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Histopathologic Study on Human Sparganosis*

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INTRODUCTION

Sparganosis is one of the well known endemic parasitic diseases in Korea. According to the review by Cho *et al.*(1975) at least 63 cases of sparganosis were reported in the Korean literature from 1917 to 1974. Among these 63 cases, more than half (34 cases) was encountered in the last ten years of their review period. The incidence of sparganosis has been evidently increased during past 15 years. Whether it is because of improved medical delivery system and health care or actual increase of cases one cannot be sure of it. Moreover as one takes count of many unreported cases of sparganosis, it would reach a considerable number.

In this connection it seems quite desirable that the Korean physicians should be well aware of various aspects of the human sparganosis, particularly of its spectrum of pathological changes in the host tissue, as this disease can endanger human life.

In 1978, we (Chi & Chi) conducted a pathological study on 258 human cysticercosis. During that reappraisal of past pathological specimens there were parasitic granulomas that were quite distinguished from cysticercosis and yet were previously diagnosed as cysticercosis. The worm morphology was different, and the tissue reaction was also distinguishable. Pathologists often confront with only one section slide for diagnosis without having any information on the gross appearance of the worm structure. And yet they have to make a best possible diagnosis.

This study was prompted by these difficult situations we encountered during the pathological examinations of parasitized tissues. Based on 16 previously unpublished cases of human sparganosis, we have studied morphological characteristics of the worms and the host tissue reactions.

MATERIALS AND METHODS

Among 16 cases 12 came from the file of Department of Pathology, College of Medicine, Seoul National University, and the remaining four cases were referred from outside Hospitals. All these 16 cases were confirmed to be sparganosis by worm morphology. In two additional cases (Case 17 and 18) *Sparganum* worm was not detected in the lesions although the histological features were quite characteristic of it. These two cases were, however, excluded from the statistics.

The pertinent informations, namely year of diagnosis, patient age, sex, worm location, signs and symptoms were briefly summarized in Table 1. In two cases out of 16 we received only the worms without accompanying host

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Table 1. Age, sex and pertinent clinical findings of human sparganosis

	Sex	Age	Worm location	Worm condition	Duration	Symptoms and signs	Accession number
Case 1	F	27	Rt. neck	section only	3mos.	mass	S71-743
Case 2	F	50	Thumb tip	section only	10yrs.	mass	S71-1670
Case 3	F	17	Epigastrium	section only	3yrs.	mass	S72-2476
Case 4	M	27	Rt. inguinal	section only	1month	lump	S72-3564
Case 5	F	45	Intermammary	section only	1month	lump	S73-1061
Case 6	F	59	Lt. breast	section only	unknown	lump	S74-689
Case 7	M	54	Rt. scrotum	section only	2yrs.	mass	S75-1811
Case 8	M	32	Trunk	viable worm	unknown	mass	S76-27
Case 9	F	17	Abd. wall	viable worm	3yrs.	mass	C77-2476
Case 10	M	48	Chest wall	viable worm	unknown	mass	S77-3687
Case 11	M	36	Epigastrium	viable worm	6mos.	mass	S78-3637
Case 12	M	40	Trunk	viable worm	unknown	mass	O78-1158
Case 13	M	43	Abd. wall	viable worm	3yrs.	mass	S79-35
Case 14	M	45	Abd. wall	viable worm	unknown	mass	O79-453
Case 15	F	30	Thigh fat	viable worm	unknown	mass	O79-490
Case 16	M	28	Eye lid	viable worm	3mos.	mass	C79-53
Case 17	M	21	Stomach wall	not identified	1week	acute abdomen	C79-130
Case 18	M	19	Tongue	not identified	1week	mass	S80-1000

tissue. In 7 cases microscopic section slides were the only available materials we had.

When the worms were only specimens submitted, they were first studied grossly. When they were viable they were observed further in saline solution. Unless they are fed to the definitive host for species identification they were fixed in 10% formalin. After fixation the worm was cut in various directions and studied microscopically with routine hematoxylin-eosin stain or with other special stains as needed. When the host tissue was sent together with the worms, the worms were partly expelled from the lesion, and the lesion with the worm in part was fixed and cut to make section slides.

