









Mammary Sparganosis: An Isolated Case

Esparganosis mamaria: un caso aislado

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Abstract

Sparganosis is an unusual zoonosis provoked by the plerocercoid larva form of the tapeworm *Spirometra*. The most frequent sparganosis infestation is subcutaneous tissue and extra-subcutaneous areas such as the breast, representing more than 2% of cases. Consumption of raw food such as frogs, snakes, and fish are a risk factor for human infection. Complete surgical removal of the parasite is the therapy of preference. We illustrate the case of a patient with a nodular tumor in the left breast who required an anatomopathological study to differentiate between benign and malignant tumors. The histological study revealed the presence of percid tapeworm larvae (Sparganosis) without evidence of the same at the systemic-visceral level, treated with surgical extraction and with a favorable outcome.

Keywords: Sparganosis; Breast; Zoonoses; Breast Neoplasms; Peru (source: MeSH NLM).

Resumen

La esparganosis es una enfermedad zoonótica inusual provocada por la forma larvaria o pleroceroide de la tenia *Spirometra*. La infestación más frecuente por esparganosis es el tejido subcutáneo y las zonas extrasubcutáneas como la mama, representando más del 2% de los casos. La ingesta de alimentos poco cocidos o crudos tales como ranas, serpientes y peces es un factor de riesgo para la contraer dicho parásito. La extirpación quirúrgica completa del parásito es la terapia de preferencia. Ilustramos el caso de una paciente con tumor nodular en mama izquierda que requirió un estudio anatomopatológico para diferenciar entre tumores benignos y malignos. El estudio histológico reveló la presencia de larvas de tenia percolde (Esparganosis) sin evidencia a nivel sistémico-visceral, tratadas con extracción quirúrgica y con evolución favorable.

Palabras clave: Esparganosis; Mama; Zoonosis; Neoplasias de la Mama; Perú (fuente: DeCS BIREME).



INTRODUCTION

Sparganosis is a rare parasitic disease caused by infection with sparganum, the larval stage of *Spirometra* species. P. Manson first described the condition in China nearly two centuries ago, and C.W. Stiles documented the first human case in 1908. This parasite is most commonly found in fat areas of the abdominal wall, limbs, genitourinary, central nervous system, eyeballs, lungs, and pleura. Breast sparganosis is even rarer, accounting for no more than 2% of all reported human cases ^(1,2).

This infectious disease is found worldwide but is more commonly seen in some regions of the Asian (Japan, China, and Korea) and Latin American continents. Humans are accidental hosts in the parasite's cycle, while some domestic and wild animals bring the parasite inside their guts and become definitive hosts ⁽³⁾.

The incubation period ranges from at least 3 weeks to 3 years. Human infection occurs through the ingestion of raw or undercooked toads, snakes, poultry, fish, or water contaminated with infected copepods. The parasite matures in the skeletal muscles, subcutaneous fat layer, urinary tract, eye, pleural and abdominal viscera. In rarer cases, other tissues, such as the breast, may also be affected ⁽⁴⁾.

These tumors are inherently linked with marked inflammation and could resemble soft-tissue neoplasms. Ultrasound imaging has played a significant role in the detection and diagnosis of this disease. We illustrate a case of breast sparganosis infection in a 47-year-old woman, discussing its management and differential diagnosis. With only 10 reported cases of breast sparganosis worldwide, this case represents a rare occurrence.

This case highlights the importance of considering a wide range of differential diagnoses in women presenting with a breast mass, especially in individuals from endemic regions. This case followed CARE guidelines for reporting and obtaining patient informed consent and approval by an ethics committee of the National Institute of Neoplastic Diseases (Instituto Nacional de Enfermedades Neoplásicas- INEN)

CASE REPORT

A 47-year-old woman from Iquitos, Loreto, Peru, who worked as a food dis-

tribution control supervisor, had a long-standing habit of consuming river fish. She denied eating raw foods such as frogs and snakes, which were commonly consumed in her region. However, she recalled an incident in early 2019 when she traveled to a remote area in her locality and was exposed to an unsanitary environment. During that time, she had contact with non-potable river water and consumed undercooked meat.

