





Apparent Sparganosis Presenting as a Palpable Neck Mass: A Case Report and Review of Literature

만져지는 경부 종괴로 나타난 현성 스파르가눔증: 증례 보고 및 문헌고찰

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Sparganosis is an uncommon human parasitic infection caused by plerocercoid cysts of the genus *Spirometra*. Sparganosis of the neck is a rare condition, thus making it difficult to diagnose. It is often initially misdiagnosed as a lymphadenopathy or a soft tissue tumor. Herein, we describe a rare case of apparent sparganosis presenting as a palpable mass in the left neck of a 53-year-old female patient. Imaging studies played a key role in the diagnosis. In this case report, we emphasize that sparganosis should be considered in the differential diagnosis of a palpable superficial mass. We also stress the importance of meticulous radiological review in the context of appropriate clinical suspicion.

Index terms Sparganosis; Neck; Breast; Ultrasonography; Computed Tomography, X-Ray

INTRODUCTION

Sparganosis is a human parasite infestation by a plerocercoid tapeworm larva (*Spirometra mansoni*) of the genus *Spirometra*. The sparganum is an organism that is wrinkled, whitish, ribbon-shaped, a few millimeters in width, and up to scores of centimeters in length (1). Sparganosis frequently occurs in people living in East and Southeast Asia due to consumption of raw frogs or snakes for traditional medical practice or drinking water contaminated with cyclops (2, 3). The majority of human infestations involve subcutaneous tissue or muscle of the chest wall, abdominal wall, or the brain. Sparganosis rarely occurs in the neck or the breast (3). Herein, we describe a rare case of ap-

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parent sparganosis in the neck presenting as a palpable mass in a 53-year-old female patient with CT and sonographic findings. We also conducted a relevant literature research regarding this disease.

CASE REPORT

This was purely an observational case study in a retrospective manner. Neither the patient's management nor outcome was altered. Therefore, no ethical approval was required for this case report. Written informed consent was obtained from the patient for publication of this case report and accompanying radiological images. However, the patient did not want us to use image materials regarding surgical excision and histopathological examination for this case report.

A 53-year-old female patient visited our hospital with a chief complaint of a palpable mass in the left neck. She said that the painless and palpable mass tended to migrate in the left lateral neck for one month. She also had a palpable mass in the left breast several years earlier which spontaneously disappeared without treatment. She had no prior medical history of this condition. The patient had frequently drunk mountain water when climbing mountains. She denied eating frogs or snakes. Physical examination revealed a soft and compressible mass with diameter of 3-cm in the left lower neck (level V). Laboratory test values were within normal reference ranges. Ultrasonography (US) was performed with a high-resolution linear array transducer. Neck US revealed a tortuous and serpiginous tubular hypoechoic lesion with ill-defined central hyperechoic linear tract that strongly suggested sparganosis (Fig. 1A). The lesion was surrounded by hyperechoic area, suggesting perilesional inflammatory change. On color Doppler US, there was no internal or perilesional vascularity. A contrast enhanced neck CT examination was also performed to evaluate the extent of the lesion and accompanying findings. CT images showed multilobulated, subcutaneous tubular lesions with central hypodensity and marginal enhancement in the left level V area well correlated with the US findings (Fig. 1B, C). Considering her statement of the preceding breast mass, mammography was also performed. On mammography, multiple linear, or tubular dense calcifications were scattered in the subcutaneous layer of the left breast which could be considered as previous tracts of the parasites (Fig. 1D). After surgical excision of the neck mass, a whitish long worm measuring 18 cm in length was found and it was confirmed as a sparganum. On pathological examination, the surrounding hyperechoic area was confirmed as granulomatous inflammation along the recent migrating tract of the sparganum.

