A Case of Spontaneous Pneumothorax Complicating Gnathostomiasis*

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Infections with larvae and immature adults of the nematode Gnathostoma spinigerum, acquired by eating raw fish, are of frequent occurrence in Eastern countries;¹ in Thailand recurrent migratory cutaneous swellings are usually pathognomonic of this infection. The parasites have been observed emerging from the skin,² and from natural openings such as the vagina and urethral orifice. Some have been picked out or excised from cutaneous nodules or abscesses,², ³ others have been found in the abdominal cavity³, ⁴ and vitreous chamber of the eye.⁵, ⁶ Although observations of transient pulmonary shadows associated with the condition and of the coughing up of the gnathostomes have been reported,², ³, ⁶ spontaneous pneumothorax as a complication of this disease has not hitherto been described.

M. S., a 33 year old Thai housewife was admitted to Siriraj Hospital on September 3, 1957, with approximately three weeks history of sharp pain and "rippling noise" in the left side of her chest.

She first attended the outpatient clinic on July 14, 1955 complaining of substernal oppression for one week. Physical examination and chest fluoroscopy at that time gave negative results. Subsequently she was seen by us and physicians elsewhere on several occasions with somewhat similar complaints. Chest roentgenogram taken on July 30, 1957, was negative.

About a month before admission she noticed a swelling over her left chest anteriorly accompanied by slight itching and some degree of lancinating pain. Three days later the swelling subsided and five firm discrete cutaneous nodules were noticed in the same area (Figure 1). She then began to feel severe sharp knife-like pain in the left side of her chest anteriorly associated with a peculiar rippling noise on changing position. She also developed productive cough of white tenacious sputum occurring especially when lying on her left side. She was otherwise afebrile and had no other constitutional symptoms. She had frequently eaten raw fermented fish.

On examination she was rather thin, and slightly pale with no apparent sign of distress. Over the left inframammary region four firm discrete cutaneous nodules of about pea size each were observed. They were not tender and the skin overlying them was of normal colour. The chest was clear to percussion and auscultation. All other systems were within normal limits.

The blood picture showed 10,400 white cells per cmm. with 35 per cent neutrophils, 45 per cent eosinophils, and 10 per cent lymphocytes. Stool and urine examinations were non-revealing. Smears of sputum were negative for tubercle bacilli, pathogenic organisms and parasites, but numerous eosinophils were repeatedly noticed; cultures revealed no pathogenic organism. An intradermal tuberculin test (10 T. U.) and several skin tests for gnathostomiasis were strongly positive.

Chest roentgenogram on August 31, 1957 disclosed a moderate degree of pneumothorax with a small amount of fluid in the left hemithorax. Pulmonary opacity was noticed over the left upper lung field (Figure 2A). During hospitalization she was afebrile. Only slight cough and a small amount of white tenacious sputum occurred when she lay on her left side. This gradually disappeared within a few days without specific treatment. Since the day of admission seven new swellings had appeared on the chest at varying intervals of hours or days, varying in size, location and symptoms (Figure 1).

Biopsy of one of the skin nodules showed a chronic granuloma infiltrated with eosinophils and a few giant cells. Serial blood pictures exhibited persistently high leukocytosis with 35-50 per cent eosinophils. Chest roentgenogram on September 4, 1957 revealed marked clearing of pulmonary shadow and some degree of re-expansion of the collapsed left lung (Figure 2B). Subsequent film on September 18, 1957 showed complete re-expansion of the left lung and the pulmonary shadow was no longer visible (Figure 2C). The patient was discharged on October 1, 1957 apparently in good health.

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Up to January 3, 1958 she had reported three times at the out-patient clinic, each time within two days of a new swelling at a new site, and without any trace of the previous one. All swellings after discharge appeared on the back below the level of the left scapula. Between January 3 and February 25, 1958 no new swelling had appeared.

Discussion

Whereas the lung is the natural habitat of *Paragonimus* (the lung fluke), it is to the nematode *Gnathostoma* but a pathway (and resting place) in its wandering.

Since Levinson7 described the first case of human gnathostomiasis in 1889, there have been only a few reports concerned directly with pulmonary lesions. Robert8 in 1922 mentioned three cases of gnathostomiasis with symptoms related to the respiratory system: the first case with frequent and violent paroxysmal coughs, the second with difficult respiration, and the last case with paroxysmal tenacious coughs. In this country Prommas and Daengsvang2 noted one patient out of their “Nine Cases” with frequent forced coughing, who subsequently expectorated a male *Gnathostoma spinigerum*. Tansurat,9 in his recent review of the subject, reported six cases in the category of pulmonary gnathostomiasis: all of them had coughed up worms and exhibited symptoms suggestive of pleuro-pulmonary involvement. In all cases previously reported there was no clearcut evidence of pulmonary invasion except for indefinite chest symptoms and the expectorated worms that might have come from the upper respiratory tract or even from the pharynx. However, Priyanonda, Pradatsundarasar, and Viranuvatti6 have added a well proved case of pulmonary gnathostomiasis to the literature.

Almost 10 years ago, in this department, an instance of spontaneous pneumothorax was observed in a patient suffering at the same time from typical recurrent migratory cutaneous swellings of gnathostomiasis.9 Un-
**Figure 2A**: August 31, 1957—Moderate degree of pneumothorax, left. Small amount of fluid at left costophrenic angle. Pulmonary shadows, left upper lung field. **Figure 2B**: September 4, 1957—Some re-expansion of left lung. Marked clearing of pulmonary shadows, left upper lung field. **Figure 2C**: September 18, 1957—Complete re-expansion of left lung. Pulmonary shadows in left upper lobe no longer visible.
fortunately there was no opportunity to investigate the case further as the patient was not hospitalized and was not followed. To the best of our knowledge, spontaneous pneumothorax occurring simultaneously with *Gnathostoma* infection has not so far been recorded in the literature.

In the case presented here there is little doubt about the diagnosis of gnathostomiasis, a disease characterized by recurrent migratory cutaneous swellings, eosinophilia and positive skin test.

Total disappearance of all swellings after a few days and the absence of any parasite exclude sparganosis. The calabar swellings of loiasis can be excluded both by geographical distribution and by the fact that they are usually much larger than gnathostomal swellings. *Wuchereria malayi*, the only filaria present in Thailand,10 does not produce subcutaneous swellings.

The transient pulmonary lesions with marked increase of eosinophils in both blood and sputum (Loeffler's Syndrome) in our case indicate local reaction to one or more parasites. Repeated negative results in the search of the sputum for other parasites and the simultaneous presence of subcutaneous swellings, hydropneumothorax and ipsilateral pulmonary lesions are strong evidence of the passage of one or more gnathostomes through the lung and visceral pleura. The sequence of events may have been as follows. The worm entered from the outside, first through the parietal pleura into the pleural cavity where it had freedom of movement and rapidly produced pleural effusion, then on through the visceral pleura into the lung, thus producing spontaneous pneumothorax (and the rippling noise). The somewhat long duration of the pulmonary shadows lends further support to this hypothesis concerning the route of passage of the worm.

The worms that fail to find an exit are supposed to die after a hitherto unknown life span. The longest duration of gnathostomiasis on record is 10 years. This unusually long duration may be explained by repeated infections. Up to February 25, 1958 our patient had had a remission of 56 days from the last occurrence of subcutaneous swellings. She was still free from chest complaint.

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REFERENCES


