# Pulmonary Sparganosis : A Case Report with Five Years Follow-Up

ANAKAPONG PHUNMANEE, M.D.*,
BENJAMARD INDHARAPOKA, M.D.*,
JURAIRAT KULARBKEAW, M.D.***
WATCHARA BOONSAWAT, M.D., Ph.D.*,
CHERDCHAI TUNTISIRIN, M.D.**,

**Abstract**

Sparganosis has a world wide distribution, but only a few patients have pulmonary involvement. The term sparganosis is defined as an infection by the larva of parasitic tapeworms of Spirometra species. We present here-in a patient, who was infected by this parasite and had pulmonary symptoms. The chest roentgenography revealed diffuse multiple nodular infiltration with cavitations. Bronchoscopy with a transbronchial lung biopsy was nondiagnostic. Finally, open lung biopsy was performed, and the histologic examination revealed plerocercoid larva of sparganum. The patient was treated with mebendazole 40 mg/kg/day for 6 months and his symptoms and pulmonary function improved. In the 5th year of follow-up, he presented with more progressive dyspnea and developed cor pulmonale, and finally died from pneumonia with sepsis.

The objective of this report was to present a rare manifestation of sparganosis and it's clinical course. Currently, there is no known effective treatment for this disease.

**Key word :** Pulmonary Sparganosis, Sparganum, Clinical Course

**PHUNMANEE A, BOONSAWAT W, INDHARAPOKA B, TUNTISIRIN C, KULARBKEAW J**
**J Med Assoc Thai 2001; 84: 130-135**

Human infection by Sparganum, or plerocercoid larva, the infective stage of Spirometra species is called Sparganosis(1). In Thailand, the first care was reported by Daengsvang and Tansurat in 1943(2). The predominant site of the infection is subcutaneous tissue. Pulmonary

* Division of Pulmonary and Critical Care Medicine, Department of Medicine,
** Division of Cardiothoracic Surgery, Department of Surgery,
*** Department of Pathology, Faculty of Medicine, Khon Kaen University, Khon Kaen 40002, Thailand.

involvement with sparganum is uncommon. From the literature in Thailand, there is only one case in which Sparganum involved intrathoracic organs (3).

## CASE REPORT

A 25-year-old Thai man from Udon-Thani presented with a cough and low grade fever for 3 months. He had sometimes noticed some bloody streaks in his sputum. He was treated at the district hospital, and the symptoms did not improve. Seven years previously, he was diagnosed with tuberculous spondylitis and treated with antituberculous drugs for one year. He usually ate raw cook beef and pork but had no history of eating raw flesh of frogs or snakes. He usually drank unboiled water from the lake near his home.

On physical examination, body temperature was 37°C, respiratory rate 20/min, pulse rate 80/min and blood pressure 110/70 mmHg. He had a post-operative scar of cleft lip and cleft palate. Chest auscultation revealed fine crepitation in both lower lung fields. Lymph nodes could not be palpated. Other parts were within normal limits.

Laboratory tests revealed hematocrit 35 per cent, white blood cell 11,200/$mm^3$, platelet normal. Differential count was polymorphonuclears 60 per cent, lymphocyte 23 per cent, monocyte 6 per cent and eosinophils 10 per cent. Urine examination and blood chemistry tests were normal. Chest roentgenography revealed multiple patchy infiltration with cavitation in both lungs. (Fig. 1A) Bronchoscopy was performed, and was normal. Transbronchial lung biopsy was non-diagnostic.

The patient was empirically treated for tuberculosis for two months, and the symptoms and chest roentgenography showed no improvement. The patient did not follow-up for 8 months. In that period, the patient took no medications and his symptoms and chest roentgenography revealed no improvement. (Fig. 1B) Finally, open lung biopsy was performed, and a histologic examination of the lung revealed larva of spirometra sp. (Fig. 2)

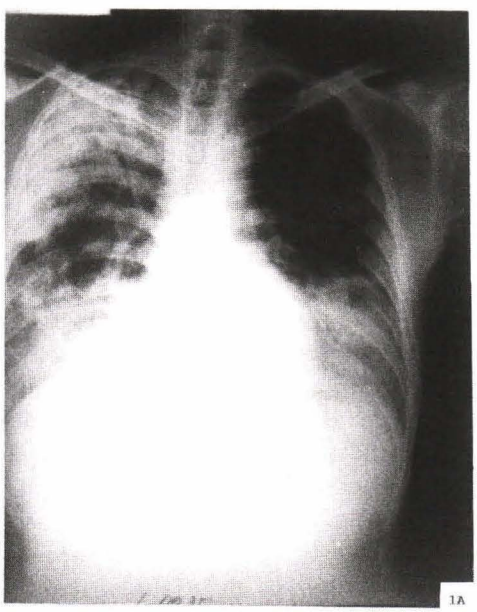


**Fig. 1A. Chest radiograph of a patient with pulmonary sparganosis demonstrating multiple patchy infiltrations which were initially greater in the right lung.**

