Vol.2, No.9, 530-534 (2013)
http://dx.doi.org/10.4236/crcm.2013.29138

Subcutaneous larva migrans of a sparganum in a gnathostomiasis-endemic area*

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Received 27 September 2013; revised 20 October 2013; accepted 19 November 2013

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ABSTRACT

We report a case of subcutaneous larva migrans caused by a tape worm (sparganum), a rare and under-recognized food-borne parasitic zoonosis, in Nan Province, Northern Thailand. An obese 56-year-old female presented a 3-month history of an intermittent migrating subcutaneous nodule in the abdomen associated with pain and itching. Physical examination showed a tubal resection scar on the lower abdomen with a swelling and movable irregular, and firm nodule with ill-defined border at the right lower quadrant of abdomen. Blood examination showed slight increase of eosinophil levels (6%). Excisional biopsy showed a viable non-segmented tape worm in the subcutaneous tissue identified by morphological examination and Western blot technique as a sparganum. Surgical removal is recommended for a worm in the subcutaneous connective tissues and superficial muscles.

Keywords: Subcutaneous Larva Migrans; Sparganosis; Zoonosis; Northern Thailand

1. INTRODUCTION

Thailand is located in tropical Southeast Asia, a region that is both hot and humid, which is suitable for the life cycles of many parasites, including protozoa, helminths and medical insects. Owing to the specific lifestyles, primitive culture and local specialty dishes of some Thai social groups, particularly poor rural communities with risky food habits, parasitic infections remain a health problem. Moreover, population migration has introduced parasites into new localities that may favor their continued existence and proliferation, such as among available intermediate hosts. Among humans infected with tissue helminths, the larval stage may migrate to where the characteristic eggs are not detected by stool examination, and be reported as a parasite in human subcutaneous tissues. In Thailand, the clinical manifestation of larva migrans is an intermittent, circumscribed swelling and this is normally a manifestation of gnathostomiasis, a disease historically prevalent in the Mekong region of Southeast Asia [1]. Misdiagnosis may occur when other tissue helminths, such as spargana, infect humans in areas where raw dishes of freshwater fish and amphibians are popular [2]. Correct laboratory identification of the responsible parasite will inform accurate diagnosis, resulting in prompt and appropriate treatment and management.

2. CASE REPORT

An obese 56-year-old female (BMI 27.06), not pale, without jaundice, showing good consciousness and cooperation, who had been residing in Ban Luang District, Nan Province, Thailand, presented with a 3-month history of a migrating subcutaneous nodule in the abdominal region, with some related pain and itching. Whenever she scratched due to the itching, the nodule developed the appearance of an erythematous acne rash; some rashes also developed in the area of the swelling. She had

*Competing interests: The authors declare that they have no competing interests.

no fever, cough, or weight loss, and had started menopause in 2011. She used to consume partially cooked or raw dishes of many kinds of freshwater fish and frogs, wild boar, and cattle. She did not keep any domestic animals/pets in her house. Physical examination showed normal vital signs, no edema in the extremities, and all body systems within normal limits, except for a tubal resection scar on the lower abdomen. A swelling and movable irregular and firm nodule measuring 2 cm in diameter, with an ill-defined border in the right lower quadrant of the abdomen was neither tender nor inflamed. Complete blood count showed unremarkable values, with a total white cell count of 8.350 cells/mm^3 (6% eosinophils, 40% neutrophils, 44% lymphocytes, 9% monocytes, 1% basophils), 40% hematocrit, platelets 330,000 cells/mm^3. An excisional biopsy showed a viable non-segmented tapeworm in the subcutaneous tissue, measuring 1.92 cm × 0.18 cm (**Figure 1**). The pathological report on the surrounding subcutaneous tissues showed chronic suppurative granulomatous inflammation with foreign-body reaction; no residual parasite material was obtained. The excised parasite was identified by morphological (**Figure 2**) and Western blot techniques. The results showed the specimen to be a sparganum in the family Diphyllobothriidae, for which praziquantel or niclosamide are effective drugs [3-5]. While surgical removal of the sparganum was the most effective treatment in this case and there was no swelling and movable nodule found anywhere for the follow up of the patient. The identification of the removed worm was the definitive diagnosis, it may not be preferred if the parasite is embedded deeply in tissues and can invade vital organs [6].