DISCUSSION

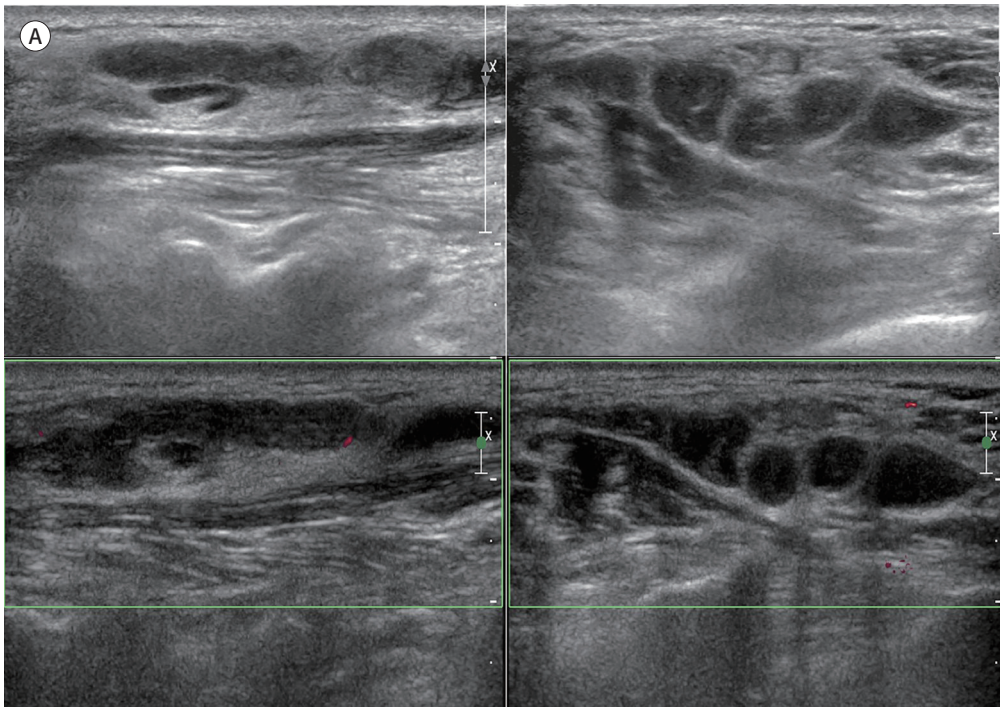
In this report, we describe the clinical and radiological findings of an apparent cervical sparganosis with probably previous migration tracts of the ipsilateral breast. Migrating nature of sparganosis has been reported in a previous study of cerebral infestation with CT findings (3).

Sparganosis is an infestation that usually occurs in subcutaneous tissues caused by larva of tapeworm in genus *Spirometra* (1). Sparganosis can involve any part of the human body in-

Fig. 1. A 53-year-old female patient with cervical sparganosis.

A. Grayscale longitudinal and transverse US images (upper row) show a well-defined, serpiginous tubular hypoechoic lesion with faint hyperechoic linear structures and surrounding indistinct hyperechoic areas in the subcutaneous layer of the left lower neck. Color Doppler US images (lower row) demonstrate no internal or peripheral vascularity.

US = ultrasonography



cluding the abdomen, chest, extremities, brain, bones, and the orbital region due to its ability to migrate. The most common location of sparganosis is the subcutaneous tissue (3). However, neck or breast involvement is a rare condition (3, 4). Although clinical manifestation of sparganosis is variable, the most frequent clinical feature is a painless or painful subcutaneous nodule.

The characteristic radiologic finding of sparganosis is a tubular appearance. This has been established in previous reports on mammography, US, CT, and MR imaging (4-6). Such a finding is produced by the empty tract and wall of a foreign body reaction (7). In this case, imaging findings were consistent with previous studies, including the following: 1) US findings demonstrated multiple, elongated, tubular, hypoechoic structures with or without internal inhomogeneous echogenicity; 2) CT findings showed tubular, central hypodense soft tissue mass with marginal enhancement; and 3) combined perilesional inflammation was observed in the surrounding fat tissue plane on both US and CT images. However, in the case of cervical sparganosis, such a tubular appearance on images should be differentiated from other mimics such as fistulous tract, abscess, or congenital cystic lesion in the neck. The fistulous tract usually has an external cutaneous opening and reveals a linear structure mostly filled with fluid on imaging studies. The abscess represents a peripherally enhancing fluid collection on CT and anechoic fluid collection with a thick wall, internal debris and posterior acoustic enhancement on US. The abscess also shows typical clinical and laboratory findings, such as

Fig. 1. A 53-year-old female patient with cervical sparganosis. **B, C.** Contrast-enhanced axial (**B**) and coronal (**C**) CT images show multilobulated, peripherally enhancing, and tubular hypodense lesions (arrows) with perilesional fat stranding in the left level V.