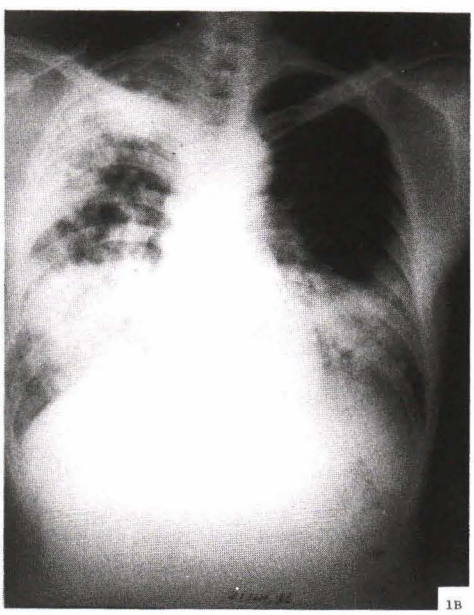


**Fig. 1B. Ten months later, after treatment with antituberculous drug for 2 months, the chest radiograph was still unchanged.**

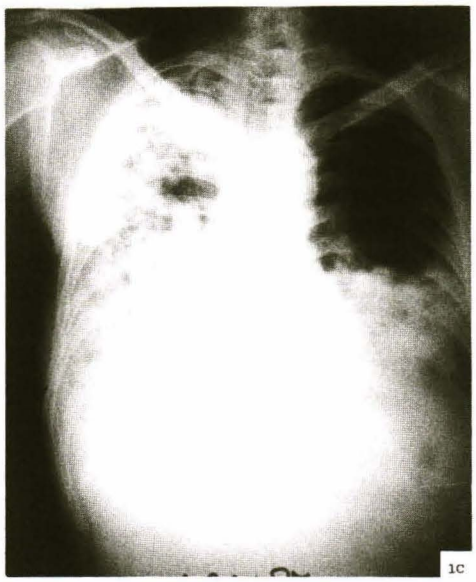

Fig. 1C. Eight months after treatment with mebendazole, despite clinical improvement, the chest radiograph revealed slightly increased infiltration in the right lung.

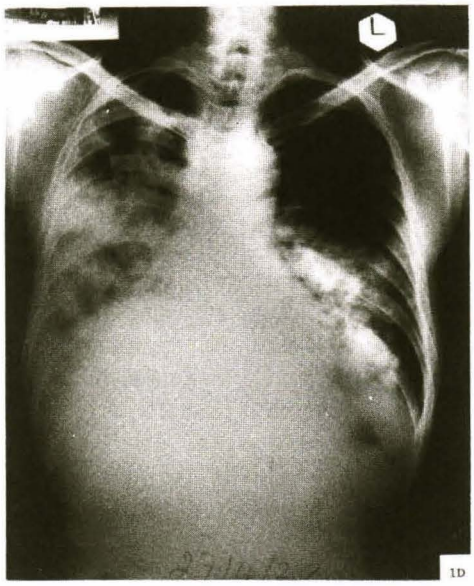

Fig. 1D. One year after treatment with mebendazole, the infiltration in the right lung decreased.

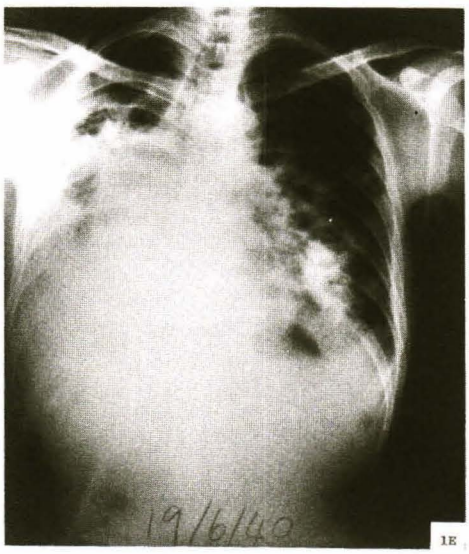

Fig. 1E. The final chest radiograph after the patient had developed severe *Klebsiella pneumoniae*.

