

Diagnostic Importance of Recognition of Calcareous Corpuscles and Larva Part in the Diagnosis of Sparganosis by Fine Needle Aspiration Cytology: Reports of Three Cases of Mammary Sparganosis

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Sparganosis of the breast is an uncommon disease. Cytological granulomatous inflammation is a common feature of sparganosis. The presence of larval fragments is necessary for a definitive diagnosis of sparganosis in a cytological specimen. However, calcareous corpuscles entrapped in necrotic debris can be very diagnostic for sparganosis in endemic areas. We experienced three cases of mammary sparganosis diagnosed by fine needle aspiration cytology before surgical excision. Two cases showed numerous calcareous corpuscles associated with necrotic debris thought to be degenerated parasitic bodies. The remaining case revealed a few degenerative calcareous corpuscles and parasitic bodies. The presence of calcareous corpuscles and/or degenerative parasitic bodies can be an aid in the differential diagnosis between sparganosis and other forms of granulomatous mastitis, particularly in endemic areas of sparganosis.

Key Words: Sparganosis; Breast; Calcareous corpuscle; Cytology

Sparganosis is a zoonotic disease reported worldwide, but is more common in Asian countries, particularly Korea, China, Japan, Vietnam, and Thailand.¹ Humans are infected by eating contaminated water or raw or improperly cooked flesh of snakes or frogs infected with plerocercoid larvae (sparganum) of *Diphyllobotrium*, particularly *Spirometra mansonioides*. The definitive parasite hosts are cats and dogs. Another possible mechanism for human infection is applying a frog or snake to open wounds, which is a folk remedy in Asian countries.² Once ingested, the larvae penetrate the intestinal wall and migrate to tissues where they grow to length of up to 14 inches. Larvae most often form a subcutaneous nodule in the chest, abdomen, inguinal area and extremities, but rarely locate in the conjunctiva, central nervous system, or peritoneum.³⁻⁵ Local tissue reactions around the parasite results in an itchy, inflamed, and painful lump. Fat necrosis and granulomatous inflammation are common histological presentations of sparganosis. A diagnostic approach using fine needle aspiration cytology (FNAC) can be effective, because the parasite often forms a mass lesion superficially. However, few reports have described the cytological features of sparganosis. We report cytomorphological findings of three cases of sparga-

nosis of the breast, which were obtained by preoperative FNAC and surgical excision, to highlight the role of FNAC in the diagnosis of sparganosis.

CASE REPORTS

Case 1

A 50-year-old female was admitted to the hospital because of a right breast mass in the upper outer quadrant for 2 months. Ultrasonography showed a 2 cm-sized mass with a focal hypoechoic area in the subcutaneous fat layer, which was suspicious of fat necrosis. FNAC was performed on the right breast mass. At first, the smears were interpreted as atypical cells with necrosis (Fig. 1A). So, an excisional biopsy was performed. The cut surface of the breast tissue showed ill defined fat necrosis. Several fragments of a white worm measuring several centimeters in length and 1 mm in width were discovered. A microscopic examination showed a branching tunnel-like structure with massive infiltration of inflammatory cells, lymphocytes,

