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【 CASE REPORT 】

Pulmonary Sparganosis: Tunnel Sign and Migrating Sign on Computed Tomography

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Abstract:
A 77-year-old woman presented at our hospital to undergo a close examination of an abnormal shadow which was observed on a chest radiograph. Contrast-enhanced computed tomography (CT) images in the lung window revealed a tortuous tunnel structure (tunnel sign), which was suspected to be the migration path of a parasite. Furthermore, CT images in the mediastinal window showed a linear filling defect from the right inferior pulmonary vein to the venous ostium in the left atrium (migrating sign), which was suspected to be a migrating parasite in the pulmonary vein. Tunnel and migrating signs on chest CT images were helpful in diagnosing pulmonary sparganosis.

Key words: pulmonary sparganosis, tunnel sign, migrating sign, computed tomography

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Introduction

Sparganosis is an infection by the plerocercoid larvae of various tapeworms belonging to the *Spirometra* genus (1). The larvae enter the human body and may migrate to the subcutaneous tissue, muscles, eyes, or the central nervous system. The involvement of the lungs and pleura is very rare (2). Computed tomography (CT) findings of pulmonary sparganosis previously reported scattered patches or nodules in the lungs (3). To our knowledge, CT images of tunnel and migrating signs reflecting a parasite's migration path in a pulmonary parenchyma and vessel have not been previously reported. We herein report the presence of tunnel and migrating signs on chest CT images which were helpful in diagnosing pulmonary sparganosis.

Case Report

A 77-year-old woman presented at the division of respiratory medicine at our hospital to undergo a close examination after her chest radiograph revealed an abnormal shadow. No signs of respiratory disease were found, and her superficial lymph nodes were not palpable. On physical examination, the patient's body temperature was 36.3°C, her respiratory sounds were unremarkable, and no skin lesions were detected. She had a past medical history of hypertension and a family history of endometrial cancer. A hematological analysis revealed a white blood cell count of $5.71\times10^3/\mu L$ with 8.4% being eosinophils, and an erythrocyte sedimentation rate of 40 mm/h. Her chest radiograph showed right hilar lymphadenopathy and thickened linear opacification in the right mid-lung zone (Fig. 1). Contrast-enhanced CT images in the lung window revealed a tortuous tunnel structure (tunnel sign), which was suspected to be the migration path of a parasite, in the lateral basal segment of the right lower lobe (Fig. 2A-E). In the periphery of the lateral basal segment, subpleural nodules with surrounding ground-glass opacity were observed. Furthermore, CT images in the mediastinal window showed a linear filling defect from the right inferior pulmonary vein to the venous ostium in the left atrium (migrating sign), which was suspected to be a migrating parasite in the pulmonary vein (Fig. 2F-J). We measured the widths and lengths of six individual slices of the linear filling defect and added the values using a workstation (ZIO station 2, ZIO soft, Tokyo, Japan). The filling defect measured 5.5 cm in length and 2.1 mm in width (Fig. 3A, B), which was considered to reflect the size of a migrating parasite.

Under a suspicion of parasite migration, a multi-dot

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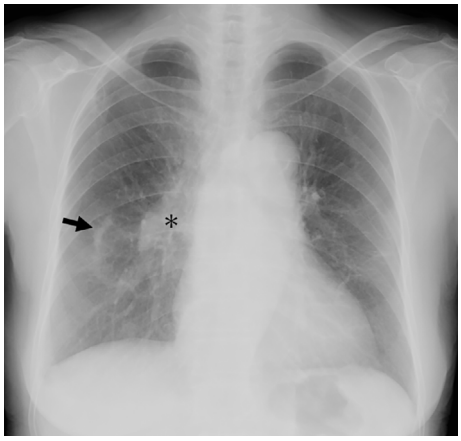

Figure 1. A chest radiograph shows right hilar lymphadenopathy (asterisk) and thickened linear opacification (arrow) in the right mid-lung zone.

enzyme-linked immunosorbent assay (ELISA) IgG was performed to detect any specific anti-parasite antibodies in the serum (Division of Parasitology, Faculty of Medicine, University of Miyazaki). The results showed that ELISA IgG was strongly positive for *Sparganum* parasites and negative for *Paragonimus westermani*, *Paragonimus miyazakiii*, *Ascaris lumbricoides*, *Dilofilaria parasites* and others. Thus, pulmonary sparganosis was diagnosed.

