

## A case report of perineal sparganosis mansoni from an endemic region

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**Abstract.** Sparganosis mansoni is a parasitic disease caused by the larva of *Spirometra mansoni*, which occurs worldwide, especially in Asian countries. In recent years, there have been cases of sparganosis reported in Guizhou province in southwest China. In this report, a case of sparganosis mansoni with a painful mass in perineal region is presented. The patient is a 49-year-old Chinese woman, she did not have a history of eating raw frogs or snakes. But she drank unboiled water from rivers and streams. Physical examination showed her vital signs were stable and blood routine examination was also regular. After surgery to remove the mass, a worm was removed from the foreign matter, which measured about 10 cm in length and 2 mm in diameter, that was identified as *S. mansoni*. One week after the operation, her symptoms changed significantly. To our knowledge, this is the first case of perineal sparganosis mansoni in Guizhou. Surgical removal is effective for the treatment of perineal sparganosis, followed by a course of therapy.

### INTRODUCTION

Sparganosis is a parasitic disease caused by the plerocercoid larvae of the genus *Spirometra* that includes at least four diphyllbothroid tapeworms (Ou *et al.*, 2010; Anantaphruti *et al.*, 2011). It was first described from China in 1882, and the first human case was reported in Florida 26 years later (Lescano & Zunt, 2013; Read, 1952). The infection is acquired by ingestion of contaminated water or a second intermediate host such as a frog or snake (Hughes & Biggs, 2002; Johnson *et al.*, 2015). Humans are accidental hosts in the life cycle, and rare infections have been reported. Although sparganosis mansoni has been reported sporadically worldwide, its prevalence is higher in several Asian countries, including South Korea, Japan, Thailand and China. Cases of sparganosis have been reported in viscera, extremities, eyes, brain, spinal canal,

but very few cases of sparganosis mansoni have been reported in perineum (Garcia & Shore, 2007; Cheng *et al.*, 2014; Li *et al.*, 2011). Here, we presented a case of perineal sparganosis mansoni from an endemic region in the southwest China.

### CASE REPORT

The patient is a 49-year-old Chinese woman, who presented with a perineal mass and paroxysmal pain. She is a farmer from a rural area in Guizhou province and never travelled overseas. She always drank unboiled water and was in contact with wild herbs gathered at the water's edge frequently. However, she had no habit of eating raw meat, freshwater fish, frogs, or snakes. About 2 weeks prior to the presentation, she was diagnosed with a mass in her perineal regions, but there was a lack of standard diagnosis and treatment

before being admitted to the hospital. On admission, her vital signs were stable with normal mentation, and neurologically she was lucid with no abnormality. Specialized medical examination presented a mass in mons pubis, that appeared as solitary and firm, and the size of the mass was about 4 cm×3 cm. The skin around the mass appeared folded, red and puffy. The mass was movable, and if touched, the pain get worse, the temperature of the skin was slightly high, but there was no laceration and oozing of blood on the surface of the perineal mass.

After relevant examination, the patient was suspected of being infected by parasites, following which she had surgery to remove the mass, the bleeding was stopped by ligature and suture, the fluid replacement was used to support symptomatic treatment. The foreign matter was sent to pathology department for examination, a worm was drawn out and removed from the mass, the extracted worm, was about 10 cm in total length and 2 mm in diameter (Figure 1), was identified as *S. mansoni*. The worm is a long band-like in shape, and it has an unsegmented body with irregularly wrinkled stripes. There is a hole in the middle of the top of the parasite that is recessed into a tunnel, the hole extends a certain distance to form a blind end. The histology structures showed the larva is acoelomate, has a characteristic body wall, and the body wall consists of the cortex and the subcutaneous layer. In the stroma (fibrous tissue), there are annular and longitudinal

muscle fibers, tissue cells, excretory duct and round or oval calcareous corpuscles (Figure 2). All the above features are similar to those descriptions about *S. mansoni* in previous cases, so the patient was further diagnosed with perineal sparganosis mansoni.

After treatment, the symptoms gradually disappeared. One week later, the materials from the physical examination showed there was no obvious abnormality in heart, lung and abdomen. There was no edema in both lower extremities, the temperature of the skin was normal, and there was no laceration and oozing of blood on the operative site. The patient presented with normal vital signs and was discharged from hospital and followed up for observation (Figure 3).