RESULTS

Annual incidence of sparganosis

Since the year of 1971 we had at least one case of sparganosis per year. The cases before the year of 1976, were detected on the base of section slides. Unfortunately gross specimens of the worms in these cases were largely unavailable.

These 16 cases of sparganosis seen during a period of 9 years in one institute indicate how prevalent this disease would be in nationwide scale. This number of sparganosis takes approximately 10% of cysticercosis encountered in the same period at the same institute, and together they represented 0.08% of a total biopsy cases examined at the Department of Pathology, Seoul National University Hospital during the same period.

Age distribution of sparganosis was relatively even from the second decade to the sixth decade. There were 9 males and 7 females in this series.

Location of the worms

The common sites of involvement were the skeletal muscle and subcutaneous tissue. The location was abdominal wall, chest wall, neck

and eye lid *etc.* (Table 1). Although the worm was not found in the lesion, the case 17 is most probable case of sparganosis involving the stomach wall. This case was referred to us by Dr. Ke Yong Song. The patient was a young soldier who was operated for acute abdomen. Gastrectomy was performed under a diagnosis of peptic ulcer perforation. Gross examination showed no

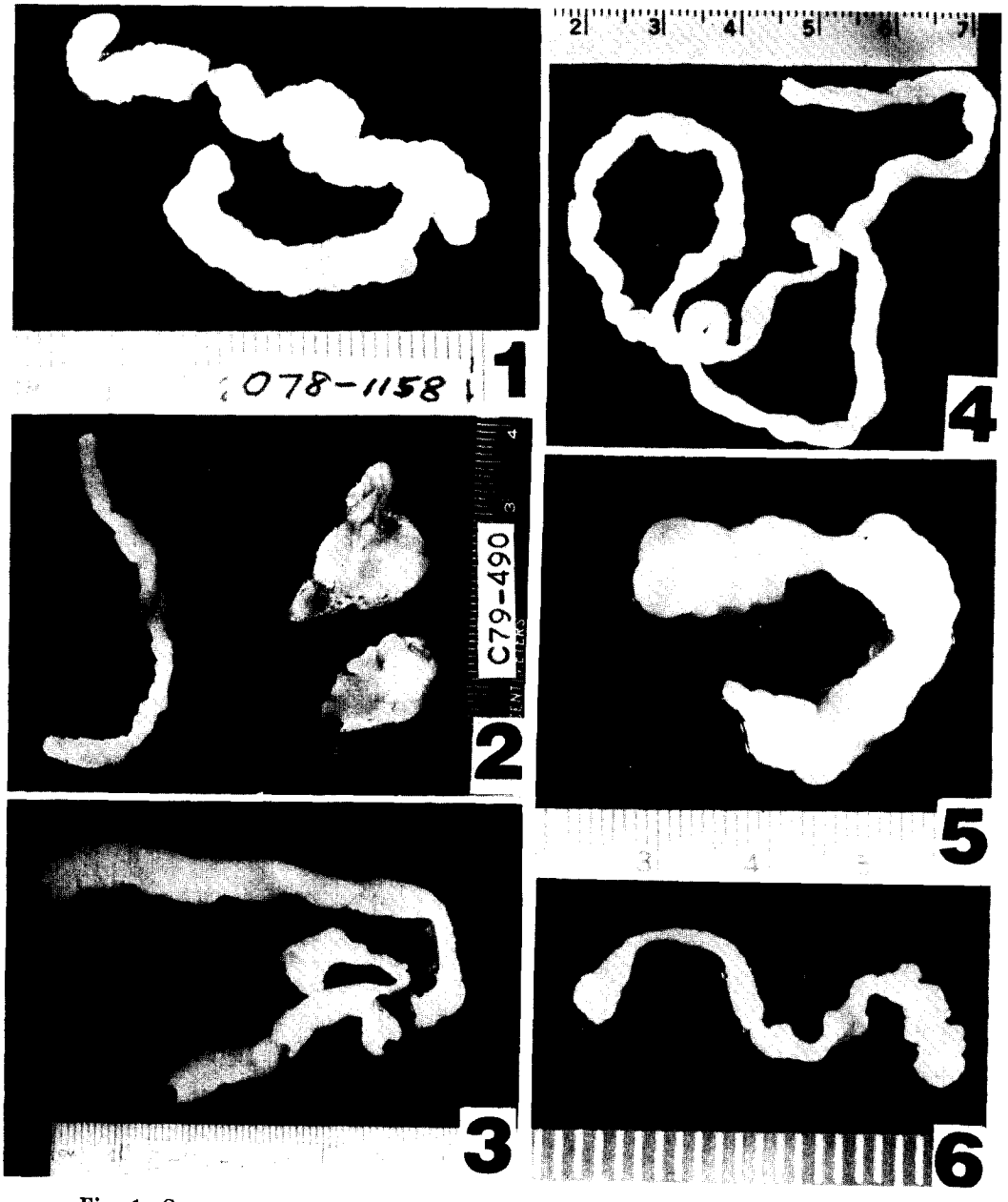

Fig. 1. Gross appearance of the worm *Sparganum* in Case 12.
Fig. 2. *Sparganum* worm and fat tissue around the worm in Case 15.
Fig. 3. *Sparganum* worm removed from chest wall in Case 16.
Fig. 4. *Sparganum* worm in Case 14.
Fig. 5. *Sparganum* worm in Case 10.
Fig. 6. Anterior portion of *Sparganum* in Case 11.

tumor but multiple tunnel-like lesions in the gastric wall. Microscopically multiple cavitating necrotizing granulomas were scattered through the entire thickness of the gastric wall. Characteristic palisading granulomas with pools of eosinophils were seen along the tracks that were thought to be made by the worm.

Gross morphology of the worm

The worm usually has a dorso-ventrally flattened ribbon-like configuration, showing pseudo-segmentation and pale milk white in color. When placed in warm saline solution the viable