The patient's history was taken again, and it was noted that she lives with her husband, who often cuts chickens, which he raises in their backyard, on the same cutting board and with the same knife that the patient used for to cut vegetables and fruits using the same knife and cutting board, and thereafter they ate the vegetables and fruits raw. Hence, we hypothesized that she acquired the infection via plerocercoid larvae that were in the chicken, which in turn contaminated the food that was prepared using the unwashed cutting board and knife. Thus, she ingested the larva orally after consuming the raw food. Her husband had no history of any parasitic disease.

To treat the parasitic infection, the patient was given praziquantel, an anti-parasitic drug. Her serum eosinophil counts normalized, and a follow-up chest radiograph indicated a reduction in the linear opacification (Fig. 4) one month after the initial presentation. Contrast-enhanced CT performed five months later revealed that the tortuous tunnel structure and linear filling defect in the pulmonary vein had disappeared. There was no evidence of any recurrence three years after treatment.

Discussion

Sparganosis is parasitic infection that rarely occurs in humans and it is caused by the larvae of a tapeworm of the genus *Spirometra* (1). While adult spargana live in the intestines of dogs and cats, the eggs are released into fresh water and hatch into coracidia, which in turn are ingested by copepod crustaceans (1). The coracidia develop into procercoid larvae in the copepod, the primary intermediate host. Infected copepods are ingested by secondary intermediate hosts including birds, reptiles, and amphibians where the procercoid develops into a plerocercoid. The plerocercoids then develop into mature worms in definitive hosts, including dogs and cats. Humans get infected by drinking untreated water containing infected copepods or ingesting raw or inadequately cooked secondary intermediate hosts, e.g. the flesh of snakes or frogs infected with the plerocercoid larvae (1). The plerocercoid larvae may invade the intestinal wall, move under the peritoneum, and migrate into various tissues, including the subcutaneous tissue, muscles, and rarely, the central nervous system, eyes, lungs, or pleura (1). The plerocercoid, or *Sparganum*, is a motile worm of varying sizes (1-50 cm), typically a few centimeters in length and 1-2 mm in width (1). The infection is prevalent in east Asian and southeast Asian countries including China, Japan, Korea, and Taiwan; however, sparganosis occasionally also occurs in North America, South America, and Europe (4, 5). Although blood eosinophilia may facilitate the diagnosis, eosinophilia has been reported in only 30% of the patients with sparganosis (6). The gold standard method for diagnosing sparganosis is identifying a *Sparganum*, which is very challenging in visceral sparganosis (3). Furthermore, incomplete surgical removal may result in recurrent sparganosis (3). ELISA using plerocercoid crude or excretory-secretory antigens has high sensitivity for the detection of human sparganosis (3). To treat the infection, the intact parasite and surrounding granuloma are removed surgically; however, anti-parasitic drugs, such as praziquantel or mebendazole, may also be effective (3), as in the present case.

Pulmonary sparganosis involves the pulmonary parenchyma, pleura, or both (3). The respiratory signs and symptoms vary and are non-specific (3). The most commonly reported initial symptoms include coughing, fever, and chest pain (3). The infection is rarely discovered incidentally on chest radiographs with no prior symptoms (3), as it was in this case. The characteristics of a chest radiograph and CT images are as follows: 1) lesions commonly distributed in the lower part of the lung, appearing as scattered patches or nodules; 2) ground-glass opacity around the nodule or mass; 3) pleural effusion or hydropneumothorax, which has been observed in 75% of the patients (3). To our knowledge, tunnel signs reflecting a migration path of the parasite in the pulmonary parenchyma have not been previously reported in pulmonary sparganosis. The tunnel signs were reported to be characteristic MR findings of cerebral sparganosis on post-contrast MRI (7, 8). In the pulmonary parasitic infections, tunnel signs or track signs were reported to be characteristic CT findings of pulmonary paragonimiasis (9, 10). Furthermore, previous studies have not reported a migrating parasite in the vessels, which was identified as a migrating sign on contrast-enhanced CT.

