

Quiz

CORRECT ANSWER TO THE QUIZ. CHECK YOUR DIAGNOSIS

CASE REPORT

SPARGANOSIS PRESENTING AS A SUBCUTANEOUS MASS OF THE THIGH

CHIA-CHIN TSAI¹, KEVA JOSEPH², I-WEI CHANG^{1,3,4,5}

¹Department of Pathology, Taipei Medical University Hospital, Taipei, Taiwan

²St. Jude Hospital, Vieux Fort, Saint Lucia, West Indies

³Department of Pathology, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

⁴Department of Clinical Pathology, Wan Fang Hospital, Taipei Medical University, Taipei, Taiwan

⁵Department of Pathology, Shuang Ho Hospital, Taipei Medical University, Taipei, Taiwan

We described a case of subcutaneous sparganosis of a 61-year-old woman, presenting with a left thigh mass. It is a rare zoonotic disease in human beings, caused by incidental infection of the genus *Spirometra* species.

Introduction

Sparganosis is a rare zoonotic disease in humans. It is caused by infection of the third-stage plerocercoid larva (sparganum) of pseudophyllidean cestodes of the genus *Spirometra*. The most common clinical manifestation is subcutaneous sparganosis, while cerebral and ocular sparganosis are more scarce [1, 2]. Most cases were reported in Eastern and Southeastern Asia [2, 3]. This zoonosis is almost neglected in Europe [4]. Histopathologically, identification of the worms is mandatory to make accurate diagnosis and treatment. Therein, we report a typical case of subcutaneous sparganosis.

Case report

A 61-year-old Taiwanese woman presented to our hospital with left thigh mass for 2 years. She had underlying diseases of type 2 diabetes mellitus and hyperlipidaemia. She also had a history of thyroid cancer and received surgical treatment. Two years earlier, she noted a lump at her left thigh. The mass was irregular without surrounding erythema. She went to a local dermatological clinic for help, and she was transferred to the Department of Plastic Surgery of our hospital. Physical examination noted a 4.5 × 3.0 cm mass at left thigh with mild tenderness and

without fluid accumulation. Laboratory data did not show remarkable findings. She received excisional biopsy. The pathological report revealed panniculitis at that time. After the surgery, she still had erythema, swelling, and pain at the same site. She came to our Department of Plastic Surgery again. Physical examination noted a 4.5 × 2.0 cm mass with tenderness. Excisional biopsy was performed this time. Gross examination of the surgical specimen revealed a tan-to-white solid lesion. Microscopically, the lesion revealed areas of fibrinoid necrosis, mixed acute and chronic inflammation, and granulomatous reaction in the dermis and subcutis (Fig. 1). Parasitic larvae were seen focally (Fig. 2A, B). The larvae consisted of external noncellular eosinophilic tegument, as well as internal pale myxoid matrix with longitudinal smooth muscle fibres (Fig. 3A). Specific ovoid bodies with concentric calcified lamellae, termed calcareous corpuscles, were noted (Fig. 3B). The histopathological features were consistent with the diagnosis of human subcutaneous sparganosis. Tracing back to the contact history, she admitted using flesh from an unknown animal as a poultice applied to a wound. She also ate raw fish and undercooked beef.

Discussion

Sparganosis is a zoonosis caused by infections by *Spirometra*, a genus of pseudophyllid cestodes. *Spirometra* species are distributed worldwide, and more than 60 species have been reported. However, only 4 were considered valid, including *S. erinaceieuropeaei*, *S. mansonioides*, *S. pretoriensis*, and *S. theileri* [5]. The first human sparganosis case was described by Dr. Patrick

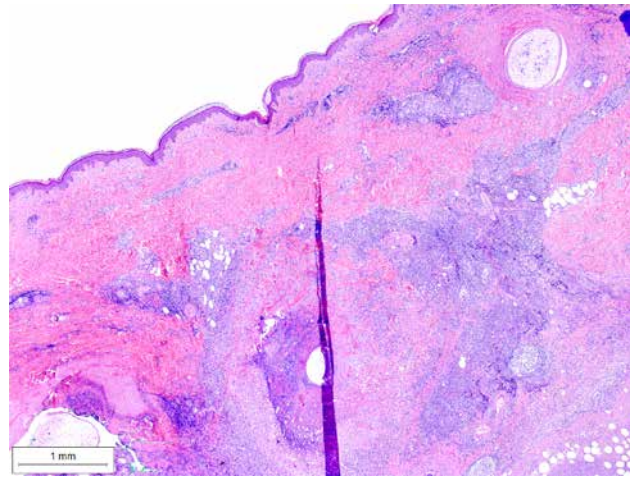


Fig. 1 Histopathologically, low-power view of the lesion revealed fibrinoid necrosis, acute and chronic inflammation, and granulomatous reaction in the dermis and subcutis (HE, magnification 20×)