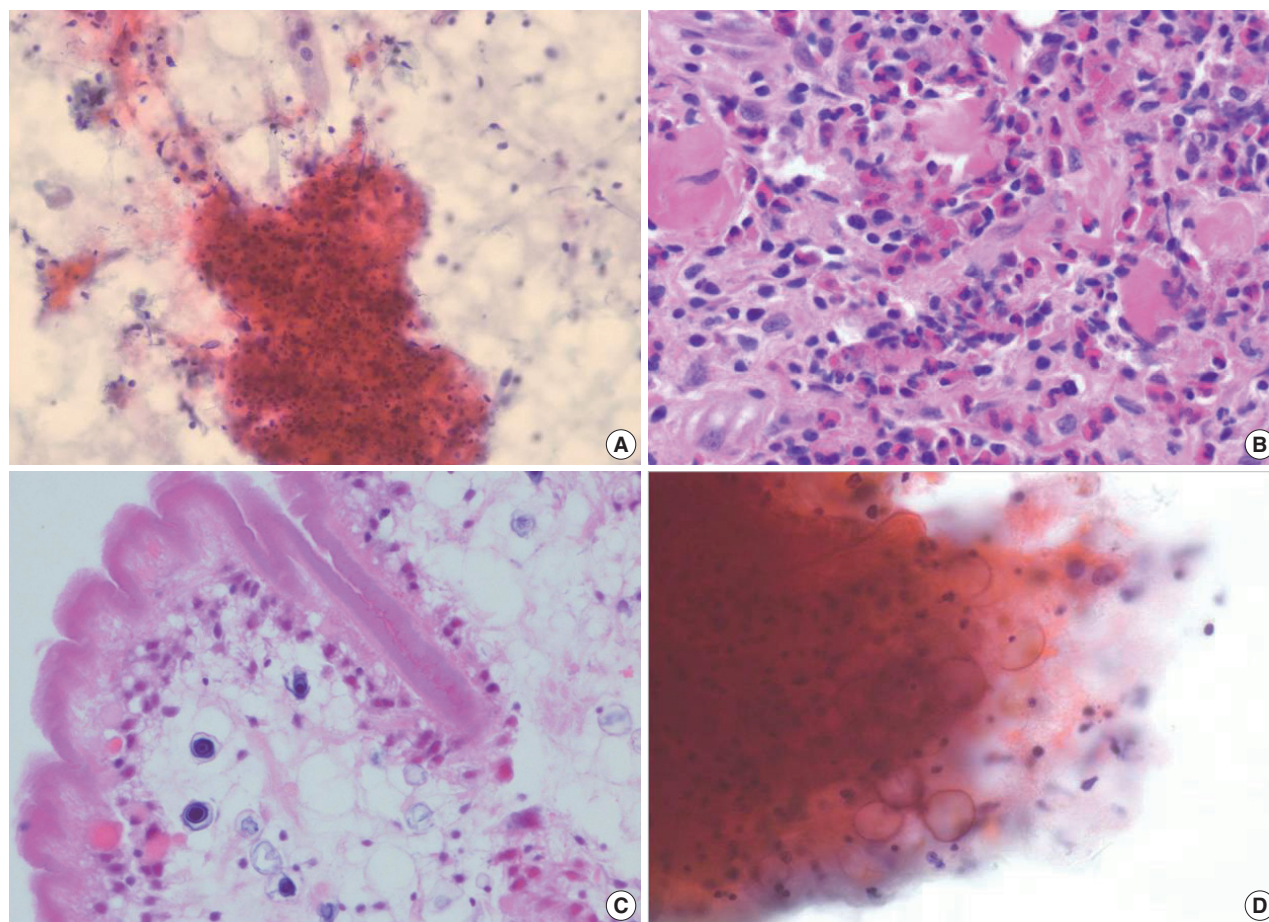


Fig. 1. Case 1. (A) Initial fine needle aspiration cytology is interpreted as atypical cells with necrosis. However, it appeared to be larvae on retrospective review. (B) Note the massive infiltration of inflammatory cells, particularly eosinophils in the resected specimen. (C) Sparganosis reveals tegumental cells and calcareous corpuscles. (D) A few degenerative calcareous corpuscles are identified upon careful examination.

and eosinophils, in the surrounding fat tissue (Fig. 1B). After recognizing that the worm was causing sparganosis (Fig. 1C), we reviewed the FNAC slides retrospectively, and found fragments of eosinophilic bodies with small dark nuclei that were smaller than those of lymphocytes (Fig. 1A, D). They appeared to be tegumental cells of the sparganum (Fig. 1C). On careful examination, a few degenerative calcific structures suspicious of calcareous corpuscle were revealed (Fig. 1D).

Case 2

A 45-year-old female visited the hospital with a 3–4 month history of a palpable right upper outer quadrant breast mass. On physical examination, a 2 cm-sized, hard, consistent, irregularly shaped, moveable mass was identified. Ultrasonography of the right breast revealed an echogenic nodule in the 12 o'clock position, consistent with fat necrosis. FNAC was performed and

showed a few benign epithelial cell clusters and fat cells with an inflammatory background. No surgical treatment was applied. Five years later, the mass had moved to the lower inner quadrant of the right breast. On repeated ultrasonographic examination, a tubular-shaped, echoic lesion was suggestive of fat necrosis or sparganosis. She had enjoyed hiking and drinking untreated water many times. A FNAC was performed repeatedly. On light microscopic examination, numerous calcareous corpuscles with granulomatous inflammation were identified in a background of the larva (Fig. 2A). This was interpreted as a parasite infection, such as sparganosis and was surgically excised. Granulomatous inflammation with lymphocytic infiltration was the dominant feature on microscopic examination of the surgical specimen (Fig. 2B). In addition, part of worm was obtained (Fig. 2C). Degenerative calcareous corpuscles were recognized on review of the first FNAC slides 5 years previously (Fig. 2D).