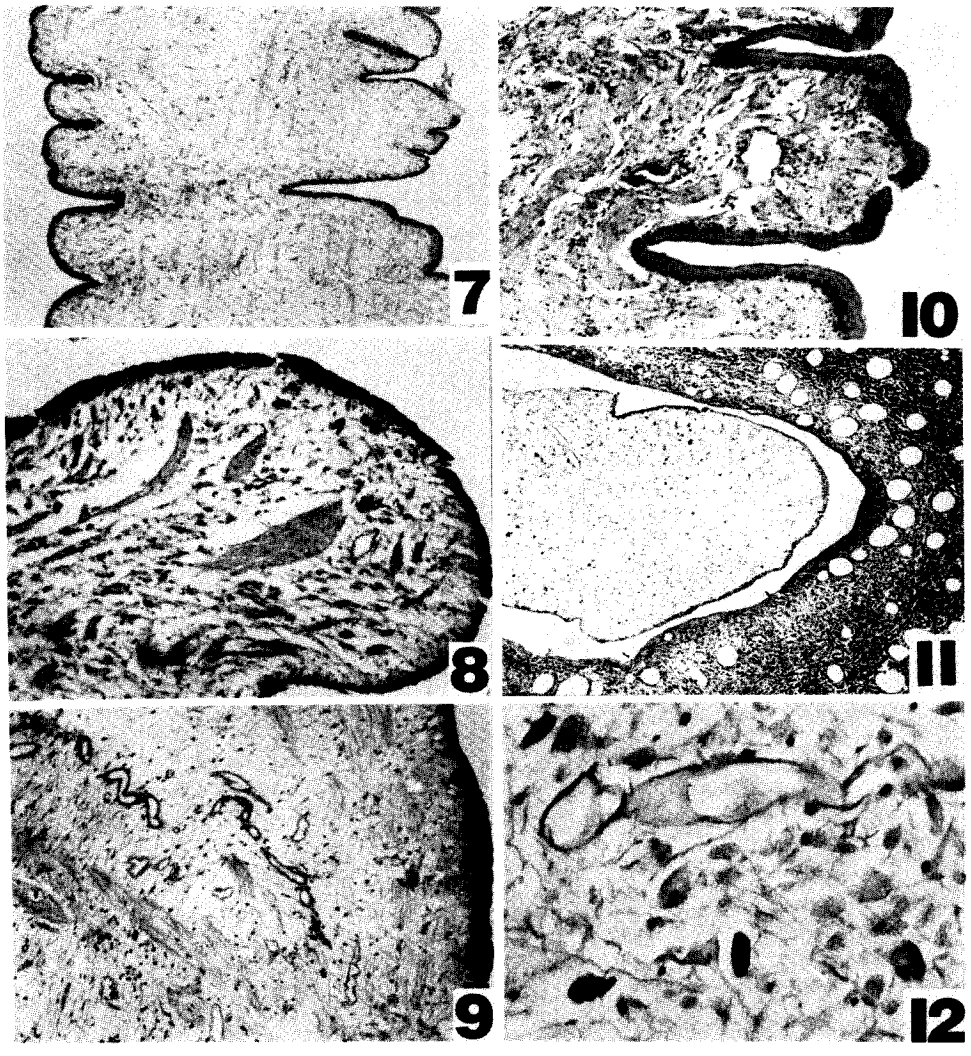

Fig. 7. Longitudinal section of the worm with characteristic muscle fibers.
Fig. 8. Oblique section of *Sparganum*, showing prominent muscle bundles.
Fig. 9. *Sparganum* worm showing excretory canals.
Fig. 10. Characteristic cuticular structure of *Sparganum*, with underlying stout muscle bundles.
Fig. 11. Cross-sectioned *Sparganum* and surrounding fat tissue with heavy inflammation.
Fig. 12. Higher magnification of the *Sparganum* worm, showing excretory system and calcospherules.

worms show active motion, typified by rapid extensile and retractile movements of the anterior portion of the organism. The anterior end of peristaltic waves connecting of the worm is thin and active when extended. Contractile segment proceeds down the length of the worm. The contractile segment becomes 3 to 4 times thicker than the original size of the worm. This strong contractility together with extremely thin anterior tip were very impressive. However, this motility was not in same degree in all cases that the worm were available for observation.

The worms often came in dead state or fragmented. When degenerated the worms became irregularly outlined, and sometimes it was very difficult to tell which is the anterior end of the worm. Degeneration here meant retrogressive changes of the worm that are not necessarily occurred inside the human body. Because it was not possible to assure that degenerative changes took place inside the human body or after it was expelled alive from the body. Accordingly definite degenerative change inside parasite body was only possible way to tell when the parasite was found inside the tissue. When fixed in formalin the worm showed contraction, and outer surface showed many transverse wrinklings. In close observation one wrinkle was further divided into finer wrinkles. This could be more clearly demonstrated under the dissection microscope.