## DISCUSSION

Sparganosis was reported sporadically around the world, and there was a higher prevalence of the disease in several Asian countries, including South Korea, Japan, Thailand, and China (Li *et al.*, 2011; Shin *et al.*, 2008; Wiwanitkit, 2005). In China, human sparganosis is endemic in south China, and majority of cases have been reported from southern provinces, such as Guangdong, Henan, Fujian, Hunan, Guangxi, and Hubei (Li *et al.*, 2011). Sparganum can live in any part of the human body including the eyes, face, neck, limbs, subcutaneous tissues of breast and abdomen, liver, lungs, and kidney (Xu

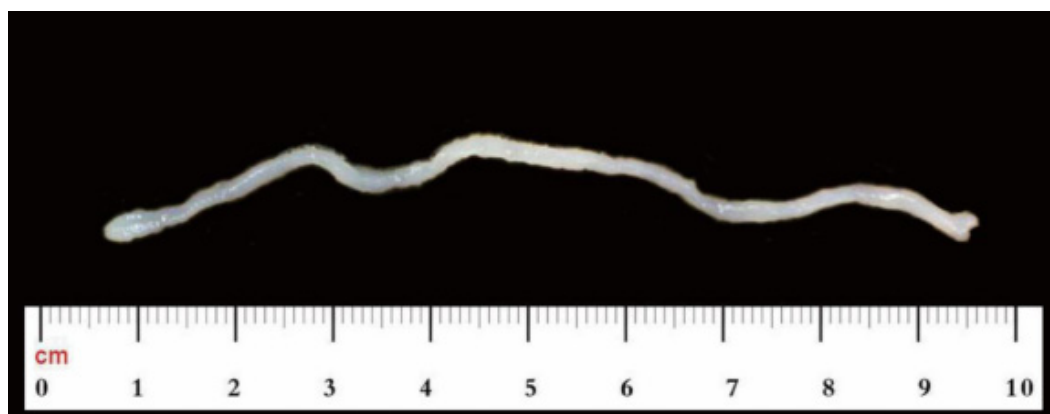


Figure 1. *Sparganum mansoni* worm extracted from the patient, measuring about 10 cm in length and 2 mm in diameter.

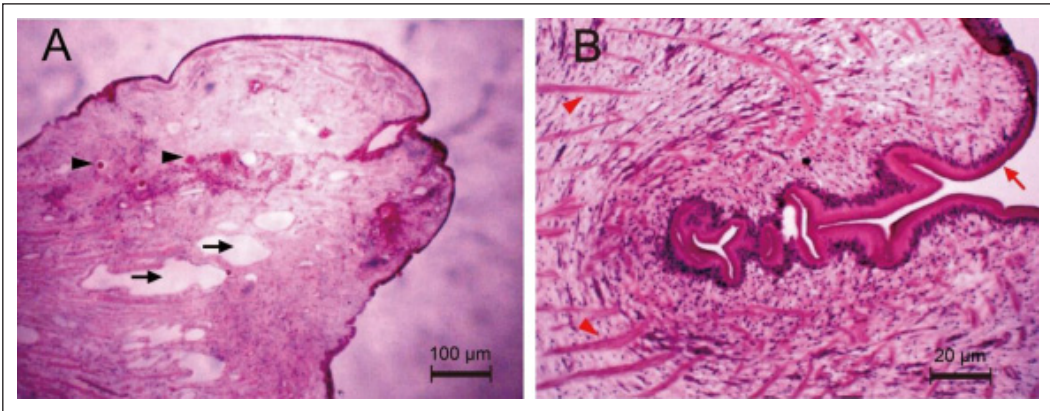


Figure 2. The histology structures of plerocercoid from perineal mass (hematoxylin and eosin stains). A, microscopic findings of sparganum indicated basophilic calcareous bodies (black arrowhead) and thin-walled excretory ducts (black arrow); B, the histology structure showed there is a hole in the middle of the top of the parasite that is recessed into a tunnel, and muscle fibers (red arrowhead) were identified in the worm, which was surrounded by a thick tegument (red arrow).