The patient was diagnosed as having pulmonary sparganosis. The patient was treated with mebendazole 40 mg/kg/day. Six months later, the cough decreased. The patient felt better and he had gained 6.5 kg. Despite the improvement of his symptoms, the chest roentgenography showed more infiltration. One year after being treated with mebendazole, the chest roentgenography improved. (Fig. 1C, 1D)

Pulmonary function tests were performed before and after treatment, which revealed severe restrictive lung disease and small airway defects that slightly improved after the first 6 months of treatment with mebendazole. The spirometry after the 2nd year and 4th year of treatment are shown in Table 1. It is important to note that the severity of restrictive lung disease, when using FVC to compare, had slightly improved after treatment. During the period of the 5 year follow-up, the patient had only symptomatic treatment. After the 5th year of treatment, the patient noticed more shortness of breath and leg edema. Echocardiography was performed which revealed dilatation of the right atrium, right ventricle, pulmonary artery and moderately severe tricuspid regurgitation. The left ventricular ejection fraction was normal (0.73). These echocardiographic findings were compatible with cor pulmonale. Five years after diagnosis, the patient

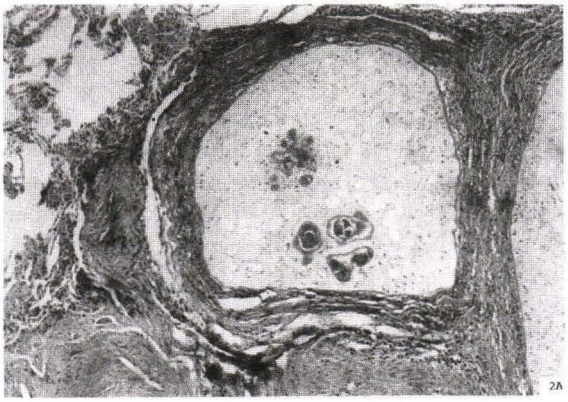

Fig. 2A. **Specimen of lung tissue demonstrating larva of sparganum surrounded by dense fibrous and inflammatory cell reactions. Morphologic features, including band of muscle, many spherical, light-to dark-staining calcareous corpuscles that have whorled appearance, indicate a diagnosis of sparganosis. (H&E x10)**

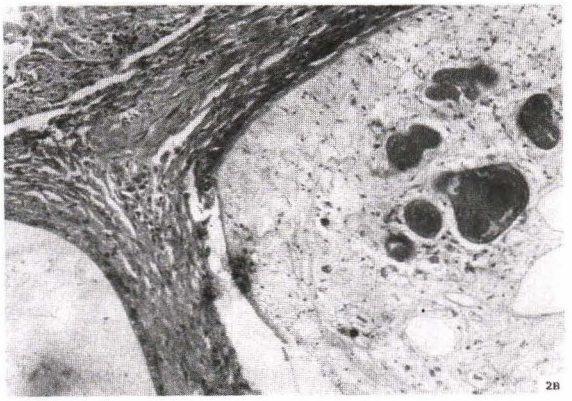

Fig. 1B. **Higher magnification, histologic section of sparganum showing inflammatory reaction. (H&E x 40).**

**Table 1. Spirometric values of the patient.**

| Parameters | Before treatment (% predicted) | 6 months after treatment (% predicted) | 2 years after treatment (% predicted) | 4 years after treatment (% predicted) |
|---|---|---|---|---|
| $FEV_1$ (L) | 1.12 (33.3) | 1.28 (38.1) | 1.06 (34.0) | 1.01 (30.0) |
| FVC (L) | 1.48 (37.8) | 1.65 (42.2) | 1.40 (37) | 1.35 (34.5) |
| $FEV_1$/FVC (%) | 75.5 | 77.5 | 76.0 | 75 |
| $FEF_{25-75\%}$ (L/sec) | 0.91 (20.6) | 0.97 (21.9) | 0.86 (19.5) | 0.79 (18) |

developed severe pneumonia with disseminated intravascular coagulation and expired due to multiple organ failure.

## DISCUSSION

Human sparganosis is caused by a larval tapeworm belonging to genus *Spirometra*[1]. The sparganum rarely involves the lung. From a previous report, the clinical presentations vary from asymptomatic, with accidental finding during an autopsy, a chronic cough and hemoptysis similar to our patient[4]. The course of this disease is chronic. The duration from the onset of the symptoms to the time of definite diagnosis is usually several years[4]. In the above case, it was about one year.

