydatid cyst(s) in the thigh [23] or gluteal muscles [22] involving the sciatic nerve, or in the pelvis [4,24] presenting with lumbar plexopathy. Postoperative temporary deficit of crural nerve following removal of hydatid cyst in the muscle was reported by Daali [17], but there were no deficit in preoperative evaluation.

To date, only a few authors have reported the occurrence of pelvic hydatid cysts causing sciatica associated with foot-drop [4,22-24]. Four of the reported cases presented with sciatic nerve deficit secondary to mass effect of the cyst located in proximity to the lumbar plexus or sciatic nerve. In one case [23] the sciatic pain that was associated with neurological sensory motor deficit along the sciatic nerve was caused by the hydatid cysts intertwined within the nerve. The present case may be the first case of isolated sensory neuropathy secondary to compression of the saphenous nerve by the hydatid cyst. Magnetic resonance imaging documented compression of the femoral neuro-vascular bundle

**Figure 1.** Coronal T1 weighted showing detached germinal layer inside of the large cyst deviating femoral neurovascular bundle.



**Figure 2.** Axial T2 weighted showing the cyst with femoral neurovascular bundle in its upper lateral wall.



by the cyst. The sensory deficit improved following removal of the cyst and disappeared one month after surgery.

The most important factor in the diagnosis of hydatid disease is an awareness of its possibility [7], especially when treating patients who lived in or have emigrated from areas where echinococcosis is prevalent [23]. Clinically, hydatidosis can mimic a soft-tissue tumour [12]. Diagnosis may be supported by laboratory tests. However, eosinophilia is seen in only one fourth of the cases with musculoskeletal echinococcosis [27]. Skin tests (Casoni's test) are nonspecific, and serological tests are often negative in the absence of hepatic and lung disease [27]. A soft-tissue hydatid cyst can be accurately diagnosed on ultrasound and computerized tomography scans, but magnetic resonance imaging studies is the gold standard [26,27,31].

Needle biopsy should be avoided as it carries a high risk of dissemination of infection and may be responsible for anaphylactic reactions [27,32]. Skeletal muscle hydatid cyst, though rare, should be considered in the differential diagnosis of soft tissue tumours, particularly in patients in endemic areas. Histological evaluation is essential for confirmation of diagnosis by demonstrating the chitinous membrane and protoscoleces.

Today, treatment options for cystic echinococcosis include surgery, PAIR (puncture, aspiration, injection, reaspiration), and chemotherapy [2,3]. In the case of isolated muscular echinococcosis, the complete removal of the unruptured hydatid cyst(s) is the treatment of choice [2,8-14]; however, perioperative dissemination of the infection and anaphylactic shock are possible complications [32]. When intraoperative opening of the cyst occurs hypertonic saline solution is used for rinsing. In such cases albendazole should be used postoperatively. If preoperative prediction of complete removal of unruptured cyst is not possible, albendazole therapy can be initiated prior to surgery.

Percutaneous aspiration of hydatid cysts has been reported in selected cases [14], but it carries a high risk of life-threatening anaphylactic shock. PAIR is indicated for inoperable patients with cystic echinococcosis and those who refuse surgery [2,3]. It has been used for the treatment of cysts in the liver, the abdominal cavity, spleen, kidney and bones, but it should not be used for lung cysts [2,3]. Medical treatment with imidazoles has little efficacy for the treatment of muscular hydatid disease and has not been routinely used in reported cases [13,21]. It is

preferred in complicated cases of large or multiple cysts for which it may be surgically difficult to achieve complete extirpation due to involvement of neurovascular structures within the cyst(s) [23].

The reported case shows that complete removal of an unruptured solitary cyst in skeletal muscle is sufficient to achieve a cure. Surgical decompression restored full sensory function in this patient. When blood investigations are normal and systemic involvement is not noted, albendazole may be avoided.

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