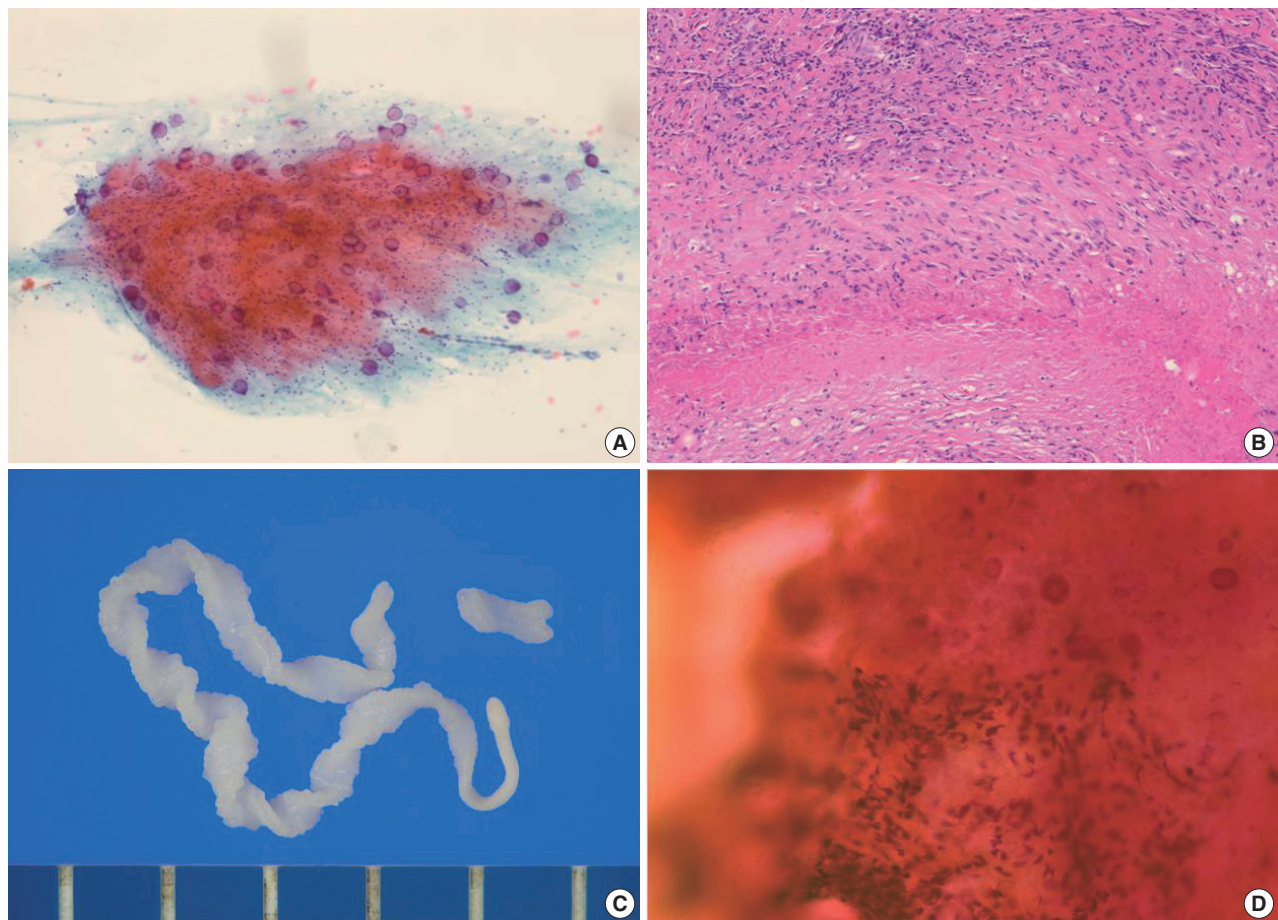


Fig. 2. Case 2. (A) Numerous calcareous corpuscles are present in the fine needle aspiration cytology (FNAC) specimen. (B) The excised breast tissue shows granulomatous inflammation. (C) The sparganum have a wrinkled, whitish, ribbon-shaped morphology. (D) Degenerative calcareous corpuscles within granulomatous inflammation are revealed upon retrospective review of the initial FNAC performed 5 years previously.