In October 2022, she noticed the presence of a lump in the left breast associated with pain and subsequent localized erythema. Therefore, she arrived at the National Institute of Neoplastic Diseases. On physical examination, a painful palpable nodule was recognized in R10-R11 of the left breast, 3 cm from the nipple. Imaging studies breast ultrasound highlighted a 17 mm nodular lesion in the left breast, BIRADS 4A type, associated with suspicious lymphadenopathies (Figure 1). An ultrasound-guided true-cut biopsy was conducted, documenting breast tissue with fibrosis and scarce ducts without evidence of malignant neoplasia. Given the discordance between the imaging study and the ana-

tomopathological result, an excisional biopsy was scheduled, and additionally, a fine needle aspiration biopsy of the left axillary region was performed, the latest being negative for malignancy.

The anatomopathological report, following a lumpectomy (excisional biopsy), confirmed the presence of percooid parasitic larvae with histological features consistent with sparganosis. The findings were associated with an exacerbated chronic inflammatory process, including foreign body-like giant cells, with no evidence of malignant neoplasm (Figure 2). Subsequent imaging studies showed a high-density image compatible with seroma. Tomographic studies of the brain, thorax, and abdomen were also performed, all without substantial alterations.

Serial parasitological and immunoassays studies were negative during the infectious diseases department's evaluation. As a result, observation and monthly follow-up were recommended, with no antiparasitic treatment prescribed at that time. The patient was discharged, and the surgical wound is currently well-treated and without significant abnormalities. (Figure 3).

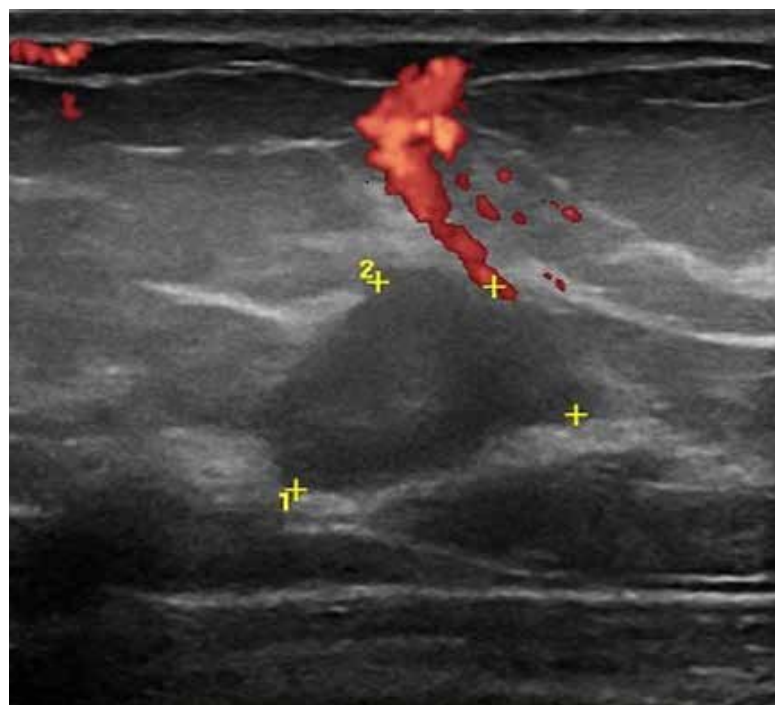


Figure 1. Ultrasound image showing suspicious lesion in left breast, pre-surgery.