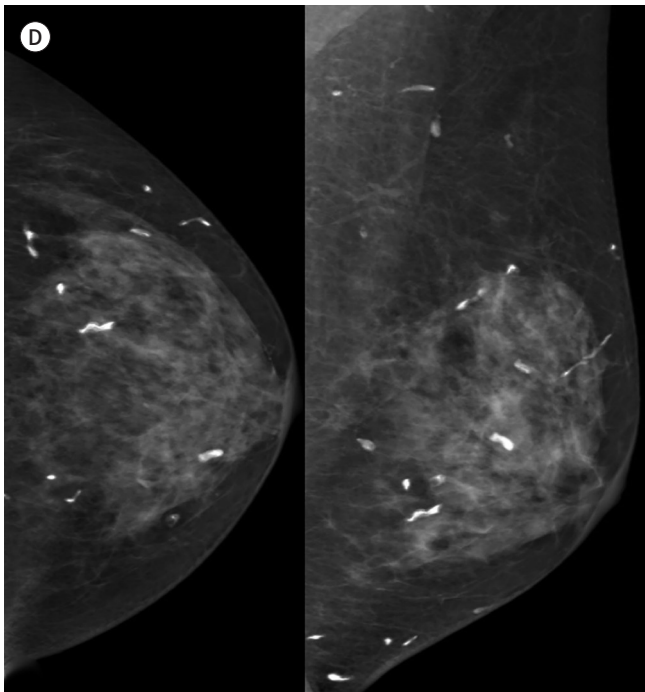


Fig. 1. A 53-year-old female patient with cervical sparganosis.

D. Craniocaudal (left) and mediolateral oblique (right) views on mammography show multiple tubular or linear calcifications, which could be the previous migration tracts of the sparganum, scattered in the subcutaneous layer of the left breast.

leukocytosis or elevated inflammatory biomarkers. The congenital cystic lesion, such as branchial cleft cyst or lymphangioma, shows a well-defined and thin-walled mass-like lesion filled with homogeneous fluid, and it usually has a typical location. However, in the case of combined complication such as infection or hemorrhage, the cyst wall is irregular, comprised of internal debris, is incompressible, and mimics a solid lesion (8).

Additionally, we presented uncommon features of linear or tubular calcifications in the ipsilateral breast. Although there has been no study of tubular calcifications in the breast sparganosis, previous studies of the sparganosis in legs have suggested that the calcification represent the sequential pathological changes after which a larva has passed (9, 10). If the larva migrates and dies, the tunnels with inflammatory foci are replaced by granulation tissue and calcium are deposited on them. Thus, multiple tubular calcification observed on plain radiographs can be considered the migration pathway along which the sparganum passed (9, 10). The findings of this case were similar to the previous case reports, and we thought that these calcified tracts of the breast seem to have been formed at least years ago, supporting previous passing tracts or the advanced age of the worm in our case.

In conclusion, we provide a case of apparent cervical sparganosis presenting as a palpable mass with clinical and radiological findings. Although imaging features of sparganosis are well-known from the literature, cervical sparganosis is often difficult to differentiate from other disease entities such as lymphadenopathy, cystic lesions, or soft tissue tumors because of its rarity and nonspecific clinical features. Through this case report, we emphasize that the migrating nature and tubular appearance can provide significant clues for diagnosing sparganosis. Therefore, clinical suspicion and awareness are critically important for accurate diagnosis and management in patients with this disease. Appropriate imaging work-up is also valuable in evaluating this systemic disease which has the possibility of concurrent or re-

mote infestation in clinical practice.

Author Contributions

Conceptualization, B.H.J.; data curation, H.M., L.S.M.; investigation, H.M., L.S.M.; project administration, B.H.J.; resources, B.H.J.; supervision, B.H.J.; visualization, H.M., L.S.M.; writing—original draft, H.M., B.H.J.; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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만져지는 경부 종괴로 나타난 현성 스파르가눔증: 증례 보고 및 문헌고찰

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스파르가눔증은 스피로메트라 아속의 유충 감염에 의해 발생하는 드문 기생충 질환이다. 경부 스파르가눔증은 드문 질환이기 때문에 림프절 병증이나 연부 조직 종양으로 오인되어 진단이 어려울 수 있다. 저자들은 영상 소견이 진단에 중요한 역할을 하였던 53세 여성 환자의 현성 경부 스파르가눔증의 증례를 보고한다. 이 증례 보고를 통해 저자들은 만져지는 경부 종괴물이 있을 때 스파르가눔증을 감별진단으로 고려해야 하며, 정확한 진단을 위해서는 스파르가눔증에 대한 임상적 의심 및 영상의학적 소견에 대한 이해가 중요함을 강조하고자 한다.

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