Clinical Features of Pulmonary Sparganosis

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Abstract: *Background:* Sparganosis is an infectious disease caused by the sparganum of *Spirometra* species, which seldom invades the respiratory system. The aim was to describe the clinical features and outcomes of pulmonary sparganosis. *Methods:* A total of 40 patients with pulmonary sparganosis were reviewed, including 12 cases known from this experience and 28 cases reported in the literature. *Results:* Among these 40 patients at an average age of 45.4 ± 11.1 years (men 29), 34 (85%) had a history of ingesting raw or undercooked meat (mainly frogs or snakes). The top 3 symptoms were coughing (60.0%), fever (57.5%) and chest pain (42.5%). Peripheral blood eosinophilia was found in 30 cases (75%). Lesions were located in lung parenchyma, airway, pleura and pulmonary vessels of the patients. Thirty-one patients (77.5%) had pleural effusion. The diagnosis was established by antisparganum antibody test in 30 cases (75%) and by pathology in 9 cases (22.5%); 1 case was not mentioned. Among the 35 cases with follow-up information, 2 treated with complete surgical removal and 31 with oral administration of praziquantel had no recurrence; the remaining 2 died without effective treatments. *Conclusions:* As an extremely rare and life-threatening parasitic zoonosis, pulmonary sparganosis should be diagnosed by combining the epidemiology, patient history, eosinophilia and the positive antisparganum antibody test result together if no worm was detected. Oral praziquantel is considered to be an effective treatment.

Key Indexing Terms: Pulmonary sparganosis; Sparganum; Plerocercoid; Pleural effusion; Eosinophilia. **[Am J Med Sci 2015;350(6):436–441.]**

"Sparganosis" is a parasitic infection caused by the plerocercoid larva (sparganum) of the genus *Spirometra*, most commonly *Spirometra erinaceieuropaei* (also called *Sparganum mansoni*).[1,2] Since the first case was detected in 1882, human sparganosis has been sporadically reported worldwide, mainly in Asian countries, but also occasionally from North America, South America, Europe, Australia and Africa.[1–5] Sparganosis poses a serious threat to human health; the larvae usually lodge in the subcutaneous tissues and muscles, but sometimes invade the brain, eyes, spinal cord and abdominal viscera, causing blindness, seizures, paralysis and even death.[1–3,5] However, pulmonary sparganosis is extremely rare.[5–7] Delayed diagnosis of pulmonary sparganosis can also pose a significant hazard to the patient.[8] So, the authors herein presented 12 cases of pulmonary sparganosis known from the experience and reviewed previous reports of such infection.

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METHODS

Subjects

Among 40 cases of pulmonary sparganosis, 12 were diagnosed and treated at Ruijin Hospital, Shanghai Jiao Tong University School of Medicine from January 1999 to December 2014, and 28 were obtained from the literature. The literature retrieval was conducted using the key word "sparganosis" in both the English and Chinese languages in PubMed, the China Academic Journals Full-text Database (CAJ), Google and Chinese Wangfang Database. Among more than 1,500 reported cases of sparganosis, only 23 articles concerned pulmonary sparganosis (32 cases) with full texts, including 11 in English,[5–15] 9 in Chinese,[16–24] 2 in Japanese[25,26] and 1 in Korean.[27] Four cases were excluded due to insufficient clinical information,[16,18,22,23] and the other 28 were finally identified (Table 1). The protocol was approved by the Institutional Review Board and Ethics Committee of Shanghai Ruijin Hospital.

Statistical Analyses

Continuous variables served as mean and standard deviation values (mean ± SD). The authors used independent-sample *t* test to compare the difference between pretreatment and posttreatment. Because all measurement values for all subjects were not available, the sample sizes for different results were somewhat different. All analyses were performed using SPSS (13.0; Chicago, IL), and $P < 0.05$ was considered as significant.

RESULTS

Clinical Presentation

The 40 cases aged between 45.4 ± 11.1 years (range: 24–67 years), including 29 men and 11 women, were from China (26 cases), Japan (8 cases), South Korea (5 cases) and Thailand (1 case). Behavioral risk factors were associated with pulmonary sparganosis: 34 cases (85%) had the history of eating raw or undercooked meat (mainly frogs or snakes), including 21 (52.5%) who had ingested them frequently. Six cases had direct contact with contaminated stream (Table 1). For these patients with pulmonary sparganosis, respiratory signs and symptoms were found to be variable and non-specific. The most common initial symptoms reported were coughing, fever and chest pain (Table 2). The patients mostly expectorated small amounts of white phlegm and 12 cases (30.0%) suffered from high fever. Chest radiographs showed the presence of lung nodules in 3 cases without any symptoms.[5,9,13]

