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A Case Report and Literature Review of Breast Sparganosis

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Abbreviations:

CP: Cysteine Protease; CT: Computed Tomography; MRI: Magnetic Resonance; PET/CT: Positron Emissiontomography–Computed Tomographic; PCR: Polymerase Chain Reaction; ELISA: Serum Enzyme-Linked Immunosorptive Assay

1. Abstract

Sparganosis is a global zoonotic parasitic disease caused by sparganum, a plerocercoid tapeworm larva of the genus Spirometra. Among them, breast sparganosis is a rare disease, accounting for less than 2% of the total number of human cases. The major common clinical presentation is the breast tumor-like mass, and preoperative diagnosis is typically challenging in most cases. A thorough history examination and imaging analysis are crucial basic foundation for preoperative diagnosis. Complete surgical excision is a radical and effective treatment. Postoperative pathology usually reveals a foreign body reaction, a granulomatous inflammation, along a tract-like, elongated cavity through which the worms have passed. Whether or not post-operative medication is necessary is up for dispute. We report a case of breast sparganosis that was completely surgically excised and confirmed.

2. Background

Sparganosis is a global zoonotic parasitic disease caused by sparganum, a plerocercoid tapeworm larva of the genus Spirometra [1]. Since the first human case was reported in 1908 by Charles Wardle Stiles of Florida, most cases have occurred in China, South Korea, and Japan [2]. The Spirometra tapeworm requires two intermediate hosts and one definitive host throughout its life cycle. Sparganum can grow into adults in the intestines of its definitive hosts, carnivorous domesticated, and wild animals (such as cats and dogs). The eggs produced by the adults are excreted in the feces and hatch into procercariae under appropriate conditions. After being consumed by the first host (the cyclops), it passes through the intestinal wall into the bloodstream and develops into a larva of procercaria. Then it will develop into the sparganum when the second intermediate host (including snakes, frogs, fish, birds, and many other animals) swallows the infected Cyclops. Eventually, when the final host swallows sparganum, it develops into an adult in the gut and continues to the next life cycle [3]. Humans become infected as accidental secondary hosts when they prey on first or second intermediate hosts, drink untreated water, or use raw meat as a topical medicine. The spargana are released in the gut and pass through the intestinal wall. However, it cannot develop in the human body as an adult and migrate to subcutaneous and other tissues (e.g., eyes, brain, abdominal cavity, and spinal cord) [4]. It can induce local tissue damage, blindness, paralysis, and possible death. The spargana have been reported to survive in humans for up to 20 years [5]. Therefore, the most common clinical manifestations are slow-growing, non-fluid, or rubbery subcutaneous nodules. Therefore, the most common clinical manifestations are slow-growing, non-fluid, or rubbery subcutaneous nodules. When larvae invade human tissues, they cause inflammatory edema in the stroma. When the larvae die, they cause intense local inflammation and necrosis. Among these, breast sparganosis is a rare disease, accounting for less than 2% of the total number of human cases [6]. The main common clinical presentation is a breast tumor-like mass, and preoperative diagnosis is very difficult in most

cases [7]. Here we report a case of breast sparganosis caused by a sparganum of Spirometra.

3. Case Presentation

A 46-year-old Chinese woman presented with a painful mass of the left breast 1 month ago. She reported that the mass was bigger when she felt pain and did not have any nipple discharge or systemic symptoms. Physical examination revealed a mobile, tough mass of approximately 4cmx3cm in the upper lateral quadrant of the left breast without any palpable axillary lymph nodes. The skin was locally intact, but the boundary of the mass was unclear. Breast ultrasonography showed a hypoechoic mass of 41mmx13mmx27mm from the subcutaneous to glandular layer in the upper outer quadrant of the left breast (BIRADS category 4b). This was a slight increase from 38mmx11mmx30mm one month earlier. Mammography suggested space-occupying lesions in the left outer upper quadrant with local glandular gathering, BI-RADS category 4c (Figure 1). The mass was highly suspicious of malignancy and was subsequently surgically excised to confirm the diagnosis. Sparganum-like worms measuring 16cm in length and 0.4cm in diameter were detected in the fibrous sac (Figure 2). The organism was referred for gross pathological examination and was identified as a tapeworm. The patient recovered well after surgery and did not receive antiparasitic treatment. No discomfort was observed at 21 months of follow-up.