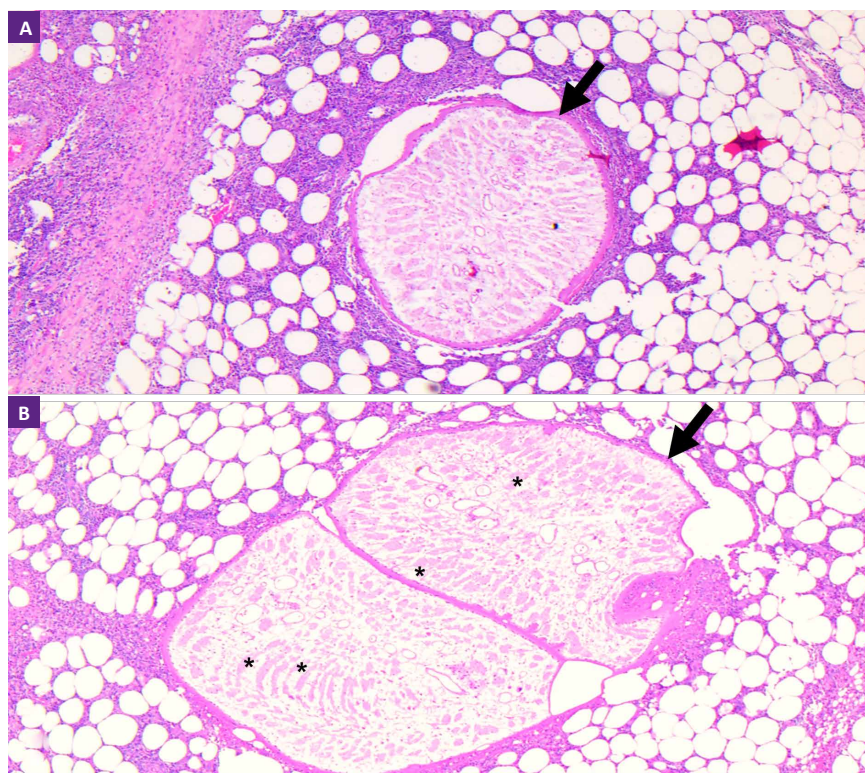


Figure 2. A. The cestode (arrow) shows a pale myxoid matrix with longitudinal smooth muscle fibers and calcareous corpuscles (mineralized concretions), surrounded by a non-cellular eosinophilic tegument. Surrounding tissue may show chronic inflammation, cyst formation, granulomatous inflammation (HE 200X). **B.** The thin longitudinal smooth muscle fibers (*) within the myxoid matrix of the cestode larval area are the distinguishing feature for identification of the organism. Scolex is absent, compared to other parasitosis (HE 400X).

DISCUSSION

Sparganosis is a zoonotic disease caused by the cestode larvae of *Spirometra* species, belonging to the *Diphylloboba-*

tridae family and *Pseudophyllidea* order. The larval stage, known as sparganum, gives the disease its name^(5,6). Several tapeworms of the *Spirometra* species, such

as *mansoni*, *erinacei*, *erinaceiueuropaei*, and *mansonoides*, can cause disease in mammals; however, the larval form of the cestodes *Spirometra mansoni* and *Spirometra erinacei* causes the most infestations in humans by far^(7,8). *S. erinaceiueuropaei* is typically found in Africa and Asia, while *S. mansonoides* is mainly reported in the United States. In Latin America, including Perú, the predominant species is *S. mansoni*⁽⁷⁾.

In Peru, human sparganosis has been reported in Loreto, where the patient under study comes from. The Peruvian Amazon rainforest is rich in fauna, which allows the parasite to sustain its life cycle. As a result, human sparganosis in this area may be more significant than currently recognized⁽⁹⁾. In the Peruvian Amazon region, this parasite is most frequently acquired through the ingestion of contaminated water or raw or undercooked meat contaminated with plerocercoid larvae, typically fish, snakes, or frogs. Once ingested, the larvae develop into a human infestation. These food sources are a regular part of the diet for people living in these area⁽¹⁰⁾.

The adult definitive form of this parasite grows in the gut of certain wild animals and domestic mammals (cats and dogs). The first intermediate hosts are copepods that thrive in freshwater, while the second intermediate hosts include amphibians (frogs, toads) and reptiles (snakes, turtles, crocodiles), which serve as reservoirs for the parasite. Humans are incidental hosts, becoming infected by consuming raw or undercooked meat containing plerocercoid larvae, such as toad or snake meat. Infection may also occur through traditional practices, such as using infected meat as a topical treatment for skin disorders or inflammatory eye diseases. However, the most common route of transmission is the ingestion of contaminated water containing copepods harboring the parasite. After the ingestion, the larvae perforate the intestinal wall, enter the peritoneal cavity, and invade subcutaneous fat tissue, skeletal muscle, and other organs. The adult definitive form of this parasite is a pale characteristic, flat, pseudosegmented helminth and ranges from 3 to 30 cm long. Plerocercoids cannot enter through intact skin but can ingress via superficial skin lesions, eyes, and mucosa-exposed^(5,7).



Figure 3. Partial mastectomy. Postoperative scar in affected breast.