The size of the worms varied greatly by different case. It seemed to depend on the sort of tissue where the worm was found and, probably on the viability of the worm. In this series the cases 10 showed the entire length of the worm to be approximately 2.5 cm in length and 0.2 cm in width. This worm showed evidences of degeneration microscopically and was partly absorbed inside the host tissue. The longest one (Case 14) measured 45 cm in length and 0.8 cm in width, and it came from abdominal wall and actively motile at the time of recovery.

Microscopic findings of the viable *Sparganum* worm

Sections of the worm showed dorsoventral flattening, thick, slightly wavy eosinophilic tegument overlaying a layer of radially oriented subparenchymal cells, well developed longitudinal bundles of smooth muscles consisted of dorsoventral and transverse fibers, arranged at right angles to each other. However, the major component was longitudinal direction. Longitudinal excretory canals were identifiable in the lateral regions of the worms parenchyme. Calcareous corpuscles (calcospherules) were almost always found in the parenchyme intermixed with muscle bundles running perpendicular to the surface of the parasite. Longitudinal sections show characteristic shallow invagination of the *Sparganum* integument as well as the longitudinal excretory canals and nerve cords.

Host tissue reaction around the worms

Pathologically host tissue reaction against *Sparganum* was fairly characteristic. The type of tissue reaction was directly related to the moving organism, and relatively localized around the parasite. No demonstrable remote change was encountered. Along the tract of moving parasite the host tissues were destroyed and became necrotic with numerous nuclear debris, fibrin deposits and inflammatory cells.

Grossly the lesion was composed of elongated tract-like cavity with or without tape-like parasite.

