CASE REPORT

OCULAR SPARGANOSIS IN THAILAND

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Human sparganosis which was first reported by Patrick Manson in 1882 (Beaver, 1984) is a rare parasitic infection caused by the migrating plerocercoid larva or sparganum of a certain group of pseudophyllidean cestodes, namely in the genus *Spirometra* Mueller, 1937. The adults of these tapeworms are generally found in the intestinal lumen of domestic, feral, or wild canids and felids (Daly, 1982). Spargana, a term originally used to describe any unknown pseudophyllidean larvae, do not have adequate morphological characteristics for differentiation. Therefore, they must be fed to a definitive host (dog or cat) to grow to adult worms which have morphological differences that can be used for taxonomic separation. This, however, has not been accomplished for spargana from most of human sparganoses. Thus, an opinion as to species of the sparganum was usually based on the general nature of the larva, knowledge of adult forms in hosts from that area, or previously reported sparganosis cases from that area. This leads to invalidity of species in many cases because certain forms are sometimes found in the same geographical regions, such as *S. mansoni* and *S. mansonoides* in North and South America (Mueller *et al,* 1975) and/or knowledge of adult forms in those areas is inadequate. Furthermore, the variability in adult worm morphology as well as lack of communication and agreement among the ivestigators, has led to confusion about the classification of this group of tapeworms.

Human sparganosis has been reported sporadically worldwide, from every continent but most commonly from East and Southeast Asia (Japan, China, Korea, Taiwan, Vietnam, Thailand). There are two types of the disease, namely nonproliferative and proliferative sparganosis. Review of the literature revealed more than 300 cases of nonproliferative sparganosis in humans but there have been only 11 well-documented and 2 other suspected cases of proliferative type (La Chance *et al,* 1983; Nakamura *et al,* 1990).

A 33-year-old female Thai had had history of pain, swelling, and redness of her left eye since June 1990. She went to medical clinics twice and the last doctor suggested she come to Maharaj Nakhon Chiang Mai Hospital, Faculty of Medicine, Chiang Mai University. At Ophthalmologic OPD, the diagnosis of allergic blepharitis and conjunctivitis was made but showed no clinical improvement after standard treatment. Follow-up examination revealed localized conjunctival swelling in the temporal region of her left eye. Gnathostomiasis was suspected. Palliative therapy was given with some degree of improvement and the patient had been absent for 2 years. She revisited in October 1992. A left subconjunctival mass was noted. Other findings were unremarkable except the presence of *Strongyloides stercoralis* larvae in the stool. No preoperative hemogram was done. Exploration of the lesion revealed a white ribbon-like cestode larva. ELISA using 36 and 31 kDa sparganum antigens (kindly donated by Dr Yoon Kong) gave a positive antibody value of 0.263 (cut-off = 0.22); 3 days after surgery; which was negative 4 months later. The patient's serum was also tested for gnathostomiasis 7 days postoperatively. The ELISA value was 0.273 (cut-off = 0.44). The WBC count 2 weeks after surgery was 14.1 $\times$ 10^9/l of which 2% were eosinophils. She was well on follow-up at 4-month.

The parasite was sent to The Department of Parasitology for identification. It measured 3.2 $\times$ 1 mm in 10% formalin fixative (Fig 1). Slight enlargement of one end suggested it be a scolex. A piece of the worm was processed for histological sections and stained with hematoxylin-eosin. The tegument of 5 - 15 μm thickness, a single layer of

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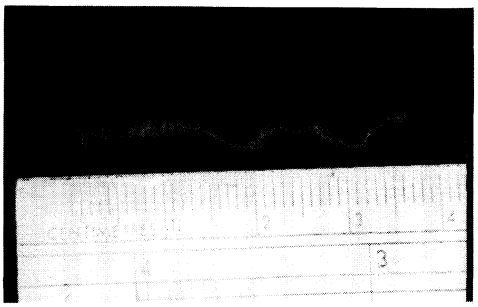

Fig 1—A Sparganum removed from the patient's eye. Bulging of the end on the left is the parasite's scolex.

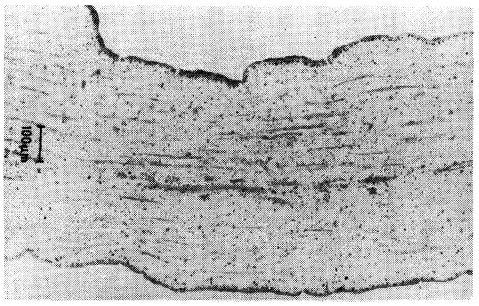

Fig 2—Histological section revealed typical fetures of a sparganum. H and E (x10).

subtegumental cells, loose parenchyma with diffuse parenchymal cells, laminated calcareous corpuscles, excretory channels, and presence of longitudinal muscle bundles throughout its stroma were definite for diagnosis of a sparganum (Fig 2).

