Case Report: An Unusual Cause of a Breast Mass in a Patient from China

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Abstract. Sparganosis is a parasitic infection caused by *Spirometra* spp. and often presents as a subcutaneous swelling, most commonly noticed in the abdominal wall or extremities. Amphibians such as frogs ingest infected copepods (crustaceans that have ingested coracidia, i.e., *Spirometra* spp. embryos) and serve as a secondary intermediate host. Complete surgical excision is recommended for definitive diagnosis and treatment. Granulomatous inflammation is the most common histologic finding. Although dissemination can occur, most cases are localized. Serum enzyme-linked immunosorbent assay (ELISA) has been suggested as a potential surveillance tool. Medical therapy with antiparasitic agents, such as praziquantel, is not typically recommended but may be effective at high doses. Preventing recurrence thus depends on adequate surgical removal of the parasite. We report a case of a breast mass caused by sparganosis infection in a Chinese female whose likely exposure was due to frog consumption. The diagnosis was confirmed on surgical excision and no systemic antiparasitic therapy was required.

INTRODUCTION

Sparganosis is a rare parasitic cause of breast masses and often presents as a subcutaneous swelling. The infection derives its name from the sparganum, the plerocercoid larva of a cestode, which belongs to the genus *Spirometra*. Although *Spirometra* can be found worldwide, the majority of human cases occur in China, Korea, and Japan.¹ Amphibians such as frogs ingest infected copepods (crustaceans that have ingested coracidia, i.e., *Spirometra* spp. embryos) and serve as a secondary intermediate host. The definitive hosts for the adult worm are domestic dogs and cats. Humans can develop sparganosis by ingesting contaminated water-containing larvae or by directly ingesting raw amphibians or reptiles.² Infection can also be acquired through direct inoculation of the organisms onto a wound using a poultice prepared from infected amphibians or reptiles, which constitutes a practice that was previously more prevalent in China.³ Our case describes the issues surrounding diagnosis and management of sparganosis manifesting as a breast mass in a Chinese female.

CASE REPORT

A 58-year-old female presented with a 2-week history of a palpable left breast mass. Her past medical history was significant for a left breast mass 2 years prior, which was biopsied and demonstrated a larval form. The lesion spontaneously regressed and she was lost to follow-up until she developed swelling at the prior biopsy site. She otherwise denied systemic symptoms including fevers, chills, night sweats, and changes in appetite or weight and had no other localizing symptoms. She was born in rural China but immigrated to the United States 30 years ago. She reported consumption of frogs purchased from a Chinese grocer as recently as 8 weeks before presentation.

Physical examination revealed a 2 × 0.5 cm soft, non-tender mass, with overlying erythema. Mammography demonstrated a 1.2 × 0.6 cm area of focal asymmetry, with a corresponding irregular, serpiginous, hypoechoic lesion noted on ultrasound

(Figure 1). Her white blood cell count was 4.1 K/ μ L (range: 4–11 K/ μ L) with an elevated eosinophil percentage of 12.1 (range: 0–4%). No ova or parasites were seen on stool studies. Wire-localized surgical excision revealed a live 3-cm tapeworm (demonstrated alongside the excised breast tissue in Figure 2).

The organism was submitted for gross pathologic examination and identified as *Spirometra* spp., consistent with a diagnosis of sparganosis. Histopathologic examination of the excised breast tissue revealed acute and chronic inflammation, focal lymphoid follicle formation, histiocytes, eosinophils, and foreign body giant cell reaction (Figure 3). The patient did well postoperatively, and antiparasitic therapy was not required. At her 3-month follow-up appointment, there was initial concern for some recurrent swelling in the form of a linear area of prominence close to her surgical incision but this was found to be consistent with lymphangitis, which has since resolved.

DISCUSSION

The *Spirometra* spp. larvae most commonly migrate within the subcutaneous tissue to the abdominal wall, extremities, or urogenital system, followed by the central nervous system and orbital area. Cases of breast sparganosis have only been intermittently reported and typically present with a mass, which may be migratory in nature.² Given the indolent nature of these breast masses and usual lack of signs of an inflammatory reaction, malignancy is usually the initial diagnostic concern for both patients and providers. Within the breast, the worm will characteristically be in the subcutaneous fat layer and most commonly occurs in the upper outer quadrant.² Ultrasonography typically demonstrates amorphous, heterogeneous, hyperechoic masses with tunnel-like internal hypoechoic tracts.⁴ Mammography findings are less specific and demonstrate lobulated soft tissue masses that could resemble benign or malignant breast tumors. Calcification can occur and can be misdiagnosed as cysticercosis.⁵ Dissemination is uncommon, and, therefore, diagnostic imaging evaluation should focus on symptomatic areas.

Complete surgical excision is recommended for definitive diagnosis and treatment. There are arguments that preoperative biopsies may fragment the worm, thereby limiting the success of complete surgical removal and potentially promoting recurrence,² which may have been the situation for our

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FIGURE 1. Left breast ultrasound. Targeted ultrasound demonstrated an irregular serpiginous hypoechoic lesion just beneath the skin measuring $1.4 \times 0.4 \times 0.5$ cm with internal curvilinear forms. There is an increased subcutaneous fat echogenicity and 3-mm focal skin thickening consistent with inflammation.

patient. This case adds to the limited body of literature by describing the natural history of sparganosis breast lesions that are not initially surgically excised. Granulomatous inflammation is the most common histologic finding but other morphologic characteristics include a cellular tegument with eosinophilic infiltrations and the absence of a bladder-like center.⁶ Gross pathologic exam reveals features consistent with the larval form of a cestode, which is a parasitic tapeworm with a ribbon-like body that does not possess an intestinal canal. *Spirometra* spp., also known as *Sparganum* spp. (most commonly *mansoni* or *mansonoides*) is typically a white ribbon-shaped wrinkled muscular worm of variable length and width. Multiple worms may be seen.² Other parasites to be kept in the differential diagnosis for infectious causes of breast masses include *Dirofilaria* and *Echinococcus*.⁶

Molecular identification using polymerase chain reaction (PCR) may also be helpful for diagnosis. If *Sparganum proliferum* is identified, further evaluation for the prolifera-

FIGURE 2. Gross pathology specimen. The fragments of the 3 cm visible tapeworm are demonstrated above excised breast mass. This was identified as *Spirometra* spp. based on its white ribbon-like muscular appearance.

FIGURE 3. Histopathology slide. This demonstrates a cyst with associated lymphoid follicle containing interspersed eosinophils, histiocytes, and foreign body giant cell reaction. The organism wall is composed of an eosinophilic tegument. In other areas, calcareous bodies were present in the parenchyma. The inflammatory infiltrate is noted to be granulomatous in a few areas and this finding is most prominent in this section of the tissue where a portion of the organism is seen.

tive or disseminated form of disease and closer surveillance may be warranted.^{7,8} Serum enzyme-linked immunosorbent assay (ELISA) has been suggested as a potential surveillance tool that could be used pre- and postoperatively^{2,9}; however, this test is not validated in most U.S. laboratories.

Medical therapy with antiparasitic agents, such as praziquantel, is not typically recommended,¹⁰ and there have been reports of resistance when immature parasite forms are present.¹ However, some cases where surgical intervention was not pursued have demonstrated therapeutic success with higher doses of praziquantel.¹¹ Preventing recurrence thus depends on adequate surgical removal of the parasite. Although breast sparganosis is a rare diagnosis, in our increasingly globalized society we frequently encounter patients from other countries, and it is important to understand their epidemiologic risks for parasitic infections and to be able to appraise the limited available literature to determine best management using a multidisciplinary team approach.

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