The lesion was not encapsulated. Although definite fibrous capsule was seen in one of the cases, often the worm parasite could be expelled from the lesion without attached tissue. The cavities probably made by the moving worm were quite characteristic. In three points these were different from those of ordinary bacterial abscess where the tissue underwent liquefaction necrosis by purulent inflammation. First, the lumina of these cavities were generally empty

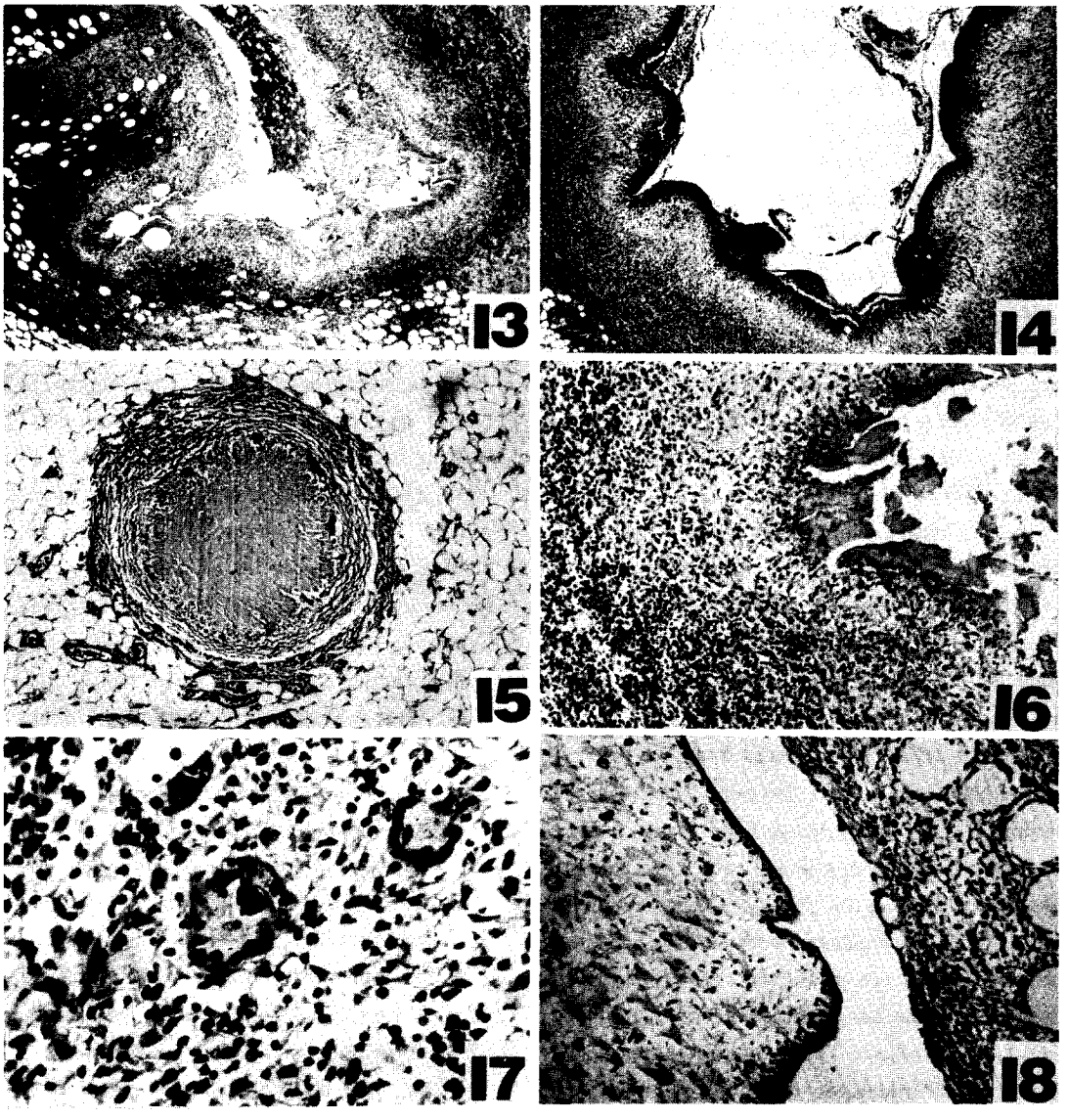

Fig. 13. Sparganosis. Irregular cavitary necrotic lesion seen in fibrofatty tissue. Note also reaction zones around the cavity. H&E ×40

Fig. 14. Another case of sparganosis in subcutaneous tissue, showing irregular scalloping margin with active necrosis. No worm is seen. H&E ×40

Fig. 15. A small focus of granuloma with degenerated *Sparganum* in the middle. H&E ×40