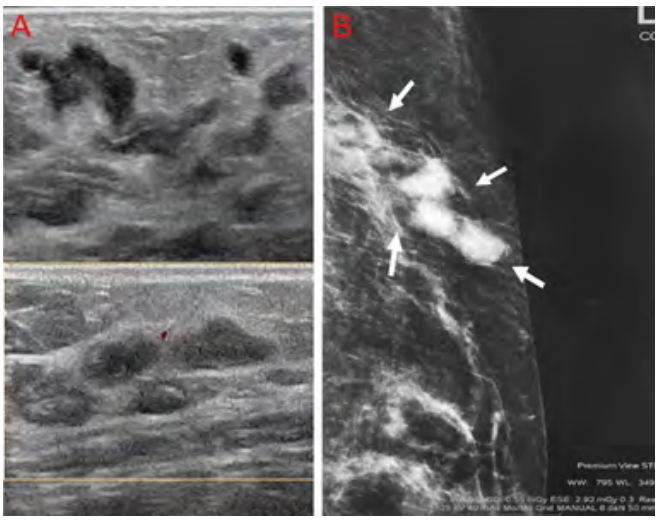

Figure 1: Imaging finding of sparganosis of the breast. (A) Ultrasound finding: sheet or strip area of low echo is seen at the subcutaneous to glandular layer with internal hyperechogenicity; (B) Mammography finding: a tubular elongated curved structure (arrows) sapparent in the subcutaneous fat layer.

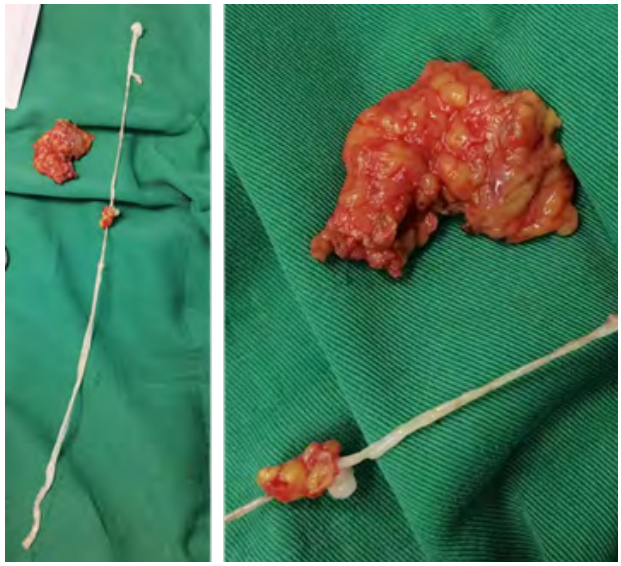

Figure 2: Sparganum-like worms 16cm in length and 0.4cm in diameter were detected in the fibrous sac during the operation.

4. Discussion

The larva of the Spirometra tapeworm is the cause of Sparganosis, which can affect any part of the human body and prefers subcutaneous invasion and migration [8]. There have been more than 60 different Spirometra species reported worldwide, making species identification difficult. Spirometra tapeworms are members of the Diphyllobothriidae family [9]. In addition, mitochondrial DNA, such as the cytochrome c oxidase subunits 1 and 3, and NADH dehydrogenase subunits 1 and 4, is regarded as a useful molecular marker for molecular identification and population genetics studies in spargana [10, 11]. Sparganosis is scattered around the world, mainly prevalent in China, Japan, South Korea, Thailand, and Southeast Asian countries [12]. Since the first case of sparganosis was originally documented in Xiamen, Fujian Province, China in 1882, More than 1,600 human cases of sparganosis have been reported in China, accounting for more than 80% of all reported cases globally [13].

Some people apply fresh snake or frog meat to relieve skin sores and eye inflammation due to different habits and cultural practices, while others choose to consume it raw. Therefore, frogs and snakes become the main source of infection [14]. Spargana actively penetrates the intestinal wall when it enters the human gut, migrating to various tissues and organs [2]. As a result, Spargana produces several proteins that aid in its ability to invade host tissue and evade host immune surveillance to reach its target spot. In the precercarial stage of different hosts, but not in the adult and egg, cysteine protease (CP), a proteolytic enzyme with cysteine residues in the active center, is expressed. Studies have shown that recombinant Spirometra erinaceieuropaei plerocercoids secrete abundant CP to hydrolyze hemoglobin, IgG, and fibronectin. Therefore, it may play a potential role in evading host immune

response, tissue invasion, and migration [15]. Liu et al. also identified Spirometra erinaceieuropaei CP of the sparganum excretory-secretory proteins recognized by early infected sera and found 15 potential antigenic epitopes. This information provides an important foundation for studying the diagnostic antigens and target molecules of antiparasitic drugs [16]. Furthermore, the tegument enolase of Schistosoma mansoni is an adherent-related factor that binds to plasminogen, enhancing plasminogen activation and promoting parasite invasion, and may even be a potential diagnostic molecule for infection [17, 18]. Additionally, it has been claimed that spargana produce a "Sparganum growth factor" or "plerocercoid growth factor" with growth hormone-like effects [19].

