Primary hydatid cyst of the thigh: a case report of an unusual localization

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The sartorius muscle is a rare location of primary hydatid cyst. In this report, the clinical, pathologic, and radiologic features are discussed, with special emphasis on the pathologic and radiographic features.

We report a case of hydatid cyst of the proximal thigh in an eight-year-old girl. Magnetic resonance (MR) images revealed muscular hydatid cyst in the sartorius muscle, with a striking appearance of daughter cysts. We did not find any visceral organ involvement. Wide excision was performed without destroying the cyst wall.

Primary muscular hydatidosis should be kept in mind in the differential diagnosis of a cystic mass of a skeletal muscle.

Key words: echinococcosis, muscle hydatidosis, primary, magnetic resonance imaging.

Human hydatid disease caused by Echinococcus granulosus is of worldwide importance and presents medical, veterinary and economic problems in developing countries. In humans, the infestation is usually located in the liver (65%) or lungs (25%), and rarely involves the brain, heart, bone, or other organs[1]. Primary muscular hydatid cysts comprise less than 0.5% of the cases in endemic populations[2]. These cysts appear as slow-growing masses of soft tissue, sometimes with inflammatory signs and fistulization.

In this report, we present a rare case of muscular hydatid disease in which preoperative diagnosis was achieved by characteristic appearance of the cyst in magnetic resonance (MR) images. Preoperative diagnosis of hydatid disease is essential because rupture and dissemination of the cyst may result in recurrence, and intraoperative spillage of the antigenic cyst fluid may lead to severe anaphylactic response.

Case Report

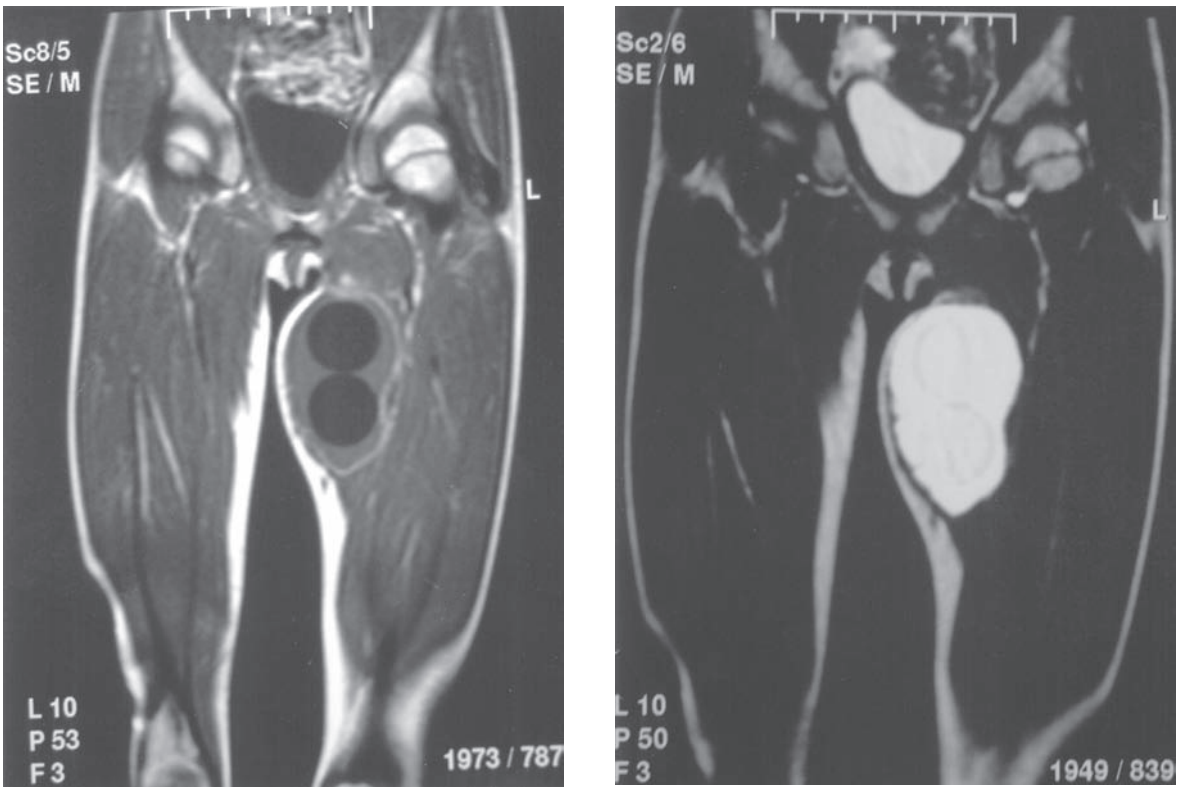
An eight-year-old girl living in a rural area admitted to hospital with the complaint of a slow-growing, painless mass in her left thigh. There was no history of abdominal pain, chest pain, hemoptysis, cough, fever, chills, weight loss, or urticaria.

