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Case report

Primary muscular hydatidosis in an endemic area of Peru: report of two cases

Plinio CONDOR[1], Jose ARGANDOÑA[2], Kevin FLORES-LOVON[3], Dayana TICONA[3], Nicolás CUBA-CÁCERES4, Ericson L. GUTIERREZ[5,6]

[1]Hospital "Ernesto German Guzman Gonzales" – MINSA, Oxapampa, Pasco, Perú
[2]Hospital II EsSalud - Huánuco. Huánuco, Perú
[3]Universidad Nacional de San Agustín de Arequipa, Perú
[4]Universidad de San Martin de Porres, Faculty of Human Medicine, Lima, Perú
[5]Insituto Nacional de Salud, Lima, Perú
[6]Universidad San Ignacio de Loyola, Unidad de Investigación para la Generación y Síntesis de Evidencias en Salud, Lima, Perú

Corresponding Author: Ericson Gutierrez Ingunza; e-mail: eringunza@yahoo.es

ABSTRACT. Primary muscular hydatidosis is an infrequent parasitic infection, caused by *Echinococcus granulosus*. We report 2 cases of female patients with diagnosis of primary muscular hydatidosis of the thigh. Both hail from an endemic area for hydatidosis, and had no pulmonary, hepatic, or systemic involvement. Cyst extraction was performed after the patients were given hypertonic solution, and antiparasitic treatment.

Keywords: cystic echinococcosis, hydatidosis, Peru

Introduction

Cystic echinococcosis is a parasitic zoonosis caused by the larval stage of tapeworm *Echinococcus granulosus* [1]. It is endemic in cattle raising countries such as Peru, Chile, Argentina, Uruguay, Brazil, some countries in Africa and Western China [2]. In Peru, studies show an incidence rate between 14 and 34 cases per 100 000 habitants, in the Junín, Pasco and Huancavelica Regions [3].

The most frequent definitive host of adult tapeworms of *E. granulosus* are dogs. Humans are accidental hosts. Infections in humans is caused by the ingestion of food contaminated with eggs of *E. granulosus*. When these eggs reach the intestine, they hatch and release oncospheres. The oncospheres cross the intestine and pass into the bloodstream and reach different organs, mainly the liver and lungs, where they become larvae [4]. Later, a cyst develops in the affected organ [5].

Intramuscular hydatid cyst presentation is uncommon, especially the primary form [5]. This condition is usually asymptomatic and is reported in 1–4% of cases of echinococcal infection [6].

Due to the occasional appearance of intramuscular hydatid cyst, early diagnosis and subsequent treatment can be difficult in patients. Interdisciplinary management is often necessary [7]. We present two cases of patients with primary muscular hydatidosis who were not diagnosed early due to their chronic evolution and having few symptoms. Both cases are described below.

Case report 1

A 33-year-old female patient goes to an outpatient surgery clinic with a disease time of 2 years. Refers tumor in anterior muscular region of right thigh, with progressive growth that causes pain and functional limitation when walking. The patient is originally from Santa Ana de Tusi, Pasco Región; a livestock area. The physical examination shows deformity and increased volume in the proximal third of the anterior right thigh, where a partially mobile tumor of approximately 10×6 cm in