Fig. 2 Spargana, plerocercoid larvae of *Spirometra*, were focally found in the lesion (HE, magnification 100×)

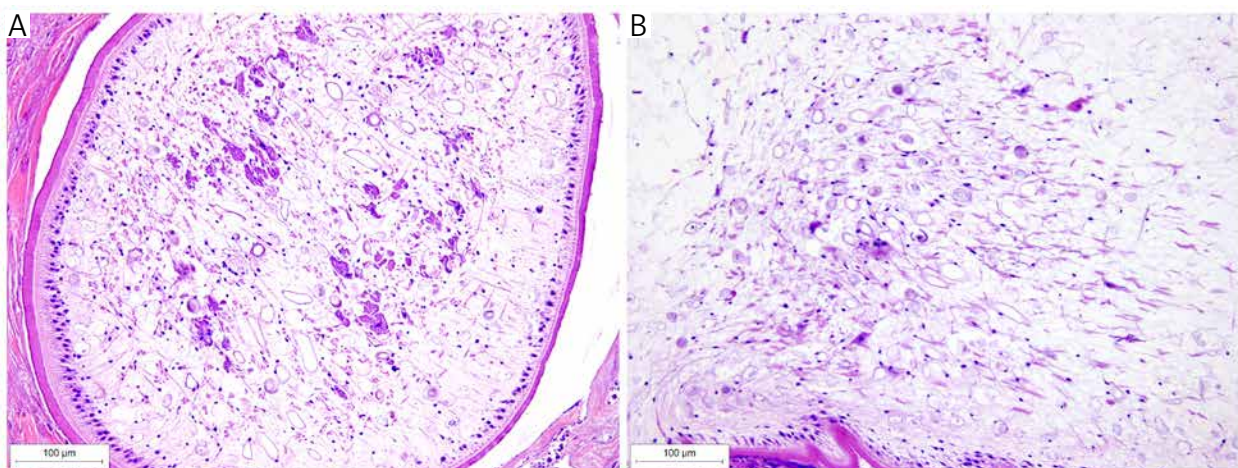


Fig. 3 A) Microscopically, the larvae consisted of external noncellular eosinophilic tegument, and internal pale myxoid matrix with longitudinal smooth muscle fibres. B) Note the specific ovoid bodies with concentric calcified lamellae, termed calcareous corpuscles (HE, magnification 200×)

Manson in 1882 from a Chinese patient in Xiamen, China [6]. The adult worms of *Spirometra* species reside in the small intestine of their definitive host, which can be a dog, cat, raccoon, or other mammal, for a period

of up to 9 years. When the host defecates, the eggs are expelled from the body through faeces. Once in fresh water, the eggs hatch and release coracidia. These coracidia are then consumed by copepods, which act

as the first intermediate hosts. Inside the copepods, the coracidia develop into procercoid larvae and dwell within the body cavity until the life cycle progresses. The second intermediate hosts in this cycle are fish, reptiles, or amphibians that ingest the copepods while drinking water. The larvae penetrate the intestinal tract of the second intermediate host and transform into plerocercoid larvae. They migrate and encyst themselves within the subcutaneous tissues and muscles of the host. If this second intermediate host is consumed by a paratenic host, the plerocercoid larvae do not advance to the next developmental stage but instead re-encyst themselves in the subcutaneous tissues and muscles of the new host. If the second intermediate hosts are eventually consumed by definitive host predators, typically cats and dogs, the cycle begins anew. Humans are accidental hosts in this cycle and can become infected with the plerocercoid larvae by ingesting either the first or second intermediate hosts. Once inside a human, the larvae migrate to the subcutaneous tissues or other organs. However, no further development occurs, and humans are incapable of transmitting the disease [1, 4, 7].

Histopathological examination is the gold standard for the diagnosis of sparganosis. Serological tests provide valuable support in diagnosing sparganosis; however, it is important to note that these tests may produce cross-reactivity with other cestodes that are prevalent in regions where sparganosis is endemic [8]. Complete surgical removal of spargana is the most effective therapy. If the infectious site is not suitable for surgery, anthelmintic drugs should be administered [1]. Although most reported cases were found in Eastern Asia and generally neglected in Europe, a few European autochthonous human sparganosis cases were described, including in Poland [9]. It is important for histopathologists to identify this infectious disease, make an accurate diagnosis, and subsequently help the patients with adequate therapies.

Disclosures

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4. Conflicts of interest: None.

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Address for correspondence:

Dr. I-Wei Chang, MD, PhD
 Department of Pathology
 Taipei Medical University
 250, Wuxing Street, Xinyi District
 Taipei 110, Taiwan
 Phone: +886-2-27361661 ext. 3140
 Fax: +886-2-23770054
 e-mail: 108098@w.tmu.edu.tw