3. DISCUSSION

This case illustrates sparganum as a visceral larva migrans that can cause misdiagnosis of gnathostomiasis in endemic areas such as Thailand. Owing to their similar life cycles and infective stages, where both worms can inhabit the same group of second intermediate hosts or paratenic hosts, humans remain a potential target for infection by both sparganum and *Gnathostoma*, or even both worms at once. Laboratory investigation can be used to distinguish between these two helminthic diseases, to facilitate diagnosis and proper treatment. Treatment failure with albendazole can occur, as benzimidazoles are not recommended drugs for use with sparganum [3-5]. This case highlights how a migrating subcutaneous nodule of sparganum can be an important etiologic pathogen of differential diagnosis. It is a challenge for clinicians in remote areas to perform a tissue excisional biopsy, where indirect confirmatory tests are not available. Western blot shows a weakly positive reaction at 35 kDa specific reacting band of specific IgG4 antibodies against antigens derived from *Spirometra erinacei* sparganum after investigation of the patient's serum (Dekumyoy, personal contact and unpublished data). Identification after excisional removal of the parasite in tissue specimens for detection of eosinophilia is a more reliable approach for confirmation of sparganosis [7]. In this study, results of the pathological report of the tissue biopsy did not show any residual parasitic material, which could exclude rare sparganosis prolifera of the budding larvae as new spargana in the tissues. As sparganosis and gnathostomiasis are both food-borne zoonotic diseases, they can be exported to other countries via international travel. Both these helminthic diseases can spread into non-endemic areas.

Sparganum is the generic name of the plerocercoid, which is a tissue-migrating larva of canine/feline tapeworms of the *Diphyllobothrium* and *Spirometra* species. Sparganum refers specifically to larvae of *Spirometra*

Figure 1. Excisional biopsy of right lower quadrant abdomen showing sparganum.

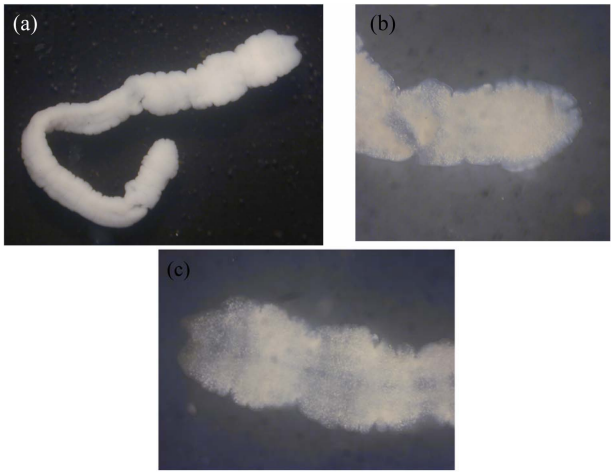

Figure 2. Morphology of sparganum. (a) Elongate, cream-white and ribbon-like unbranched sparganum, measuring 1.92 cm in length and 0.18 cm in width; (b) a candal part with no internal structures or other morphologic features that can be distinguished for species identification; (c) an anteroposterior polarity, called a pseudosucker with no scolex.

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spp. in humans, which are known to cause sparganosis of subcutaneous and visceral larva migrans [8]. *Spirometra* adult worms inhabit the small intestine of dogs, cats and wild carnivores, but rarely humans (spirometrosis). Copepods (freshwater crustaceans) are the first intermediate host for development of procercoid; various kinds of vertebrates (amphibians, reptiles, birds and mammals) then serve as the second intermediate host for development of plerocercoid, to complete the life cycle [9]. Sporadic cases of sparganosis in humans have been reported, mainly because of their scarcity but also because of the difficulties around identification; there were only 52 cases of human sparganosis reported in Thailand between 1943 and 2010 [10]. Although sparganosis is generally thought to be distributed all over the world, *Spirometra* spp. seem to be responsible for the larval plerocercoid stage and in humans rarely develop to adulthood; it appears parasitic in muscles or body cavities. However, ocular and cerebral involvements have been reported in relation to sparganosis lesions found in Thai patients particularly cerebral sparganosis accompanied by brain abscess, seizures, hemiparesis has poor prognosis [10]. The larval plerocercoid can mostly be seen as a white, string-like creature in the muscles or body cavities of birds (chickens, ducks), reptiles (snakes), amphibians (frogs), and small rodents, which are considered to be important sources of infection. In addition, these hosts and humans can also be infected by drinking contaminated water with cyclops-carrying procercoids, consuming inadequately cooked meat containing the second-intermediate or paratenic host carrying plerocercoids, and also suffer penetration of cutaneous tissues from poultices made of the flesh of frogs or snakes as dressings for open wounds and eye sores [9,11,12]. Lesions and clinical symptoms of sparganosis have much in common with those of gnathostomiasis spinigerum; the subcutaneous migrans is usually found as a migrating nodule, varying in size, with occasional slow migration.