The chest roentgenography finding of this disease was multiple patchy infiltration and nodular lesions with or without cavitations[4]. The radiographic finding resembled pulmonary paragonimiasis, tuberculosis and metastatic cancer. This prolonged clinical course was unlike metastatic cancer, and the epidemiological data of our region showed that pulmonary tuberculosis was a more common disease than paragonimiasis. Therefore, most pulmonary sparganosis cases were initially treated with an antituberculous drug, before having a definite diagnosis by pathologic examination[4].

A definite diagnosis of pulmonary sparganosis always requires an open lung biopsy. Immunodiagnostic procedures of this disease are still in the developmental stage[8,9]. The definitive hosts of sparganum are cats, dogs and related carnivores[1]. The transmission of human sparganosis is as follows: (1) by drinking unboiled water from natural water beds contaminated with infected cyclops, the first intermediate host containing the procercoid larva. In this way, humans serve as the second intermediate host; (2) by ingestion of raw or partially cooked flesh of a second intermediate host; such as frogs, snakes, birds and mammals, which contain sparganum. In this way, humans serve as an accidental host; (3) by local application of the flesh of an infected frog or other second intermediate host to a wound, sore eyes or vagina. Whereby, the sparganum is transferred from the second intermediate host to human tissues and encysts as a sparganum. From the history of our case, we believe that the first and second mechanism were the causes of sparganum infection.

Sparganum can migrate to all parts of the body in the same manner as *Gnathostoma* larva. Sparganum produced eosinophilic chemotactic factor, so we were able to detect eosinophilia early from laboratory tests. Afterwards, the parasite encysted, so eosinophilia subsided[5]. Sparganosis can produce the protease enzyme, which is believed to play an important role in tissue migration and parasite feeding[6]. Sparganum metabolizes arachidonic acid to prostaglandin $E_2$ ($PGE_2$), which can suppress the functions of the mononuclear cell of the host. The releasing of $PGE_2$ may be related to the escape mechanism of sparganum from the host immune system[7].

The specific treatment of sparganosis is surgical excision, but in pulmonary sparganosis, since most of the lesions are bilateral, surgical excision is not possible[4]. Medical treatment, usually involves a 4-month course of mebendazole at a dose 40 mg/kg/day or praziquantel 40 mg/kg/day in six divided doses, repeated on three occasions over a period of two weeks, but the results are poor[10]. The sparganum leads to fibrotic and calcified lesions in the lung,[4,11] which result in restrictive lung disease. Restrictive lung disease rarely causes cor pulmonale, probably due to the short life span of a patient after the diagnosis compared with cor pulmonale that results from obstructive airway disease[12]. The clinical course of our patient lasted 6 years before developing right heart failure. We found no other causes of right heart failure and found a normal ejection fraction of left ventricle. In our patient, the spirometry test indicated severe restrictive and small airway defects that had slowly progressed after treatment with mebendazole. Therefore, we assume cor pulmonale developed because of the long standing restrictive lung disease post sparganum infection.

Effective treatment to prevent the complications of pulmonary sparganosis needs to be studied. Currently, the best treatment is to prevent the disease by educating people on improving their health sanitation.

(Received for publication on May 10, 1999)

## REFERENCES

1. Tansurat P. Sparganosis. In : Raul A. Marcial-Rojas, ed. Pathology of protozoal and helminthic disease with clinical correlation. Baltimore: Williums and Wilkins 1971: 585-91.
2. Daengsvang S, Tansurat P. Textbook of parasitology. Bangkok: Pakdiprandit Press, 1943.
3. Tesjareon S. Sparganosis in Thais. Siraj Hosp Gaz 1991; 43: 743-9.
4. Lin TP, Su IJ, Lu SC, et al. Pulmonary proliferative Sparganosis. J Formos Med Assoc 1978; 77: 467-72.
5. Horri Y, Owhashi M, Ishii A, et al. Leukocyte accumulation in sparganosis : demonstration of eosinophil and neutrophil chemotactic factor from the plerocercoid of spirometra erinacei *in vivo* and *vitro*. Am J Trop Med Hyg 1984; 33: 138-43.