There are two distinct clinical kinds of Spargana infection in humans: proliferative sparganosis and non-proliferative sparganosis. The infection S. proliferum, the larval form of an unspecified spirochaeta species, which is divided into cutaneous and internal forms, causes proliferative sparganosis [20]. There have been sporadic incidents reported in Japan, Taiwan, and Thailand. They can branch out and break down into many fragments thanks to their ability to reproduce massively asexually, which facilitates their expansion to new locations. It invades multiple tissues and organs in the human body and disrupts the normal function and nutritional function of tissues and organs, which may be the main cause of poor prognosis and even death. When humans infect the larvae of other Spirometra species, non-proliferative sparganosis affects only one or a small number of sites. It is characterized by the migration of one or several plerocercoids through connective tissues and occasionally reaches the lungs, liver, eyes, and central nervous system. 30% of the population showed slow-growing rubber-like subcutaneous nodules, which were mostly distributed in the lower extremities (35%), trunk (26%), breast (17%), and other tissues [21].

Sparganosis of the breast is a rare disease that usually manifests as a painless breast lump, sometimes as a movable lump [22]. The invasion of larvae induces inflammatory edema within the stroma and results in local intense inflammation and necrosis after death [23]. It is very important to pay attention to the differentiation from clinical malignancy because it is usually the initial diagnosis concern of patients and doctors. Carefully ask the patient about their medical history first. Some patients may have directly consumed infected reptiles, amphibians, or mammals, such as snakes, frogs, or pigs. However, drinking unclean water is now the main source of infection thanks to economic growth and improved sanitation [24]. Secondly, imaging examinations are helpful for preoperative diagnosis. In ultrasound, the worm body is visualized on ultrasound as either a hypoechoic structure within a heterogeneous hyperechoic mass or a tubular hypoechoic structure with or without internal heterogeneous echo [25]. It has been suggested that the echo without internal heterogeneity is formed when the worm migrates within the breast's subcutaneous tissue or fat and may persist even after surgical excision [26]. However, it is important to note that catheter dilation, radioactive edema, and superficial thrombophlebitis may present similarly on ultrasound. A dense solid mass of irregular soft tissue without calcification is typically shown on the mammogram. In addition, there are a few incidental cases of chest CT, breast MRI, or PET/CT, although there is limited evidence of effectiveness in diagnosis or postoperative monitoring [27, 28]. As technology advances, molecular assays, such as PCR and ELISA, may also be useful to diagnose cases with vague symptoms and monitor residual disease after surgery, but they should not be utilized as routine tests [29]. Moreover, calcareous corpuscles characteristic of tapeworms and caseous necrosis have been observed in preoperative biopsy specimens [30]. However, it has been suggested that puncture biopsies may fragment the worm, thus limiting the effectiveness of complete surgical excision and possibly promoting recurrence [24].

Complete surgical excision is a radical and effective treatment. It has been reported that surgical treatment using a vacuum-assisted breast biopsy system is relatively safe and minimally invasive without serious complications(6). Postoperative pathology usually reveals a foreign body reaction, a granulomatous inflammation along a tract-like, elongated cavity through which the worms have passed [31]. There is debate about whether medication is needed after surgery. Whether or not post-operative medication is needed is up for debate. It has been suggested that medication has no part in the treatment of sparganosis [24]. However, some medications, including Praziquantel and Albendazole, have been reported to only have limited impact and are occasionally combined with surgical excision [32]. The location of the worm and incomplete clearance during initial treatment can both play a role in the rarity of postoperative recurrence. Finally, we present a case of breast sparganosis that was confirmed by complete surgical excision.

5. Conclusion

Although breast sparganosis is extremely rare, it is necessary to differentiate it from other breast tumors. The key components of a diagnosis include a preoperative examination of the mass, a detailed medical history, imaging examinations, molecular detection, and puncture biopsies. Complete surgical excision is an important treatment and a definite diagnosis. To avoid missing them when a diagnosis is made, the organs outside the breast must be thoroughly inspected.

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