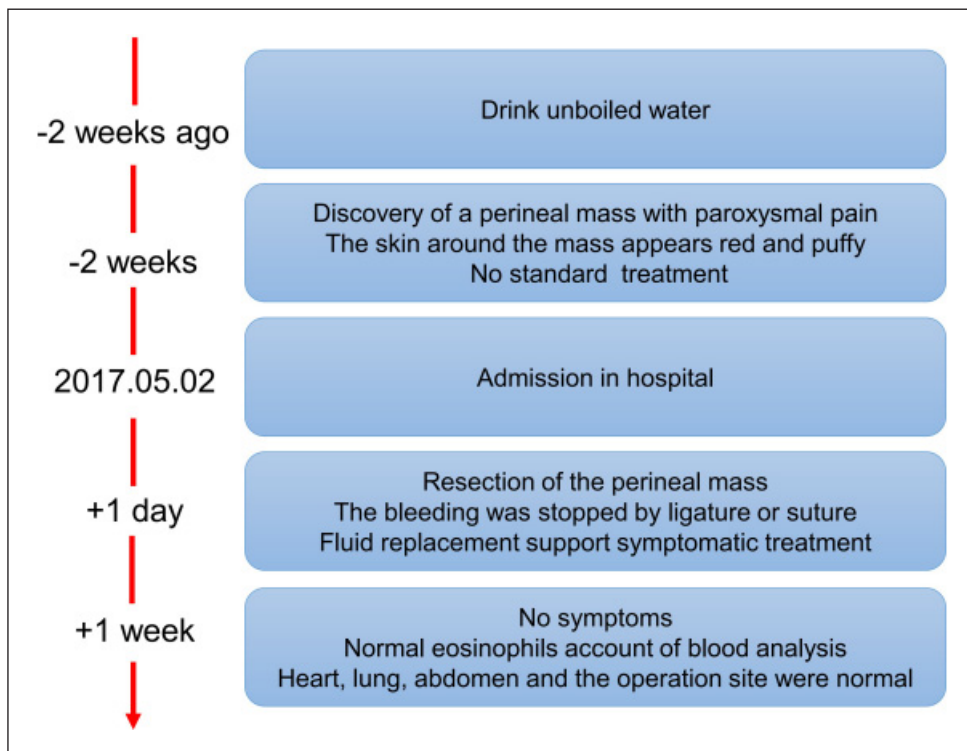


Figure 3. The timeline of diagnosis, treatment and follow-up.

*et al.*, 2009; Mo *et al.*, 2013), but rare in mons pubis and perineal regions. This case will improve the understanding about the clinical presentation of this rare cestode infection. The primary source of infection with

sparganum are by eating undercooked meat of snakes or frogs infected with sparganum, or using poultices of frog or snake flesh or skin on open wounds (Li *et al.*, 2011; Murata *et al.*, 2007). In endemic area,

Table 1. Comparison of the findings before and after correct diagnosis of patient

	History	Symptoms	Signs	Blood analysis	Physical examination	Ultrasound examination
Before diagnosis	Drink unboiled water	Painful nodule and the skin appeared red and puffy	No urinary frequency and urgency and no fever	WBC count $7.76 \times 10^9/L$ Eosinophil count $0.09 \times 10^9/L$	T 36.5°C R 20/min P 80/min BP 115/67 mmHg	No abnormal sign in heart, lung and stomach
After therapy	–	No	No	Normal	Normal	Normal

T, body temperature; R, respiratory rate; P, heart rate; BP, blood pressure.

drinking unboiled water contaminated with proceroid-infected copepods is also an important reason to cause this disease.

The diagnosis of sparganosis is usually made by recovery of a sparganum from infected tissue. The white, ribbon-like larva can measure from a few millimeters to tens of centimeters in length and up to 0.1 to 0.4 centimeters in width (Kim *et al.*, 2001). In theory, a pre-operative diagnosis could be made by identification of a single painful, migratory, subcutaneous nodule firstly. So sparganosis is typically diagnosed following surgical removal of the worms, and the infection could be further identified by the morphology and histology of parasite in a tissue specimen. If such biopsy and excision procedures are not feasible, the anti sparganum ELISA test may be used, CT and MRI scans are especially useful for diagnosis of cerebral sparganosis and visceral sparganosis (Walker & Zunt, 2005). Clinicians should be experienced in different sparganosis mansoni cases so as to make the right diagnosis.

Although anthelmintic medications such as mebendazole, albendazole, or praziquantel are used for treatment of this organism infection, complete surgical removal of the larva is the optimal treatment and is often curative especially for cutaneous sparganosis (Johnson *et al.*, 2015). In more advanced cases of proliferative or disseminated sparganosis, such as cerebral sparganosis and visceral sparganosis, these pharmacologic agents have been used in combination with surgical removal with

varied success (Ou *et al.*, 2010; Cheng *et al.*, 2014). To our knowledge, perineal sparganosis was rare reported before. Although there was no convincing references for clinical project of treatment, all the previous reports have reference value to the diagnosis and treatment of this case. The patient was properly diagnosed due to its clinical features and the patient is recovering well following treatment by surgical removal (Table 1). It indicates that surgical removal is effective for the treatment of perineal sparganosis, followed by a course of therapy. The best way to prevent the disease is for people to change their bad eating habits, such as avoiding drinking unboiled water and eating uncooked meat in endemic regions.

## CONCLUSIONS

Sparganosis is a systemic infection that can cause various symptoms, so it should be considered as a differential diagnosis. Although perineal sparganosis is rarely reported, this case suggested that *S. mansoni* can parasitize any part of human body. The treatment which was carried out can be helpful for the diagnosis of perineal sparganosis especially in endemic areas. Surgical removal of the worm is the best treatment and with good outcomes.

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