6. Kong Y, Chung YB, Cho SY, Choi SH, Kang SY. Characterisation of three neutral protease of spirometra mansoni plerocercoid. Parasitology 1994; 108: 359-68.
7. Fukushima T, Isobe A, Hojo N, Shiwaku K, Yamane Y, Torri M. The metabolism of arachidomic acid to prostaglandin E2 in plerocercoids of spirometra erinacei. Parasitol Res 1993; 79: 634-8.
8. Nishiyama T, Ide T, Himes SR Jr, et al. Immunodiagnosis of human sparganosis mansoni by microchemiluminescence enzyme linked immunoadsorbent assay. Tran Royal Soc Trop Med Hyg 1994; 88: 663-5.
9. Oh SJ, Chi JG, Lee Se. Eosinophilic cystitis caused by vesicle sparganosis, a case report. J Urol 1993; 149: 581-3.
10. Torres Jr, Noya OO, Noya BA, et al. Treatment of proliferative sparganosis with mebendazole and praziquantel. Tran Roy Soc Trop Med Hyge 1981; 75: 846-7.
11. Aoshima-M, Nakara K, Matsuoka M, Kawabata M, Nakamura T. A case of proliferative sparganosis associated with pie syndrome and pulmonary embolism. Nippon Kyobu Shikkan Gakkai Zasshi 1989; 27: 1521-7.
12. Butter J, Agostoni PG. Cor pulmonale. In : Murray JF, Nadel JA, ed. Textbook of respiratory medicine. Philadelphia: W.B. Saunders, 1994: 1778-801.

# ปอดอักเสบจากพยาธิสปาร์กานัม : รายงานผู้ป่วยและติดตามการดำเนินโรค 5 ปี

*อนัฆพงษ์ พันธุ์มณี, พ.บ.*, วัชรา บุญสวัสดิ์, พ.บ., Ph.D.*, เบญจมาศ อินทรโภคา, พ.บ.*, เชิดชัย ตันติศิรินทร์, พ.บ.**, จุไรรัตน์ กุหลาบแก้ว, พ.บ.****

ปอดอักเสบจากพยาธิสปาร์กานัม สามารถพบได้ทั่วโลก แต่ผู้ป่วย Pulmonary sparganosis พบได้น้อย sparganosis ใช้เรียกการติดเชื้อจากตัวอ่อนของพยาธิในจีนัส Spirometra ในรายงานได้นำเสนอผู้ป่วยที่ติดเชื้อพยาธิสปาร์กานัม ที่มีอาการแสดงทางปอด ภาพรังสีทรวงอกพบมีลักษณะเป็นก้อนร่วมกับโพรงกระจายทั่วไปในปอดทั้งสองข้าง ซึ่งไม่สามารถให้การวินิจฉัยได้จากการส่องกล้องตัดเนื้อปอดผ่านทางหลอดลม ผู้ป่วยได้รับการวินิจฉัยโดยการผ่าตัดเนื้อปอดและส่งตรวจทางพยาธิวิทยาพบตัวอ่อนพยาธิระยะ plerocercoid ของพยาธิ sparganum ผู้ป่วยได้รับการรักษาด้วยยา mebendazole ขนาด 40 มก./กก./วัน เป็นเวลา 6 เดือนผู้ป่วยมีอาการและการตรวจสมรรถภาพปอดดีขึ้นเล็กน้อย ในช่วงแรก ในช่วงติดตามการรักษาในปีที่ 5 พบว่าผู้ป่วยเกิดภาวะ cor pulmonale สุดท้ายผู้ป่วยเสียชีวิตจากปอดบวมและติดเชื้อในกระแสเลือด ปัจจุบันยังไม่มีการรักษาปอดอักเสบจากพยาธิสปาร์กานัมที่ได้ผลดี

**คำสำคัญ** : ปอดอักเสบ, สปาร์กาโนสิส, การดำเนินโรค

**อนัฆพงษ์ พันธุ์มณี, วัชรา บุญสวัสดิ์, เบญจมาศ อินทรโภคา, เชิดชัย ตันติศิรินทร์, จุไรรัตน์ กุหลาบแก้ว**
**จดหมายเหตุทางแพทย์ ฯ 2544; 84: 130–135**

* สาขาวิชาโรคระบบทางเดินหายใจและเวชบำบัดวิกฤติ, ภาควิชาอายุรศาสตร์,
** สาขาวิชาโรคทรวงอกและหลอดเลือด, ภาควิชาศัลยศาสตร์,
*** ภาควิชาพยาธิวิทยา, คณะแพทยศาสตร์ มหาวิทยาลัยขอนแก่น, ขอนแก่น 40002