Fig. 16. Palisading histiocytic proliferation is seen around the necrotic worm. H&E ×100

Fig. 17. Higher magnification of multinucleated giant cells engulfing characteristic calcospherules of *Sparganum* in their cytoplasms. H&E ×360

Fig. 18. A portion of *Sparganum* is seen, with adjacent host tissue showing acute inflammatory cell infiltration and fibrin exudation. H&E ×100

although sometimes they contained a small amount of serous fluid or blood. Second, the inner margin of these cavities were characteristically serrated, which probably indicating previously expanded portions of the worm that were partly collapsed following the movement of the

worm into other areas. Third, the wall of the cavities showed distinctive zonal phenomenon in all sorts of tissue it involved. A thin layer of fibrin and necrotic debris and eosinophils consisted the inner-most zone, outside of which showed a broad band of histiocytes and fibroblasts. Here the histiocytes were frequently palisaded and often accompanied with multinucleated giant cells. Sprinklings of lymphocytes could be seen in this zone. Next broad zone was extending into surrounding fibroadipose muscular tissue, consisted of heavy infiltration of eosinophils and some granulation tissue and lymphoplasmacytic cells. When the worm was found in the tract, histiocytes and neutrophils were the main components of the infiltrates, whereas histiocytes and lymphocytes predominated in the lesion without containing worm parasite.

It was interesting to note that calcospherules were occasionally seen in the cytoplasm of macrophages or multinucleated giant cells. These calcospherules were apparently derived from the worm parasite, because these areas showed degenerating parasite bodies that are in the process of absorption by the host tissue. This finding was particularly interesting because the proximal portion of this degenerating parasite was fairly well preserved with evidence of viability.

Charcot-Leyden crystals were not regularly seen in the lesion. They were seen in only two cases of subcutaneous infections in a small scale. The crystals were present among numerous eosinophils and some necrotic debris and fibroblasts. They were found singly or in loose clusters and were in the form of long slender spindles with almost straight sides, tapering from the center to a sharp point on either end. They measured approximately $3 \times 30 \mu m$. In hematoxylin-eosin stained preparations they were seen as pink color. It showed no birefringence under polarized light.

DISCUSSION

Sparganosis is human infection by a larval form of an animal tapeworm, genus *Spirometra*. The definitive hosts for the adult worm are domestic cats and dogs and their wild counterparts. The eggs are shed by the adult worm into the gastrointestinal tract of their definitive host and pass with the feces into the water. A ciliated embryo (coracidium) hatches from each egg and is ingested by the first intermediate hosts (*Cyclops* species). The procercoid larva develop in the body cavity of the *Cyclops*. When the crustacean is swallowed by the second intermediate and transport host usually a water snake, frog, mouse, raccoon, or opposum, the procercoid larva penetrates the gut wall and enters the mesentery. It travels to the root of the mesentery and thus reaches the body wall, in which it migrates to muscles or subuctaneous tissue. Here the plerocercoid larva (called a *Sparganum*) develops.

When the second intermediate host is eaten by a cat or a dog the *Sparganum* attached to the mucosa of the gut and the adult tapeworm is generated. Sparganosis occurs when man is involved in the life cycle as the second intermediate host.

Although it was generally known that the usual source of human sparganosis is the ingestion of infected *Cyclops* in contaminated water, eating of raw snake in this country seemed to be the most important source of infection (Weinstein *et al.*, 1954; Cho *et al.*, 1975). It is not known in this series how many patient had the history of eating raw snakes of other possible intermediate host. However, at least 3 patients whom the history could be checked in detail we could not elicit any positive history of consuming raw snake.

All of the 16 cases we described in this series

were diagnosed to be infected with *Sparganum mansoni* rather than *Sparganum proliferum*. This was based on worm morphology, namely the longitudinal muscles, nerve cords and excretory canals. According to Mueller(1938), spargana of the mansoni type have the dorso-ventral and transverse muscle fibers cross at right angles, when viewed in cross sections, and thus impact a rectangular pattern to the section, whereas in *S. proliferum* the muscle fibers follow no particular plan.

In general the tissue reaction in sparganosis appeared to be fairly stereotypic. There was no definite trend of "aging" phenomenon among cases we examined. This feature was quite in contrast to the cysticercosis in which definite staging, first to third, could be determined. This difference probably represents the difference in worm motility between two parasites. While *Cysticercus* worm is settled in one area and makes a cystic lesion, *Sparganum* worm seems actively moving.

