Case Report

Lower Extremity Sparganosis in a Bursa

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Received: 10 November 2010 / Accepted: 12 April 2011 / Published online: 26 April 2011
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Abstract

Background Sparganosis is a rare parasitic infection caused by the plerocercoid tapeworm larva of the genus *Spirometra*.

Case Description We report the case of a 67-year-old man with a mass over the anteromedial surface of the proximal extremity of the tibia. We surgically excised a bursa containing *Spirometra* larvae.

Literature Review Sparganosis is a rare parasitic infection. We found no cases of lower extremity sparganosis combined with bursitis reported in the literature.

Purposes and Clinical Relevance Sparganosis should be considered in the differential diagnosis of soft tissue tumors, especially among patients who frequently have consumed mountain water or raw snakes or frogs.

Introduction

Sparganosis is a rare parasitic infection caused by the plerocercoid tapeworm larva of the genus *Spirometra* [3]. The *Spirometra* larvae are white, wrinkled, ribbon-shaped, and a few millimeters to several centimeters in length. Sparganosis is transmitted to humans by drinking water contaminated with copepods containing *Spirometra* larvae, by consuming the raw flesh of a second intermediate host, such as frogs or snakes, or by placing a poultice infected with plerocercoid larvae on an open wound [2].

The clinical presentation of sparganosis occurs after the larvae migrate to a subcutaneous location. The destination of the larvae is often a tissue or muscle in the chest, abdominal wall, extremities, eyes, brain, urinary tract, spinal canal, and scrotum. The parasite triggers a local lymphocytic and eosinophilic inflammation.

We report an unusual case of sparganosis combined with bursitis involving the proximal lower extremity.

Each author certifies that he has no commercial associations that might pose a conflict of interest in connection with the submitted article.

Each author certifies that his institution approved the reporting of this case report, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

This work was performed at Incheon St Mary's Hospital, Incheon, Korea.

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Case Report

A 67-year-old man initially was evaluated for a mass of 3 months' duration over the right anteromedial surface of the proximal tibia. The mass was approximately 3 cm by 3 cm, soft, nontender, and mobile. He was treated with antiinflammatory medications for the following 3 months. The symptoms were alleviated by the treatment and the mass resolved. However, after cessation of the oral agents, the mass recurred with overlying erythema. He was referred from a private clinic for treatment of a spontaneous rupture of the gastrocnemius. He had no history of leg trauma and overseas travel and denied ingesting raw snakes or frogs. He reported a history of pulmonary tuberculosis

when he was 25 years old. He was treated for the tuberculosis and believed to have been cured. His medical history also was significant for diabetes mellitus, which developed in his sixth decade of life. The physical examination again revealed a 3 cm × 3 cm, soft, nontender mass. The mass was freely mobile and easily compressed. Laboratory studies revealed a normal leukocyte count ($5.6 \times 10^9/L$) with 66.0% neutrophils, 22.3% lymphocytes, 7.1% monocytes, and 4.3% eosinophils. The erythrocyte sedimentation rate was 81 mm/hour.

Plain radiographs of the knee showed numerous small, nodular, calcific densities in the right posterior thigh and leg, especially proximal to the popliteal fossa, suggesting sequelae of a previous inflammatory response, such as a parasite infestation or tuberculosis (Fig. 1A, B). MRI revealed multilobulated, peripheral, enhancing lesions with an adjacent soft tissue signal abnormality from the subcutaneous layer of the right proximal, anteromedial to proximal-to-mid-lower extremity involving the medial gastrocnemius muscle ($5 \times 1.5 \times 10$ cm) (Fig. 1C). The preoperative imaging diagnosis was an abscess with infectious myocellulitis.

Three months after the initial appearance, we excised the lesion, which appeared to be an enlarged bursa with a thick fibrous wall containing a living sparganum (Fig. 2) and a large amount of necrotic tissue. There was little fluid in the bursa although the pes anserinus conjoined tendon had a dark brown, puslike discharge. The histologic examination revealed multinucleated giant cells (Fig. 3). Although culture of the surgical specimen and real-time polymerase chain reaction (PCR) did not show *Mycobacterium tuberculosis* and the histology was nonspecific, we considered clinical manifestations consistent with tuberculous bursitis, especially in this region where tuberculosis is prevalent.

Antituberculous therapy consisted of isoniazid (300 mg/day), ethambutol (750 mg/day), and rifampicin

(450 mg/day). The oral antituberculous therapy continued for 9 months postoperatively. Serial ESR at 3, 6, and 10 months declined to values within the normal reference range. At the 10-month followup, the patient had no symptoms and there was no recurrence of the lesion.

Discussion

Human sparganosis is an extremely rare disease, even in endemic countries, the manifestations of which are diverse, such as nonspecific discomfort, vague pain, palpable masses, headaches, or even no symptoms, depending on the organs involved [6]. Humans can be infected by ingesting water or the raw flesh of frogs or snakes and by contact with an infected poultice. Currently, the diagnosis of sparganosis can be made based on serology. Peripheral eosinophilia is common in human sparganosis. Although the activity of eosinophils is not entirely clear, they are known to destroy parasitic organisms [1]. A competitive ELISA test using sparganum-specific monoclonal antibodies has been developed to improve the diagnostic specificity of sparganosis [5]. Human sparganosis is a surgical disease because the diagnosis depends almost entirely on observation of larvae from lesions; however, immunodiagnosis for sparganosis is recommended to establish a preoperative diagnosis when soft tissue tumors are detected in patients living in endemic areas [4]. Our patient denied ingesting raw snakes or frogs, and there was no evidence of eosinophilia. Thus, an infection by sparganum was not suspected.

Our case is unusual because our patient had pulmonary tuberculosis more than 40 years before presenting with the mass and parasitic infestation. Consequently, although the disease presumably had been cured and was inactive, we treated the patient with antituberculous chemotherapy and

Fig. 1A–C (A) Plain AP and (B) lateral radiographs of the knee show numerous, small, nodular, calcific densities in the right distal posterior thigh. (C) An axial T2-weighted MR image reveals multilobulated peripheral enhancing lesions (thin arrow) with an adjacent soft tissue signal abnormality from the subcutaneous layer of the right anteromedial proximal to the proximal-to-mid-lower extremity involving the medial gastrocnemius muscle (thick arrow).

Fig. 2A–B The photographs show (A) a sparganum in the right proximal medial lower extremity, and (B) the removed sparganum worm.

Fig. 3A–B (A) Histologic examination showed necrosis (arrow), granulomatous inflammation (Stain, hematoxylin & eosin; original magnification, $\times 100$), and (B) multinucleated giant cells (Stain, hematoxylin & eosin; original magnification, $\times 400$) in a 67-year-old patient with lower extremity Sparganosis.

surgery, presuming there had been reactivation of the tuberculosis. Although laboratory culture of the surgical specimen and real-time PCR did not detect *Mycobacterium tuberculosis*, we initiated antituberculous chemotherapy. Long-term followup should be continued because recurrences have been reported after long intervals of regression of the disease.

Sparganosis infestations should be considered in the differential diagnosis of soft tissue tumors, especially among patients who frequently consume mountain water or raw snakes or frogs.

Acknowledgment We thank SH Park MD for assistance in preparing this manuscript.

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