Case 3

A 47-year-old female presented with a palpable breast mass in the right upper outer quadrant for 3 months. She felt dull pain in the breast. A breast ultrasonography revealed a 3 cm-sized hypoechoic long tubular lesion in the right breast, so FNAC was performed. The aspiration slides demonstrated granulomatous inflammation with fat necrosis. Scattered calcareous corpuscles and fragments of amorphous eosinophilic material with small dark nucleus-like structures were characteristically present and appeared to be larvae (Fig. 3A, B). The range in the diameters of the calcareous corpuscles was 12.5-18 μ m. The FNAC was interpreted as parasitic infection consistent with sparganosis, so an excisional biopsy conducted. The surgically removed specimen included several pieces of irregularly shaped fat tissue and a 3 cm length of fragmented white worm. The branched tract was surrounded by granulation tissue with lym-

phoplasmacytic infiltration on microscopy, and granulomatous inflammation, epithelioid histiocytes, and multinucleated giant cells were present (Fig. 3C). A thick tegument with dark basophilic tegumental cells and calcareous corpuscles were identified upon microscopic examination of the worm (Fig. 3D). She had no history of ingesting snake or frog meat or drinking untreated water.

DISCUSSION

Parasitic infections of the breast are uncommon. Filariasis,⁶ cysticercosis,⁷ schistosomiasis,⁸ dirofilariasis,^{9,10} and sparganosis^{11,12} of the breast have been reported as causes of parasitic infection. The characteristic cytological features of a parasitic infection are granulomatous inflammation with various inflammatory cells, particularly eosinophils.¹³ However, granuloma-