In Thailand, there have been at least 27 reported cases of sparganosis of which 14 cases (52%) were ocular, 9 (33%) subcutaneous, 3 (11%) intracranial, and 1 (4%) intraperitoneal infections (Table 1). The mean age was 29.57 (range 11 - 60, n = 23) with male:female ratio = 1:2 (n = 27). Risk behaviors were either not recorded or ambivalent in most cases. In our case, the patient lived in Amphoe Fang, Chiang Mai. She strongly denied ingestion of any kind of raw meat but she habitually drank water from temporarily open dug wells during her working days in the rice fields which might be polluted with procercoid-containing first intermediate host. The eosinophil count ranged from normal to markedly elevated. The duration from first appearance of symptoms to settlement of the definite diagnosis varied form less than 1 month to several years depending on awareness of patients and doctors, sites of infection, and coexisting disease as in case no. 14, a medical student who presented with severe abdominal pain mimicking acute appendicitis but actually cause by a right ovarian cyst. A sparganum was coincidentally found in the pelvic cavity.

Most cases of ocular sparganosis present as extraocular infections. The early phase of the disease mimics allergic blepharitis or conjunctivitis. Eosinophilia, localized swelling, and no or little response to anti-allergic drugs favor the diagnosis of parasitic infection. The parasite may invade into the eye ball where it can cause severe inflammation and blindness (Sen *et al,* 1989). Poulticing of frog meat on the eyes may cause multiple infection (as in case no. 6).

Morphology and histology of sparganum has been intensively studied. Grossly, it cannot be differentiated from other plerocercoid-type larvae, *ie* tetrathyridium of a cyclophylidean cestode genus *Mesocestoides* (Andersen, 1983). Therefore, microscopic study is essential for diagnosis. Serological techiques were recently developed in Korea. Specific antigens, molecular weight of 31 and 36 kDa, were purified from sparganum by immunoaffinity chromatography. The antigen was tested for its sensitivity and specificity by ELISA. It exhibited 96.4% sensitivity. Cross reactivity was associated with cysticercosis (11.1%), taeniasis saginata (12.5%), paragonimiasis (6.3%) and clonorchiasis (6.7%) (Cho *et al,* 1990). Further evaluation with sera from 25 gnathostomiasis, 33 angiostrongyliasis and 22 trichinosis revealed positivity in one of each group (unpublished data). Thus, the antigen is useful in differentiating migratory swellings due to sparganum and *Gnathostoma.*

The situation of sparganosis in Thailand is unclear at this time. We have little knowledge about the responsible species, endemic foci, impact on public health, and diagnostic tools. The history and clinical picture of sparganosis are similar to gnathostomiasis. Nevertheless, the treatment may be different between the two, especially in inoperable cases, since albendazole was recently demonstrated to be effective against the latter (Kraivichian *et al,* 1992) but the appropriate drug for the former is still uncertain (Torres *et al,* 1981). Moreover, sparganosis may imitate other more common helminthiases in case of CNS

Table 1

Review of 27 human sparganoses in Thailand.

Case no.	Report year	Age (years)	Sex	Organ	Risk habit[a]	Duration	Eosinophil count[b]
1	1943	39	M	Eye	-[c]	-	-
2	1950	13	F	Abdominal wall	-	5 months	-
3	1960	14	F	Eye	D,I	5 months	33% of?
4	1960	13	F	Eye	D,I	15 days	33% of?
5	1960	11	F	Eye	-	7 days	-
6	1964	25	F	Both eyes	P	5 years	1,230
7	1964	33	F	Eye	D	8 years	458
8	1964	19	F	Abdominal wall, left thigh	-	20 days	-
9	1965	40	M	Abdominal wall	-	-	-
10	1965	18	F	Forehead	-	-	-
11	1968	-	M	Eye	-	-	-
12	1968	-	F	Eye	-	-	-
13	1971	46	F	Brain	-	3 years	-
14	1974	23	F	Peritoneal cavity	D	4 days	1,320
15	1975	24	M	Subcutis of neck	-	1 month	-
16	1978	-	M	Subcutis of scrotum	-	9 months	-
17	1978	17	F	Subcutis of groin region	-	-	-
18	1979	23	F	Subcutis of thigh	-	-	-
19	1990	60	M	Subcutis of thumb	-	-	-
20	1980	34	M	Right upper eyelid	D,I	3 months	1,185
21	1981	34	F	Right upper eyelid	-	3 years	64
22	1984	-	M	Foramen monro of brain	-	-	-
23	1985	25	F	Pontomedullary junction[d]	-	1 year	-
24	1988	51	F	Inner ear and eye	D	5 months	347
25	1989	37	M	Eye	D	1 month	-
26	1992	51	F	Eye	-	1 week	1,825
27	1993	30	F	Eye	D	3 years	282

a: D = drinking impure water, I = ingestion of frog or snake meat, P = use frog meat as poultice
b: expressed as cells per µl
c: '-' = data unavailable
d: A cysticercus was found in the adjacent area

Case no. 1,2,5,8,9,10	(Tansurat, 1966)
Case no. 3,4	(Sampavapon, 1960)
Case no. 6	(Bedavanija and Namatra, 1964)
Case no. 7	(Jipipop and Chenpanich, 1964)
Case no. 11,12,13,15,16,17,18,19	(Tesacharoen, 1980)
Case no. 14	(Khamboonruang *et al*,1974)
Case no. 20	(Masrungsan *et al*, 1980)
Case no. 21	(Masrungsan *et al*, 1981)
Case no. 22,23	(Pongprasert and Somboon, 1985)
Case no. 24	(Kittiponghansa *et al*, 1988)
Case no. 25	(Jenchitr *et al*, 1989)
Case no. 26	(Kasantikul *et al*, 1991)
Case no. 27	The present case

infection (gnathostomiasis, cysticercosis, angiostrongyliasis).

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