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Case Report

A case of breast sparganosis: with an emphasis on ultrasonographic findings ☆☆☆

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ABSTRACT

Sparganosis is a rare disease caused by the infestation of the plerocercoid tapeworm larva of the genus *Spirometra*. Human sparganosis is most commonly encountered in subcutaneous fat areas of the abdomen, limbs, and genitourinary tract. Breast sparganosis occur very rarely, accounting for less than 2% of total human sparganosis cases. Because of the disease's rarity, clinical suspicion is essential to reach the diagnosis of breast sparganosis. We present a case of mammary sparganosis in a 58 year-old woman on the ultrasonographic findings. The patient had a painless breast lump with a history of drinking impure water. On ultrasonography (US), we noted four masses, the largest lesion was suspected as sparganosis, and others tended to be benign lesions. The patient was treated following excisions by a US guided Vacuum-assisted breast biopsy system (VABB). The final diagnosis of all lesions was sparganosis.

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Introduction

Sparganosis is an infection caused by sparganum, a plerocercoid larva of a cestode belonging to the genus *Spirometra*. The first human case was reported by Charles Wardell Stiles in 1908 [1]. The incidence of sparganosis in China has gradually increased in recent years [2]. Humans acquire sparganosis by ingesting contaminated water or eating raw or partially cooked fish (typically snakes or frogs) infected with the plerocercoid larvae [3]. It can grow and migrate preferentially into

any part of the human body, such as the abdomen, bladder, epididymis, nervous system, eyeballs, limbs, and lungs [4-5]. However, breast sparganosis is a rare disease that accounts for less than 2% of the total cases of human infections [6]. It is often manifested as a migratory subcutaneous and skeletal muscle mass [7]. These masses are typically associated with inflammation and can mimic soft tissue neoplasm. Ultrasonography (US) has made a significant contribution to the detection and accurate diagnosis of the disorder. We describe a case of breast sparganosis in a 58 year-old woman and dis-

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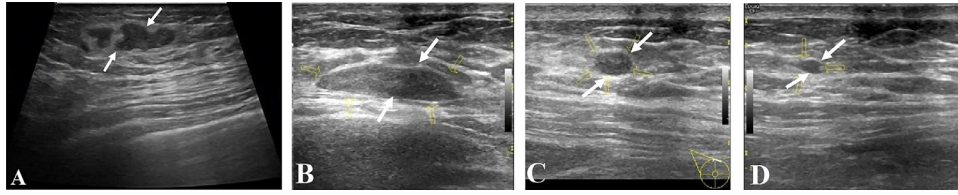


Fig. 1 – Breast ultrasonography examination showing (A- white arrow) hypoechoic, tubular structures with surrounding increased echogenicity in the upper outer quadrant of the right breast, sizing 5.0 cm × 1.7 cm × 0.7 cm, BIRADS category 4b; (B- white arrow) an oval-shaped low-echo mass with well-delineated contours (categorized as BIRADS 4a) at 6 o'clock closely attached to the pectoralis major having a size of 1.8 cm × 1.2 cm × 0.4 cm; (C, white arrow) At 11-12 o'clock, two hypoechoic masses in the fat layer sizing about 0.5 cm × 0.3 cm and (D, white arrow) 0.3 cm × 0.2 cm, respectively.

cuss the sonographic features that can help make a preoperative diagnosis.

Case report

A 58 year-old woman presented with a history of a lump in the right breast for 3 years. She denied any family history of breast cancer and had been menopausal for 5 years. The patient also denied a history of eating frogs or snakes but had a history of drinking impure water. Physical examination revealed discomfort and pain in her armpit and upper outer quadrant of the right breast with migratory pain and sensed a mobile lump.

Breast ultrasonography examination was performed with Logiq E9 (GE Medical Systems, Wauwatosa, WI, USA) equipped with a 13MHz transducer. It revealed the presence of hypoechoic, tubular structures with surrounding increased echogenicity in the upper outer quadrant of the right breast—the most prominent structure was about 5.0 cm × 1.7 cm × 0.7 cm, BIRADS category 4b (Fig. 1A). The ultrasound scan also showed an oval-shaped, low-echo mass sizing approximately 1.8 cm × 1.2 cm × 0.4 cm with well-delineated contours (categorized as BIRADS 4a), located at 6 o'clock and closely attached to the pectoralis major muscle (Fig. 1B). Two hypoechoic masses were seen at 11-12 o'clock in the mammary gland's fat layer, sizing about 0.5cm × 0.3cm (Fig. 1C) and 0.3 cm × 0.2 cm (Fig. 1D), respectively. No obvious blood flow signal was detected in the above lesions on Color Doppler Flow Imaging (CDFI). The largest lesion in the upper outer quadrant of the right breast was suspected to be sparganosis (Fig. 1A). The oval hypoechoic mass located at 6 o'clock and closely attached to the pectoralis major was suspected to be a fibroadenoma (Fig. 1B). While 2 other masses in the adipose tissue layer of the mammary gland were considered to be benign. An ultrasonography-guided 7 gauge Encor Vacuum-assisted breast biopsy system (VABB) was performed, and all the breast masses detected via the US were excised. The largest mass was excised 3 times, while the lesion next to the pectoralis major required 5 excisions, and the two small lesions required 2 passes. The inner diameter of each sample was equivalent to 5.3 mm (0.39 g). The resected specimens were directly put into the bottle with a formaldehyde solution and sent directly to the pathology de-