Physical examination revealed a firm, nontender, fixed mass of 8x5x5 cm in the anteromedial proximal left thigh. There was no fluctuation, erythema, ecchymosis, increased warmth or lymphadenopathy.

Laboratory results showed an erythrocyte sedimentation rate of 12 mm/h (Westergren) and a total leukocyte count of 7,000/mm^3.

Magnetic resonance (MR) imaging was conducted using Philips Gyroscan NT 1.5 T instrument (Philips Medical Systems, The Best, Netherlands). T1- and T2-weighted images demonstrated a well-defined oval cystic mass in the sartorius muscle, containing round-shaped daughter cysts (Fig. 1). A continuous low-intensity rim could be easily seen in T2-weighted images. The fluid in the daughter cysts showed slight hypointensity compared to the main cyst in both sequences. There was no sign of wall thickening or detached germinative membranes.

Since the MR images were suggestive of hydatid cyst, further laboratory and imaging studies were employed to support the diagnosis and to



(a) (b)

Fig. 1. T1-weighted (a) and T2-weighted (b) coronal magnetic resonance images show a well-defined cyst in the medial thigh containing daughter cysts.

detect the other sites of possible involvement. Skeletal survey radiographs, plain films of the chest, and abdominal ultrasound examination revealed no other lesions. There was a positive response to indirect hemagglutination test for hydatid disease.

Surgically, the mass was excised under general anesthesia with wide surgical margins without destroying the cyst wall, followed by irrigation with 3% hypertonic saline. Histological examination of the specimen revealed daughter cysts and fragments of the lamellar membrane of the hydatid cyst. Scoleces were also detected within the surgical specimen. No bacterial pathogen was cultivated in the cyst fluid.

Postoperative recovery was uneventful. An albendazole therapy, 200 mg twice daily, was given for six weeks. Thorough clinical, serological, radiological, and MR examinations did not reveal any recurrence or any other site of hydatid disease in the follow-up span of two years.

Discussion

The majority of hydatid cysts cause no symptoms. The manifestation of symptoms depends on the location, size, and the pressure caused by the enlarging cyst. Hydatid cysts grow slowly, like benign tumors[3,4].

In muscular hydatidosis, the primary foci are generally thoracic or abdominal organs from which the dissemination occurs during the primary surgery. Primary skeletal muscle hydatidosis without involvement of the thoracic and abdominal organs is extremely rare. However, cases involving the muscles of the vastus lateralis[5], supraspinatus[6], biceps brachii[7], pectoralis major[8], gracilis[9] and quadriceps[10] have been reported in the literature. The muscle is considered an unfavorable site for hydatidosis because of its high lactic acid level[7].

This report presents a very unusual case of primary intramuscular infestation of Echinococcus granulosus. There have been only

two cases reported as primary intramuscular hydatidosis of the sartorius muscle in the literature[11,12].

Given its relative rarity, the diagnosis of soft tissue hydatid cysts requires a high index of suspicion. A preoperative radiological work-up may reveal the characteristic features of a hydatid cyst; radiological findings range from purely cystic lesion to a completely solid appearance[13]. A continuous low-intensity rim with a maximum thickness of 4-5 mm, multicystic appearance, a homogeneous signal intensity of the cystic fluid and presence of daughter cysts are the most characteristic MR imaging findings[14]. The detachment of the germinative membrane from pericyst (water-lily sign) is considered to be pathognomonic[11]. MR imaging is capable of adequately demonstrating these features and is useful in monitoring the response to treatment. MR imaging can also assist in defining treatment strategy, by showing extent and location of the lesion. Computerized tomography (CT) has been used in the diagnosis of subcutaneous hydatidosis[15], but MR imaging is superior to CT in the definitive radiological diagnosis of subcutaneous and muscular hydatidosis[11,14].

Since the disease is generally located in other parts of the body, primarily in hepatic or pulmonary regions, the patients with echinococcosis must undergo a thorough systemic examination[16-18]. A chest radiograph, ultrasonography of the abdomen, and bone X-rays did not reveal any additional lesions in our patient. Indirect hemagglutination test result was positive for hydatid cyst; however, due to the high rate of false-positive and negative results, serological tests including indirect hemagglutination, latex agglutination, enzyme-linked immunosorbent assay and immunoelectrophoresis are controversial in the initial diagnosis of hydatid disease.

Surgery is the most effective way to treat hydatid cysts. Typically, medical treatment alone is not effective in hydatid disease[19,20]. Complete surgical resection plus medical therapy is the preferred treatment for isolated echinococcosis. Rupture or spillage of cysts should be avoided to prevent local or distant dissemination and immediate anaphylaxis[9,18,21].

In conclusion, hydatid cyst with origin in the sartorius muscle is a rare entity with important surgical implications. It must be kept in mind in the differential diagnosis of a slow-growing soft tissue mass of the extremities, especially in regions where this zoonosis is endemic. The characteristic radiological findings may help in preoperative diagnosis of the disease.

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