We have no definite information on the longevity of the *Sparganum* in human body. Mueller (1938) reported that one feature of human *Sparganum* not shared by other hosts of spargana is his greater longevity. Using monkeys Mueller could confirm that larva well over a year old showed no evidence of senility. He also described that there was every indication that they were able to survive for a period of years, perhaps indefinitely.

Fragmentation of the worm takes place in the worm in its entirety or in part. When this process occurs in a portion of a worm it may be still possible for the parasite to maintain its life if the scolex portion is not affected. Mueller described that this fragmentation of the organism is apparently due to lesser motility of the posterior part of the larva compared to the great activity of the head. Once fragmented this portion is partly phagocytosed by macrophages. In this regard laminated calcospherules often found in the cytoplasm of the proliferating macrophages and multinucleate giant cells give an important clue of sparganosis, particularly when the tissue reaction is accompanied with the tunnel-like lesion. The calcospherules are unique and important structures in Cestode worms, and are apparent in larval worms as well (Slais, 1970). Although calcospherules were often found in the parenchymal(scolex and spiral canal) portion of *Cysticercus cellulosae*, they were seldom seen in the bladder portion of the worm. And in cases they were found in the bladder they were not laminated(Chi & Chi, 1978). These findings together with characteristic muscle bundles and tegumental structure of the worms enabled us to make diagnosis of sparganosis.

Since most lesions in this series were extirpated at Outpatient Clinic under local anesthesia hematological data of these patients were not available. However, Mueller(1938) reported 25 to 30 per cent eosinophilia in early stage of experimental infection of procercoids in rhesus monkeys.

SUMMARY

Based on 16 cases of human sparganosis, a histopathological study was made. There was a striking similarity among histological features of sparganosis involving different tissues.

The histological change of the affected tissues was characterized by a necrotizing and granulomatous inflammation with or without worm parasite in the lesions. There was also a remarkable polymorphonuclear leukocytic mobilization, predominantly of eosinophils, plasma cells and lymphocytes in and near the lesions. Tunnel formation lined by palisading histiocytes was another characteristic feature of the host tissue reaction. These findings were quite distinguishable from those of cysticercosis which were more

localized and self-limited.

Several features that were prominent in section slides of *Sparganum* worm parasite were also noted. Laminated calcospherules found in the cytoplasm of the proliferating macrophages and giant cells were of diagnostic value of sparganosis in the absence of the worm, particularly when these were accompanied with tunnel-like lesion in the host tissue.

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＝國文抄錄＝

人體 스팔가눔症의 病理組織學的 檢索

서울大學校, 中央大學校 醫科大學 病理學敎室 및 寄生蟲學敎室

池 堤 根·池 賢 淑·李 純 炯

韓國에서 드물지 않게 발생하고 있는 스팔가눔症은 有鉤嚢尾蟲症 및 肺吸蟲症의 異所寄生例와 더불어 중요한 組織寄生蠕蟲症이 되고 있다. 우리나라 사람들의 몇가지 習慣的 행위로 말미암아 本症은 앞으로도 續出할 展望이며, 따라서 이에 關한 正確한 知識이 醫療人들에게 필요하게 되었다.

著者등은 1971년부터 1979년까지의 기간동안 서울大學校 醫科大學 病理學敎室에서 檢査된 16예의 스팔가눔症을 토대로 하여 病理組織學的 檢索을 시행하였다. 스팔가눔 蟲體 및 주위 조직의 반응으로 보아 本症은 病理學的으로도 상당히 特徵的 所見들을 보였고 이들 모든 例를 綜合하건데 顯微鏡 標本만 가지고도 診斷에 상당히 가깝게 갈 수 있음을 알 수 있었다.

一般的으로 스팔가눔症의 病理組織學的 特徵은 多發性 空洞性 壞死性 肉芽腫性 炎症으로서 이는 蟲體의 有無와 相關없이 나타났다. 이러한 變化는 疾病期間과도 直接 關聯이 없는 듯 하였으며, 다만 蟲體가 生體內에서 變性되는 경우는 宿主組織에 의하여 處理되는 現象을 보였다.