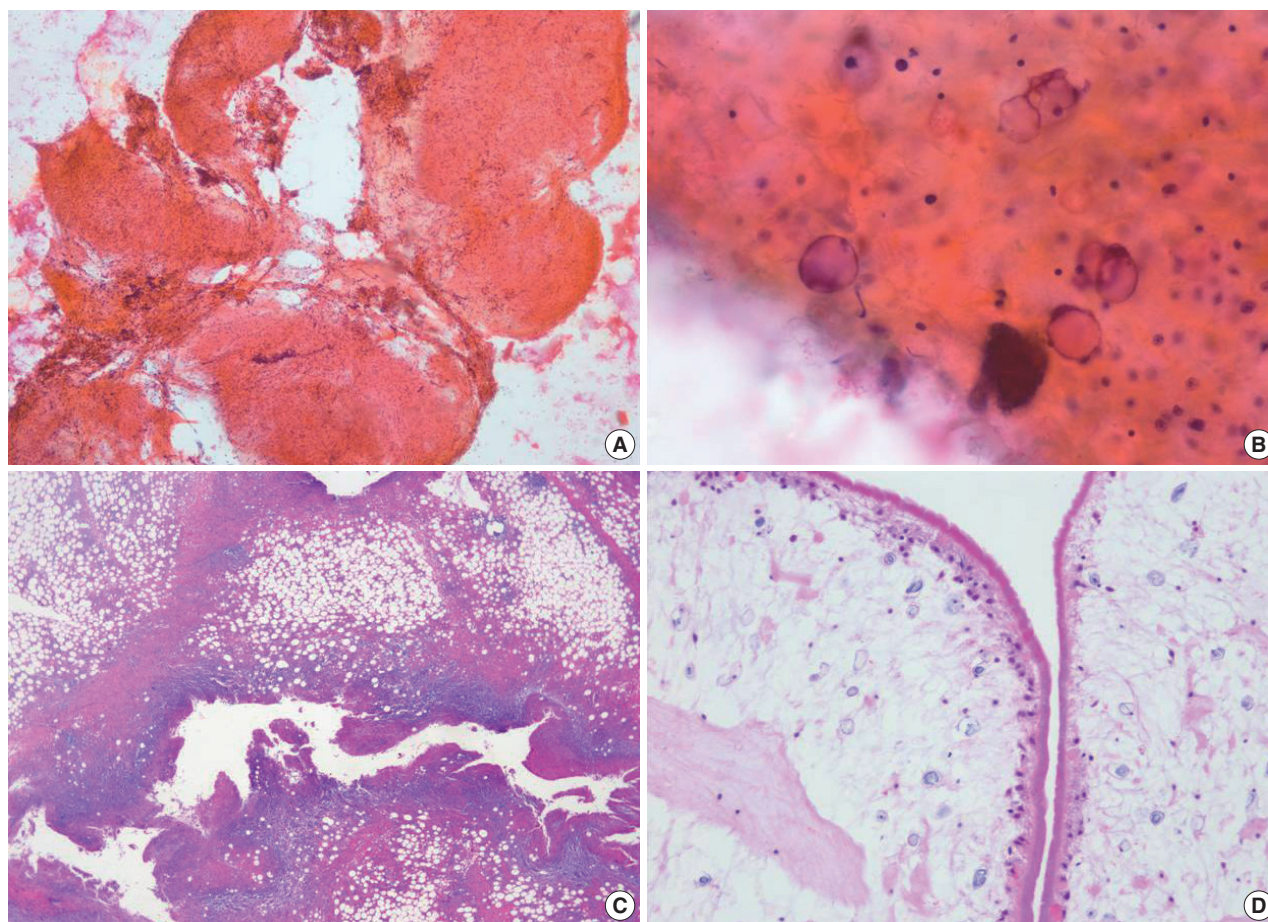


Fig. 3. Case 4. (A) Ill-defined segmented amorphous eosinophilic material is observed on fine needle aspiration, which seemed to be larva, however, it is identified in a necrotic background on low magnification. (B) A few scattered calcareous corpuscles are present within the larva. (C) The branched tract surrounded by lymphoplasmacytic infiltration is observed in the surgical specimen. (D) A thick tegument, tegumental cells and calcareous corpuscles are revealed on a histological section of the sparganum.

tous inflammation of the breast can be associated with tuberculosis,¹⁴ sarcoidosis,¹⁵ fungal infection,^{11,14} implantation of foreign material, idiopathic inflammation or breast carcinoma. The common finding of all previously reported cases of sparganosis is granulomatous inflammation.^{5,7,11,12,16,17} But, the calcareous corpuscles or larval section was not identified in all cases. Although identifying parasite fragments is most important for a diagnosis of sparganosis, it is difficult to use a worm as a cytology specimen. Actually, we missed the degenerated parasite in the FNAC specimen during the initial examination of cases 1 and 2. We confused the peculiar pattern of crowded tegumental cells with aggregated lymphocytes and failed to notice the scattered calcareous corpuscles. Calcareous corpuscles are present as either intact or a small number of degenerative features. The former is easy to recognize but the latter is not. But, these calcareous corpuscles can be found during a careful microscopic examination when cytopathologists suspect sparganosis.

We think that the calcareous corpuscles are relatively easy to recognize compared to other sparganosis findings. Cysticercosis also shows calcareous corpuscles, but the presence of suckers and hooklets is a unique finding of cysticercosis.^{7,13,16} A history of association with hogs, the characteristic cystic structure, and a serological test can help make the diagnosis of cysticercosis.⁷ The presence of the calcareous corpuscle alone, without other morphological features of the parasite, is an aid to the pathological diagnosis of sparganosis in an endemic area.¹⁷ Large larval fragments reveal an eosinophilic segmented structure with tegumental cells and calcareous corpuscles. However, the small larval fragments are often mistaken for inflammation. The tegumental cells have a small dark basophilic nucleus, which appears like lymphocytes, whereas tegumental cells have a smaller sized nucleus than those of lymphocytes and usually present in an eosinophilic larval background. Cytopathologists should attempt to find clues of a parasitic infection such as larval frag-

ments and calcareous corpuscles when they discover granulomatous inflammation in the breast, particularly in an endemic area.

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