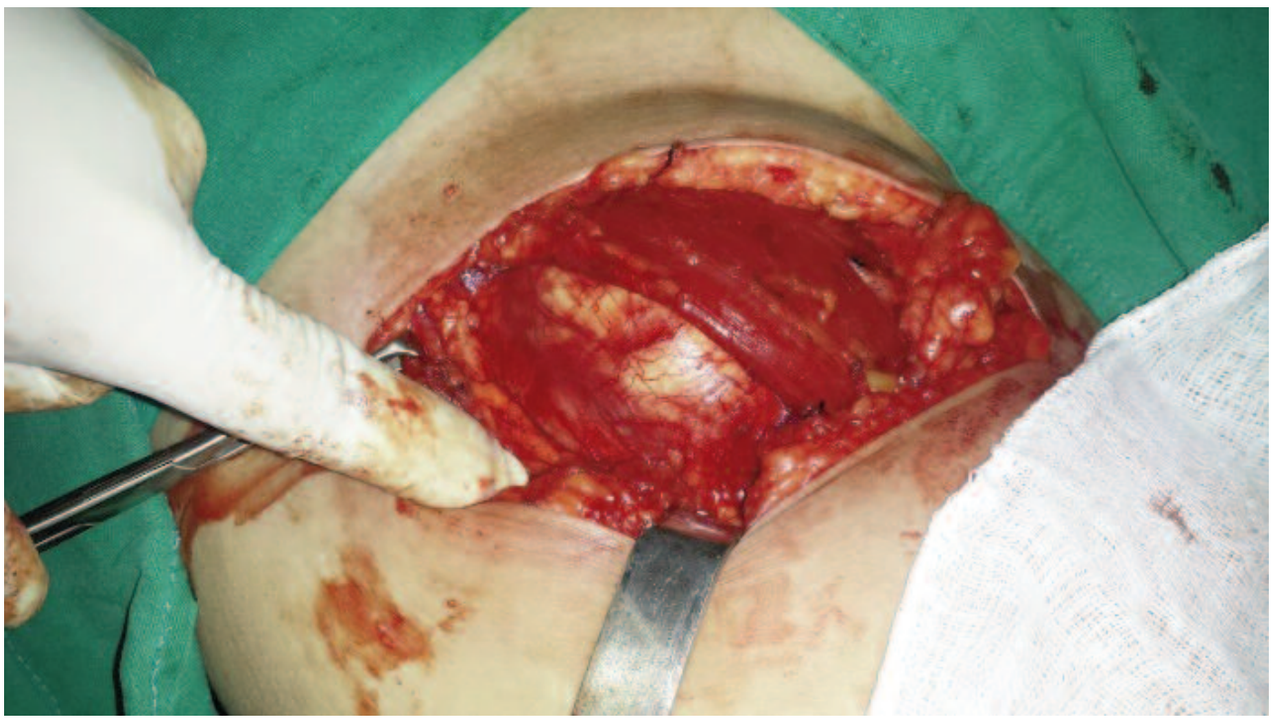

Figure 1a. Cyst is visualized below the sartorius and vast medial muscles

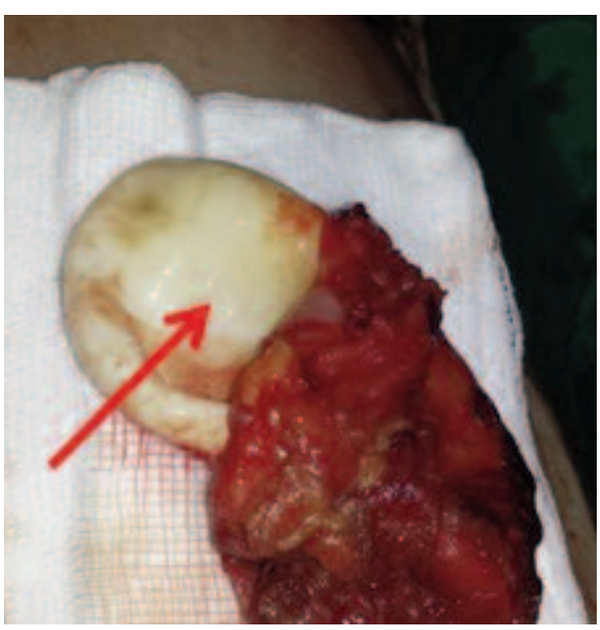

Figure 1b. Fibrous tissue cyst wall called: adventitia, periquistic or ectocyst

diameter is palpated. Soft tissue ultrasound showed a cystic lesion suggestive of muscular hydatidosis. Abdominal ultrasound, chest radiography and other preoperative exams showed no alteration (Tab. 1). Subsequently, hypertonic solution is given. The surgeon used an oblique incision parallel to inguinal line (Figs 1a and 1b).

Case report 2

A 30-year-old female patient goes to an outpatient surgery clinic with a disease time of 18 months. Refers tumor in posterior region of left thigh of slow and progressive growth associated with intermittent pain. She comes from the district of Ninacaca, a livestock area of the Pasco region. She refers to raising sheep and dogs. She lived in the countryside until she was 7 years old, and then moved to the city of Cerro de Pasco. The physical examination shows a partially mobile tumor of approximately 12×8 cm. The tumor is located in the distal third of the posterolateral left thigh. Soft tissue ultrasound shows inflammation of the semitendinous muscle. Chest radiography and abdominal ultrasound showed no alteration. The CBC showed a left shift due to leukocytosis (Tab. 1). Subsequently, an excision was performed with an oblique incision posterior area left thigh (Figs 2a and 2b).

Both patients evolved favorably and were discharged. In both cases, antiparasitic treatment consisted of albendazole 400mg orally every 12 hours for 90 days.