Fig. 2 – Biopsy of the largest mass revealed multiple spargana with a length of 18 cm, 16 cm, 11 cm, respectively.

partment for testing. Pathologic examination of the largest lesion surrounding the worm revealed inflammatory cells' infiltration, including neutrophils and eosinophils. The worm microscopic findings showed characteristic thick eosinophilic tegument, subtegumental calcareous bodies, and longitudinal strips of muscle, thus, was confirmed as sparganum. It revealed multiple spargana with a length of 18 cm, 16 cm, 11 cm, respectively (Fig. 2). The other three masses were confirmed as parasite eggs. The final diagnosis showed breast parasitosis.

Discussion

Sparganosis mansoni is widely distributed in China, and the foci are more common in southern provinces [8]. Upon entry of sparganum to the human body, it maintains its larval state and migration. When invading human tissues, larvae induce inflammatory edema within the stroma. When the larva dies, it causes strong local inflammation and necrosis [9]. Sparganosis in the breast is a rare disease, in the vast majority of which the migration is invisible. Only irregular masses with local swelling and pain are present [10]. As sparganosis of the breast might be seen as a soft tissue mass, it is sometimes difficult to distinguish from the malignant tumors [11]. Here we reported a rare case of breast sparganosis presented

with a painless lump, among which one lump on ultrasonography was suspected as sparganosis, the other solid mass attached to the pectoralis major muscle as fibroadenoma, and other two masses in the fat layer of the mammary gland as benign lesions.

As we know that the larva prefers subcutaneous migration [12], ultrasonography can be helpful in the diagnosis. On ultrasonography, the hypoechoic, tubular structures are empty tracts resulting from the migration of the spargana larvae, and the echogenic nodules are the larvae themselves or their debris. Some researchers have suggested that the absence of internal heterogenic echogenicity indicates a tract passed through by the worm, which may persist even after surgical removal. Granulation tissue around these structures was observed as a hyperechoic mass [13]. Nevertheless, the atypical imaging manifestations, the varied parasitic sites, and the lack of specific manifestations bring great difficulty in diagnosis. Thus, the differential diagnosis of other tumors is also essential. Clinical suspicion is paramount to reach the diagnosis. In this case, the patient had a history of drinking impure water, and US helped analyzing and characterizing four breast masses. The largest mass in the upper outer quadrant of the right breast displayed hypoechoic, tubular structures with surrounding increased echogenicity, which contributed to the diagnosis of sparganosis. However, no tapeworm movement was evident, potentially due to the worm having remained quiescent during evaluation. The other three lesions' US characteristics were not typical and were small in size; thus, we suspected fibroadenoma and other benign lesions.

Surgical treatment of sparganosis is a fundamental and effective. Anthelmintic medication (eg, praziquantel) is sometimes administered alongside surgical excision of the sparganum [14]. Few cases of recurrence after surgical treatment were reported [15]. Recurrence depends on the worm's location and the its incomplete removal in the primary treatment. VABB is regarded as a feasible, effective, minimally invasive, and safe method for the removal of benign breast lesions, without the occurrence of serious complications [16]. The patient was treated via a US guided Vacuum-assisted breast biopsy system (VABB). Follow-up was done for up to 2 years postoperatively and no specific findings were observed.

Conclusion

In summary, a migrating subcutaneous mass with characteristic US findings such as elongated, folded, tubular structures in a hypo or hyperechoic mass with or without specific clinical history may be a preoperative diagnostic clue of sparganosis [17]. Because of the rarity of the disease, clinical suspicion is vital to reach the diagnosis. This case reminds us when the ultrasound performance is not typical, it should be combined with comprehensive clinical analysis. Furthermore, the

disease's possibility and multiple lesions of the parasite in the ipsilateral breast should also be considered and ruled out accordingly.

Patient consent

The study was approved by the institutional review board of our hospital, and written informed consent for publication of this report was obtained from the patient.

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