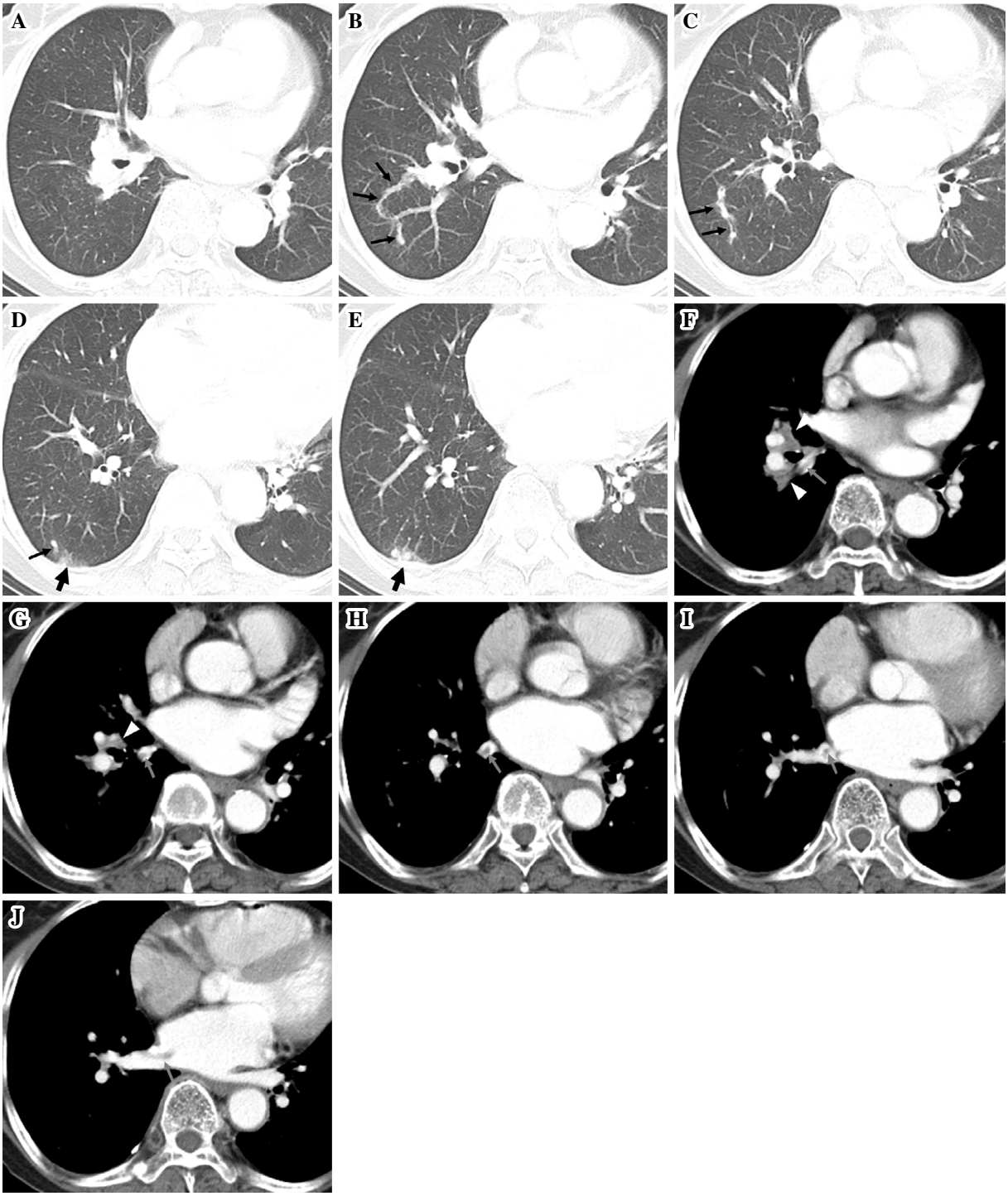

Figure 2. **A-E: Axial 5-mm contrast-enhanced computed tomography in the lung window shows a tortuous tunnel structure (arrows). Continually, subpleural nodules with surrounding ground-glass opacity were observed (D, E: thick arrows). The tunnel structure is suspected to be the parasite migration path (tunnel sign). F-J: (Fig. 2F at the same level as Fig. 2A) Axial 5-mm contrast-enhanced computed tomography in the mediastinal window shows a linear defect in right inferior pulmonary vein and venous ostium in the left atrium. Enlarged right hilar lymph nodes (arrowheads) are also observed. The linear defect is suspected to be a migrating parasite in the vessel (migrating sign).**

Conclusions

Previously, CT findings of pulmonary sparganosis was reported as scattered patches or nodules in the lung. To our knowledge, tunnel signs reflecting a migration path and a migrating sign reflecting a migrating