Discussion

The cases of two women who presented primary

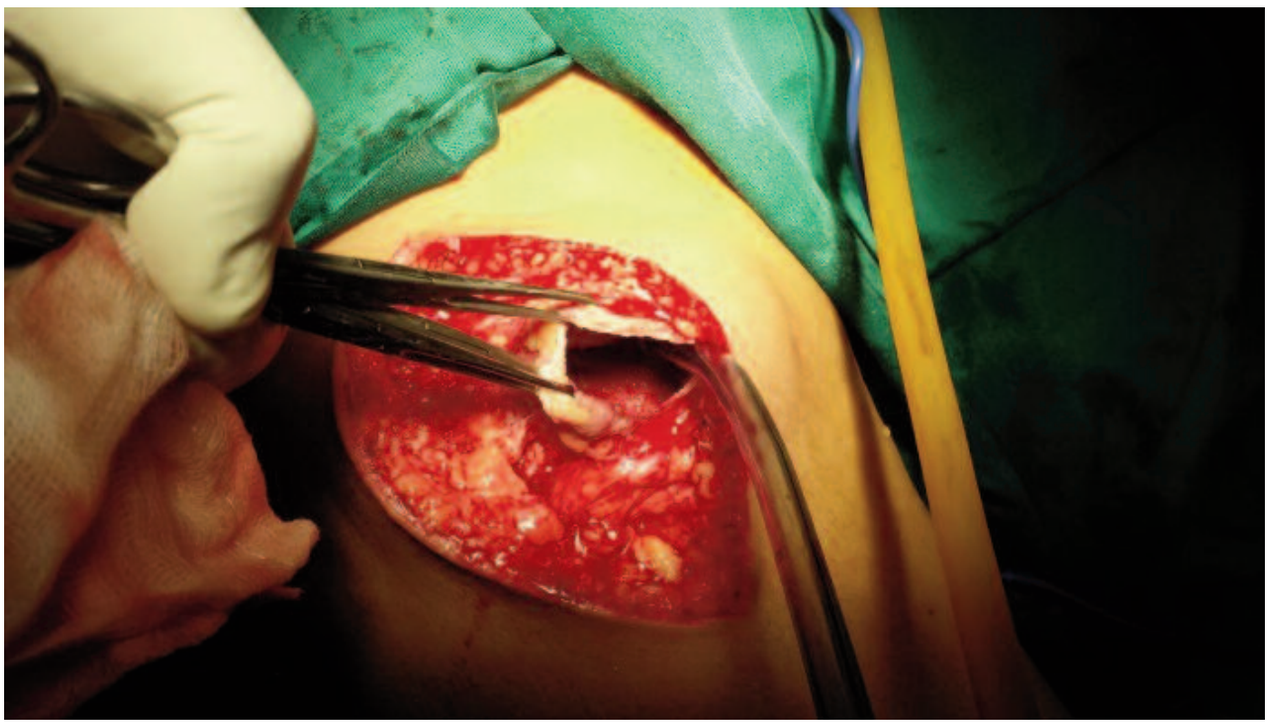

Figure 2a. Cystic tumor approximately 12×8 cm in diameter, thick hydatid membrane

Figure 2b. Cystic content: abundant daughter vesicles plus liquid „rock crystal”

muscular hydatidosis in lower limbs. A patient reported by Botello [8] with a history of 4 years, also has symptoms of pain, functional limitation when walking and volume increase in lateral region of left thigh that ended in the cutaneous fistula. Another patient reported by Petit [9] with a one-year history, presented a mass in the lateral region of the right thigh and progressive pain that worsened during exercise. Likewise, Roldan-Aviña [10] presented the case of a patient with a history of 5 months with intermittent claudication and a mass in the anterior of the right thigh. In these cases, as in this case report, no history of previous hydatidosis was found.

Table 1. Summary chart of general features of the patients

	Age(y)/Sex	Disease time (years)	Sites/Number of lesion	Cyst dimensions (cm)	Clinical manifestations	Complete (CBC) Parameter	Blood count	Soft tissue ultrasound findings on the thigh
1	33/F	2	Right front thigh/single	10×6	Pain and functional limitation when walking	WBC	6,560 cells/mcl	Cystic lesion suggestive of muscular hydatidosis
						Lymphocytes	43 (pct)	
						Monocytes	2 (pct)	
						Eosinophils	1(pct)	
						Segmented neutrophils	48 (pct)	
						Band neutrophils	6 (pct)	
						Hb	17,4 g/dl	
2	30/F	1½	Left back thigh/single	12×8	Pain	WBC	12,180 ells/mcl	Semitendinosus muscle inflammation
						Lymphocytes	21 (pct)	
						Monocytes	3 (pct)	
						Eosinophils	1 (pct)	
						Segmented neutrophils	73 (pct)	
						Band neutrophils	3 (pct)	
						Hb	16.5 g/dl	

The primary muscular location is very rare; presenting between 1–4% of total skeleto-muscular hydatidosis tumors [6]. 45% of the primary muscular tumors are located in lower limbs and 77% of the lower limb tumors are in the thigh [6]. The symptoms of this form of hydatidosis are nonspecific, such as pain, fever and tumor [11].

The most sensitive serological tests used are enzyme immunoassay (ELISA), indirect hemaglutination and latex agglutination, with western blot and immunoelectrophoresis tests being more specific [12]. These tests have high sensitivity (80–100%) and specificity (88–96%) in cases of liver disease; being less sensitive (25–56%) when the condition is in other tissues, including muscles [12].

Imaging tests are important to establish a differential diagnosis with soft tissue tumors and abscesses [13]. Ultrasound is able to detect most of the characteristics of cyst. However, magnetic resonance imaging (MRI) is the preferred diagnostic imaging method since it provides a better evaluation of the locoregional extent of the lesion and the relationships with the nerve and vascular pedicles [5].

Surgery is the most effective way to treat hydatid cysts. Hypertonic saline should be used before the surgery to eliminate scolices and prevent the rupture of cysts to prevent local or distant dissemination. Medical antiparasitic therapy lowers the recurrence rate, being albendazole and mebendazole the drugs of choice [14].

As limitations of the case report, the serotypes of *Echinococcus granulosus* were not identified to demonstrate its relationship with muscular location and its probable resistance to antiparasitic treatment. We highlight the infrequent location of the cyst and that presurgical imaging allows the correct therapeutic approach.

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