Table 1. Clinical-pathological characteristics of reported cases of breast sparganosis.

Author	Year	Sex and age	Procedence	Characteristics	Treatment	Pathology
Ordoñez-Chinguel, <i>et al.</i>	2025	47-year-old woman.	Loreto, Iquitos Province, Peru	<ul style="list-style-type: none"> Painful nodule in R10-R11, left breast, 3 cm from the nipple Ultrasound: 17 mm nodular lesion, BIRADS 4A, suspicious lymphadenopathies 	<ul style="list-style-type: none"> Lumpectomy Antiparasitic therapy was not needed 	<ul style="list-style-type: none"> Plerocercoid parasitic larvae compatible with sparganosis Chronic inflammatory process with foreign body-like giant cells No malignant neoplasm
Zhang Y, Tian Y. ⁽²⁾	2021	58-year-old woman	Wuhan, Hubei Province, China	<ul style="list-style-type: none"> Ultrasound: Nodule in the right breast 5x1.7x0.7 cm. BIRADS category 4B Another nodule in 6 o'clock 1.8 cm x 1.2 cm x 0.4 cm BIRADS 4A Two hypoechoic masses 0.5 cm x 0.3 cm at 11-12 o'clock, and 0.3 cm x 0.2 cm. BIRADS 4A 	<ul style="list-style-type: none"> Vacuum-assisted breast biopsy system 	<ul style="list-style-type: none"> The most extensive lesion surrounding the worm revealed inflammatory cell infiltration and thick eosinophilic tegument Multiple tapeworms were 18 cm, 16 cm, and 11 cm long. The other masses were parasite eggs
Okino T. <i>et al.</i> ⁽¹³⁾	2021	92-year-old woman	Okayama Prefecture, the western part of Japan	<ul style="list-style-type: none"> A nodule in the left breast (11.4 x 8.6 x 9.5 mm) was surgically removed in 2017 One year later, a new mass (8.9 x 4.5 x 6.7 mm) appeared 	<ul style="list-style-type: none"> Lumpectomy 	<ul style="list-style-type: none"> Genomic DNA revealed Mitochondrial cytochrome and was sequenced: <i>Spirometra</i> Type I
Oh MY. <i>et al.</i> ⁽¹⁴⁾	2019	69-year-old woman 46-year-old woman	Seoul, Korea Seoul, Korea	<ul style="list-style-type: none"> Painless tumor of the left breast Mammography: benign calcifications Ultrasonography: A hypoechoic lesion in the subcutaneous layer Painless tumor (2 cm) in the right breast Ultrasonography: Anoechoic tubular mass 	<ul style="list-style-type: none"> Excisional biopsy Excisional biopsy 	<ul style="list-style-type: none"> Features of sparganum, tegumental brush border, longitudinal muscle fibers, and absence of gastrointestinal tract Sparganum was confirmed: thick eosinophilic tegument, calcareous bodies, and longitudinal trends of muscles
Kazemi A, Awosika O, Burgess C. ⁽¹⁵⁾	2018	77-year-old woman	Washington	<ul style="list-style-type: none"> Two firm nodules in the right breast 	<ul style="list-style-type: none"> Single-dose therapy with oral praziquantel 600mg 	<ul style="list-style-type: none"> Punch biopsy: thin-walled vascular channels, loose inflamed stroma, fibrosis, calcareous corpuscles. Elevated Immunoglobulin E (811 IU/mL)
Kim HS. <i>et al.</i> ⁽¹⁶⁾	2017	62-year-old woman	Seoul, Korea	<ul style="list-style-type: none"> A firm tumor in the Right breast Mammography: Tubular lesion (5,5 cm) Ultrasonography: multiple track-like tubular lesions 	<ul style="list-style-type: none"> Lumpectomy 	<ul style="list-style-type: none"> Distinctive attributes of a sparganum and foreign body reaction Anti-sparganum-specific IgG levels: 1.22 (calculated by ELISA)
Nathavitharana RR, <i>et al.</i> ⁽¹⁷⁾	2015	58-year-old woman	She was born in China, immigrated to the United States more than 30 years ago	<ul style="list-style-type: none"> Left breast mass: 2 x 0.5 cm Mammography: area of focal asymmetry 1.2 cm Ultrasound: Irregular, serpiginous, hypoechoic nodule 	<ul style="list-style-type: none"> Lumpectomy 	<ul style="list-style-type: none"> Inflammation, lymphoid follicle shape, histiocytes, eosinophils, and foreign body giant cell response
Min KW. <i>et al.</i> ⁽¹⁸⁾	2013	76-year-old man	Seoul, Korea	<ul style="list-style-type: none"> Tumor of the right breast, it appears 1 month ago Ultrasound: Tumor with serpentine tubular low echoic pattern 	<ul style="list-style-type: none"> Lumpectomy Albendazole/praziquantel for 3 days 	<ul style="list-style-type: none"> Dilated tubules, infiltration of lymphocytes, eosinophils, and plasma cells The worm demonstrated the typical attributes of sparganum
Park JH. <i>et al.</i> ⁽¹⁹⁾	2006	59-year-old woman	Seoul, Korea	<ul style="list-style-type: none"> Palpable masse in the left breast Mammography: continuous cord-like structures Ultrasonography: tubular mass, folded band-like tracts, tubule-in-tubule impression 	<ul style="list-style-type: none"> Lumpectomy 	<ul style="list-style-type: none"> <i>Spirometra sp.</i>, total length of 20.3 cm
Sim S. <i>et al.</i> ⁽²⁰⁾	2002	29-year-old women	Seoul, Korea	<ul style="list-style-type: none"> Palpable itching tumor on the right breast Ultrasound: serpiginous hypoechoic tubular mass, partial fat necrosis 	<ul style="list-style-type: none"> Lumpectomy 	<ul style="list-style-type: none"> Ivory, pale opaque ribbon-like worm (16.5 x 0.5 cm)

