Sparganosis in Male Breast

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Abstract: Humans can acquire infection by sparganum or plerocercoid larva of the tapeworm from drinking water containing infected *Cyclops* species and by ingesting infected snakes, birds, or other mammals. Once infected, the plerocercoid larva migrate slowly in the tissues and present as a subcutaneous lesion, making the correct diagnosis difficult. A 38-year-old man visited our clinic due to a breast mass. Due to suspicion of gynecomastia or breast cancer, he underwent tumor excision. Unexpectedly, a long white worm was found in the breast tissue and the pathologic finding showed sparganosis. After the surgery, mebendazole was prescribed for 1 week. No recurrence of the sparganosis was found during 1-year follow-up.

Key words: Breast; Case reports; Differential diagnosis; Sparganosis; Spirometra

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Sparganosis is an incidental extraintestinal infection caused by plerocercoid larva that is associated with the development of spargana or its cysts in the subcutaneous connective tissue and superficial muscles. ¹⁻³ This uncommon disease is found throughout the world. Once sparganum larvae infection occurs, the larvae proliferate, often in the subcutaneous tissues, and become encysted in large nodules. These nodular lesions develop slowly, may be found in any part of the body, and may resemble the subcutaneous tumor like a lipoma, fibroma, or sebaceous cyst. ⁴ We report a case of sparganosis in the left breast tissue that was initially treated as suspected gynecomastia or breast cancer.

Case Report

A 38-year-old man visited our clinic in July 2002 because of a painless and firm mass in the left breast for 1 month. Physical examination revealed a $2.0 \times 2.0 \times 1.0$ cm tumor at 2 o'clock position of the left breast. Chest film showed no definite active lung lesion and normal heart size and configuration. Under the impression of gynecomastia or breast cancer, an excision biopsy was performed in August 2001, which revealed a long white worm in the breast mass, 25 cm in length. The frozen pathologic examination showed sparganosis with acute and chronic inflammation in the breast tissue. The anatomic structures of sparganum were seen, including the bothrium

(Fig. 1),⁵ tegument with microvilli, the layer of smooth muscle, tegumental cell, and calcareous body (Fig. 2).⁵ After the surgery, mebendazole 100 mg twice a day was prescribed for an additional 1 week. The postoperative course was uneventful, and he was discharged 3 days later. Tracing back the history of this patient, he had lived Canton, China on a business assignment during the two years before admission and reported having eaten uncooked snake gallbladder in May 2002. He returned to Taiwan in June 2002. No recurrence of the sparganosis was found during outpatient department follow-up for 1 year.

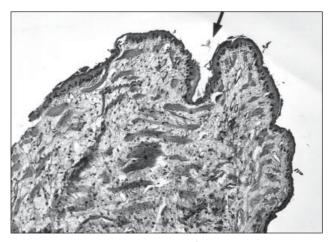


Fig. 1. Microscopic appearance of the surgical specimen. Section through the bothrium (arrow) of the sparganum (hematoxylin and eosin stain, \times 100).

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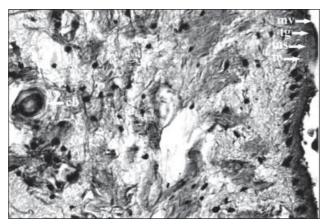


Fig. 2. Microscopic appearance of the surgical specimen. The body wall of sparganum consists of a tegument (tg) with microvilli (mv), the layer of smooth muscle (ms), and tegmental cells (tc). Calcareous bodies (cb) are usually present in the parenchyma (hematoxylin and eosin stain, \times 400).

Discussion

The spargana are stages in the life cycle of a tapeworm. The cycle requires 2 intermediate hosts. The first intermediate host is a copepod (planktonic crustacean) that ingests ciliated embryos (coracoidium) developing from the tapeworm's eggs,⁵ and the second is a fresh-water fish, frog or snake that ingests the infected copepod with procercoid. Sparganum are the larvae of tapeworms of the genus Spirometra that are common in various canines and felines. Ingestion of these larvae produces sparganosis in humans, since the larvae cannot mature in this abnormal host.⁶ In addition, humans can be infected by the sparganum or plerocercoid larva of another diphyllobothrium tapeworm. Humans acquire sparganosis mainly by ingestion of larvae contained in raw or insufficiently cooked meat of animals, which were infected with spargana, such as amphibians, reptiles, birds, pigs and other wild mammals.7 Infection could also be acquired by drinking water containing infected Cyclops species. In China or other areas of Asia, men could be infected by the traditional practice of using frog meat to cover wounds or eyes. In our patient, ingestion of the infected raw gallbladder of snake may have been the pathway of infection. Once infection occurs, the worm migrates slowly in the subcutaneous tissue, and the infection commonly presents as a subcutaneous nodule and resembles the subcutaneous tumors like a lipoma, fibroma, or sebaceous cyst.¹⁻⁴ The nodule may be itchy, inflamed or painful. Other tissues, like lung or periorbital tissues, have been reported to be involved, and pulmonary or ocular sparganosis may destroy the lung⁸ or the eye.⁷ Migration of the plerocercoid to the breast tissue and formation encysted nodule is rare, which makes the differential diagnosis with other breast diseases, like breast cancer, difficult but important.⁹⁻¹²

Physical examination or image studies, such as sonogram or computed tomography, may facilitate the correct diagnosis of sparganosis. The definite diagnosis requires pathologic examination, and treatment with surgical excision of the encysted nodules of the sparganosis is advised. However, the key to prevention of parasite infection is to avoid ingestion of contaminated water and uncooked food.

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