Gnathostomiasis is an important zoonosis with wide distribution from tropical to temperate zones; it can be fatal to humans if vital organs are affected [3]. The disease is widespread across Ecuador and Mexico in Latin America [13,14], and Laos, Vietnam and Thailand in Southeast Asia [2,15]. *Gnathostoma spinigerum* is particularly prevalent, yet remains the only etiologic agent of human infection in Thailand so far [16]. Adult worms are generally 2 - 3 cm long, with spines covering the cephalic bulb to the posterior end, and inhabit tumors of the gastrointestinal wall in fish-eating mammals. Common intermediate hosts are cyclops and freshwater fish (eels, catfish, snake-head fish), reptiles (snakes) and amphibians (frogs), which develop by hatching from eggs into infective advanced third-stage larvae, which themselves sometimes may be transferred to paratenic hosts such as chickens, ducks, or pigs. Humans mostly get infected accidentally by eating the infected flesh of intermediate and paratenic hosts, in whom worms do not develop into the adult stage. There are two other routes of infection, as well, via skin penetration and prenatal infection [2]. The most common manifestation is intermittent migratory circumscribed swelling, with associated redness, pain and itching in the subcutaneous tissues; this tends to subside and reappear elsewhere near the original site. In rare cases, the patient may suffer seizures, paralysis, unconsciousness, or even death due to severe damage of the CNS; invasion of the eye can also result in visual impairment [17]. Meanwhile, immature worms have been found using excisional biopsy methods in some patients, as migratory swelling tracks have been left behind after the worms' movement. Although worm removal is a good method of treating gnathostomiasis, it remains impractical as it often fails to remove all of the etiologic worms present. Therefore, indirect investigation in the laboratory is performed to give a tentative diagnosis of gnathostomiasis. One method is to use specific monoclonal antibodies to detect circulating antigens to the *Gnathostoma* worm; another is to use Western blot to detect specific antibodies. However, such tests are not available in remote areas of Thailand. Indeed, tests like these are most frequently carried out in university hospitals, such as the Hospital for Tropical Diseases, Mahidol University. About 1000 suspected cases are diagnosed clinically each year after being sent for confirmatory tests of gnathostomiasis, according to the annual report of the Faculty of Tropical Medicine [18]. Clinicians might decide to start treatment with albendazole (800 mg) for 21 days [19], and observe clinical symptoms with a follow-up drop in *Gnathostoma* antibody titer. It is challenging to perform an excisional biopsy at the swelling area, as worms can always escape from the swollen migration tracks. For our patient, who had been residing in rural areas of Ban Luang District, Nan Province, Thailand, an excisional biopsy was attempted, and was successful in removing the invading sparganum (**Figure 3**). Recently, an alternative treatment with praziquantel (75 mg/kg/day), given in three consecutive doses, has been attempted and the result was successful for visceral sparganosis [5].

4. CONCLUSION

The sparganum can be an important etiologic pathogen of differential diagnosis in migrating subcutaneous nodules in endemic areas like Thailand. Clinical manifestations of gnathostomiasis and sparganosis are alike, in terms of the migration of the immature worm, known as subcutaneous larva migrans. This remains a cause of migratory swelling in patients, which is the common symptoms presented, and is the result of increased glob-

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Figure 3. Post-operation showed no swelling subcutaneous tissue (arrow) with distended lower abdomen.

alization and proliferation of food-borne zoonotic diseases. The challenges surround the difficulties of performing tissue excisional biopsies, and indirect confirmatory tests are not available in remote areas. Rapid and accurate diagnosis is important for prompt treatment and appropriate case management, so imported cases can be dealt with efficiently by physicians in non-endemic areas and countries.

5. CONSENT

Informed consent form was required and written by the patient for the allowance to be published in the journal.

6. ACKNOWLEDGEMENTS

The authors owe very special thanks to the Faculty of Tropical Medicine, Mahidol University for supporting the budgets for field trip survey. The appreciation goes to Dr. Saravuth Konggarnkha for pathological examination of the biopsied tissue. Many thanks also to Mr. Paul Adams for editing the manuscript.

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