The most common presentation of sparganosis involves subcutaneous and extra subcutaneous disorders, which can emerge in different parts of the body, preferentially skeletal muscles, eyeball, tongue, breast, lungs and pleura, viscera of the abdomen, urogenital, pericardium and mediastinum, neck, spine, bone, and central nervous system ^(6,11). The manifestation of the infestation depends on the number and location of larvae within the human body and may remain asymptomatic in the early phases of infection. Tissue migration and perforation by the plerocercoids usually trigger a localized inflammatory response with perilesional erythema or oedema, as observed in our patient (Table 1) ⁽⁶⁾.

The typical manifestation of sparganosis consists of migrating subcutaneous nodules that periodically appear and disappear. These lesions are intensely pruritic, inflamed, and erythematous, often becoming tender and accompanied by painful edema. In some cases, systemic symptoms such as chills and fever may occur. Eosinophilia is a common characteristic in affected patients. Nevertheless, our patient did not present with eosinophilia, and imaging studies showed no evidence of visceral involvement ⁽⁶⁾.

The diagnosis of sparganosis is problematic because it is not pathognomonic based on signs and symptoms. The definitive diagnosis is performed by biopsy or excision of the parasite in the subcutaneous tissue; however, cerebral, soft tissue, breast, or visceral sparganosis presents a greater challenge, as it often requires surgical removal of the larva. Serological tests are the most utilized for diagnosis ⁽⁶⁾. The pathological examination revealed a chronic inflammatory process with a granulomatous response, the presence of mature B-lymphocytes, eosinophils, and significant collagen deposition surrounding the tapeworm. These findings are consistent with the pathological results observed in our patient ⁽¹²⁾.

Surgical removal of the larval form is the preferred treatment for subcutaneous, ocular, breast, oral cavity, and cerebral sparganosis. It is crucial to ensure complete extraction of the parasite, as any remaining scolex can lead to recurrence of the disease. Praziquantel is the preferred treatment in case of considerable fat nodules or visceral sparganosis; the recommended dose is 120-130 mg/kg separated into two or three doses ⁽¹²⁾. In cases of deep-seated infections, albendazole is considered as an alternative treatment ⁽¹¹⁾.

In conclusion, parasitic infection by sparganosis is rare in our setting, and even more uncommon when confined to the mammary parenchyma without involvement of other body sites. In subcutaneous tissue tumors, it is essential to consider a differential diagnosis that includes rare infections or granulomatous responses to uncommon pathogens, such as in this case. Recurrence depends on the parasite's location and incomplete removal during the initial treatment.

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