parasite in the vessel have not been previously reported in pulmonary sparganosis. In the present case, these signs were helpful in the diagnosis

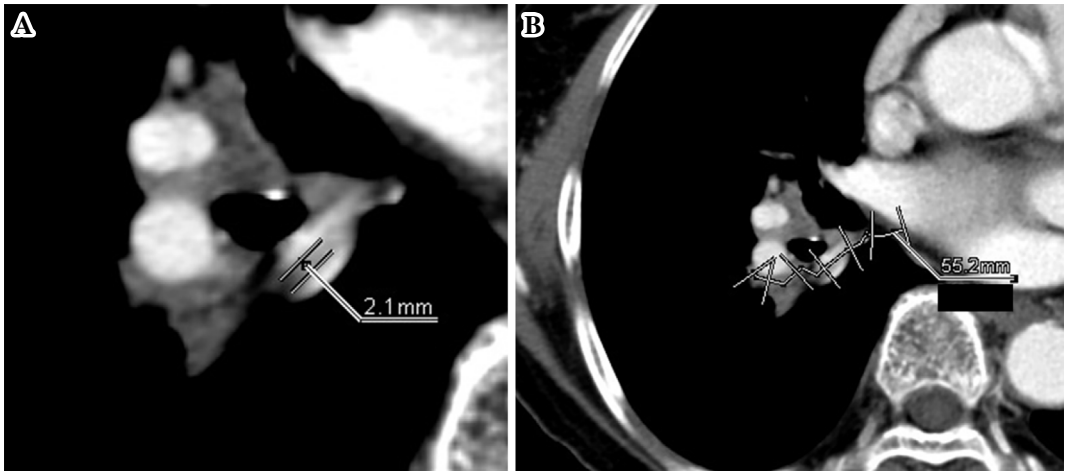

Figure 3. **A, B: Using a workstation (ZIO station 2, ZIO soft, Tokyo, Japan), the width of the linear filling defect was measured (A), and the length in six slices was measured and summed (B). The filling defect measured 5.5 cm in length and 2.1 mm in width, which is considered to reflect the size of migrating parasite.**

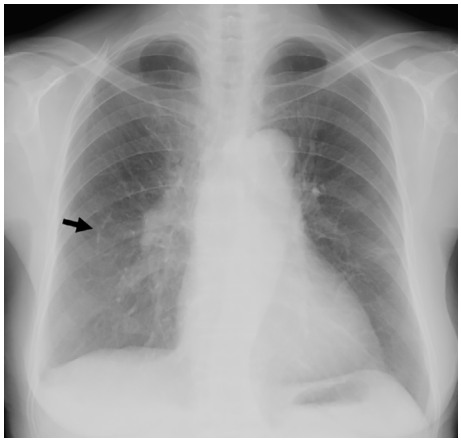

Figure 4. **A chest radiograph one month later shows a reduction of the linear opacification (arrow) in the right mid-lung zone.**

of pulmonary sparganosis. Additional studies that focus on these diagnostic techniques are needed to support the results presented in this report.

The authors state that they have no Conflict of Interest (COI).

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