Laboratory Tests

The mean white blood cell count was (8.81 ± 3.09) × 10^9/L in patients at the onset of pulmonary sparganosis, including 9 cases presented with high white blood cell count (10.0 × 10^9/L − 17.9 × 10^9/L). Peripheral blood eosinophilia was observed in 30 cases (75.0%), and the mean eosinophil count was (1.47 ± 1.51) × 10^9/L (range: 0.04 × 10^9/L − 7.78 × 10^9/L). Levels of erythrocyte sedimentation rate and C-reactive protein were elevated (35.4 ± 12.0) mm per one hour and (43.0 ± 71.1) mg/L, respectively. Total serum immunoglobulin

TABLE 1. Summary of all cases with pulmonary sparganosis

No.	Reference	Sex/age	Suspicious raw food intake	Location	Diagnosis	Treatment	Outcome
1	Lin et al[9]	F/28	N/A	Lung	Lung biopsy	Untreated	N/A
2	Aoshima et al[25]	M/48	N/A	Lung, pleura pulmonary vessel and pelvis	Mass biopsy, AD (serum)	Thiabendazole	2 months after operation, dead
3	Shuya et al[10]	M/47	Boar and chicken	Pleura	AD (serum, pleural effusion)	PZQ	6 months, nonrecurrence
4	Ishii et al[11]	M/62	Frog, crab, boar and contaminated water	Pleura	AD (serum, pleural effusion)	PZQ	2 months, nonrecurrence
5	Phunmanee et al[12]	M/37	N/A	Lung	Lung biopsy	Untreated	5 years, dead
6	Kamiya et al[26]	M/61	Frog, snake, pheasant and boar	Pleura	AD (serum, pleural effusion)	PZQ	Nonrecurremce
7	Kamiya et al[26]	M/46	Frog	Pleura	AD	PZQ	N/A
8	Kamiya et al[26]	F/43	Frog	Pleura	N/A	N/A	N/A
9	Kamiya et al[26]	F/43	Snake and chicken	Pleura	AD	PZQ	N/A
10	Iwatani et al[13]	M/57	Frog, snake and chicken	Lung	Surgery, AD (serum)	Surgical removal	1 year, nonrecurrence
11	Zheng et al[17]	M/39	Snake	Lung, pleura, abdominal ST and liver	AD (serum)	PZQ, mebendazole	3 months, nonrecurrence
12	Oh et al[27]	M/44	Frog and snake	Lung, ST	Surgery	Surgical removal	4 months, nonrecurrence
13	Xu et al[19]	M/33	Frog	Pleura, abdominal ST and parotid gland	AD (serum)	PZQ	1 year, nonrecurrence
14	Dai et al[20]	F/31	Snake and contaminated water	Pleura	Pleural biopsy	PZQ	6 months, nonrecurrence
15	Cheng et al[7]	F/32	Frog and bullfrog	Lung	AD (serum, BALF)	PZQ	5 months, nonrecurrence
16	Lee et al[14]	F/53	Frog	Pleura and pericardium	AD (serum)	PZQ	4 years, nonrecurrence
17	Bai et al[15]	M/67	Frog, snake, boar, shrimp, sheep, snails and contaminated water	Bronchus	Spargana found by bronchoscopy	PZQ	5 months, nonrecurrence
18	Chung et al[5] group A	M/57	Snake, boar and contaminated water	Lung and pleura	Lung biopsy, AD (serum)	PZQ	9 months, nonrecurrence
19	Chung et al[5] group B	M/61	Contaminated water	Lung	Lung biopsy, AD (serum)	N/A	N/A
20	Huang et al[8]	M/43	Snake and crab	Lung, pleura abdominal and inguinal ST	Mass biopsy, AD (serum)	PZQ	1 month, nonrecurrence
21	Wang et al[21]	M/53	Frog	Lung and pleura	AD (serum)	PZQ	10 months, nonrecurrence
22	Oh et al[6]	M/45	Frog and snake	Pleura	Spargana found by thoracic close drainage	PZQ	1 month, nonrecurrence
23–28	Zhao and Wang[24]	M3 and F3: average 30 years	Frog and snake	Lung including 5 cases with pleura	AD (serum) including one case with pleural biopsy	PZQ	Nonrecurrence
29	Present case 1	M/54	N/A	Lung and pleura	AD (serum, pleural effusion)	PZQ	41 months, nonrecurrence
30	Present case 2	M/42	Frog and crab	Lung and pleura	AD (serum)	PZQ	28 months, nonrecurrence

(*Continued*)