Reminder of important clinical lesson

Sparganosis of liver: a rare entity and review of literature

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Summary

Humans are known to serve as the secondary intermediate host for some larval forms of canine or feline tapeworms. Sparganosis is caused by plerocercoid larva of *Spirometra* of which there are three main species; *Spirometra mansoni* (or *Spirometra erinaceieuropaei*), *Spirometra mansonioides* and *Spirometra proliferum*. A one-and-half-year-old patient presenting with febrile illness was diagnosed radiologically as a case of liver abscess. The aspirate from the abscess cavity had a thin thread-like worm which was identified as a larval stage of Pseudophyllidea that was further confirmed as belonging to genus *Spirometra*. Aspiration of the worm and antiparasitic therapy with metronidazole led to complete recovery. Reports of sparganosis from Indian subcontinent are scanty and these cases had involvement of brain, kidney and eye. To the best of our knowledge this is the first case of hepatic sparganosis from India.

BACKGROUND

Humans are known to serve as the secondary intermediate host for some cestodes of the order Pseudophyllidea.^{1 2} Sparganosis is caused by plerocercoid larva of genus *Spirometra* of which there are three main species; *S. mansoni* (or *S. erinaceieuropaei*) causing infection mainly in Asia, *S. mansonioides* mainly in Americas and *S. proliferum* which is a serious migratory form. In the year 1882, Patrick Manson reported the first case of sparganosis caused by *S. mansoni* in Amoy, China. In the USA, the first case of sparganosis caused by *S. proliferum* and *S. mansonioides* was reported by Stiles (1908) and Muller (1935), respectively.

The disease is generally localised to the subcutaneous connective tissue and usually self-limiting except in case of *S. proliferum* where it disseminates to other organs. On rare occasions, it involves the sites like eye and periorbital tissue, brain and abdominal viscera. Humans act as the accidental host and can acquire infection by one of the many ways including drinking water containing infected copepods, eating uncooked or undercooked amphibian, reptile or rodent meat or using those meats as a poultice, a traditional treatment in some parts of Asia. Humans accidentally harbour the larval stage (plerocercoid-sparganum) of the helminth. Clinical presentation depends on the site of involvement. Diagnosis can be made by demonstration of the larval stage of worm from biopsy specimen but species differentiation is not possible. No medical therapy has proven useful and surgical removal is the treatment of choice. Sparganosis has been reported sporadically around the world, and a higher prevalence of the disease occurs in Asian countries, including South Korea, Japan, Thailand and China. Reports from Indian subcontinent are scanty and most of the cases had involvement of brain, few had renal and eye involvement.^{3 4} Here we report the first case of hepatic sparganosis from India.

CASE PRESENTATION

A one-and-half-year-old girl presented to local private hospital with complaints of high fever, vomiting, poor

feeding and irritability for 4–5 days. Vomiting was non-projectile and non-bilious. Fever episodes were not associated with rash, bleeding or chills and rigor. Initial haemogram revealed leucocytosis with predominant polymorphonuclear cells and marginally raised eosinophils (total leukocyte count: 12.6×10^9 , differential leukocyte count: polymorphs—65%, lymphocytes—29%, monocytes—2% and eosinophils—4%). Peripheral blood examination showed moderate red blood cell microcytaemia with mild decrease in cell mass. Cerebrospinal fluid examination did not reveal any abnormality. Patient was started on cefoperazone-sulbactam and amikacin along with supportive treatment for 1 week. Despite therapy, the patient continued to have febrile episodes. Subsequently an ultrasonography (USG) was done on day 3 of hospital admission (day 8 of fever) which revealed a well-defined heterogeneous hypoechoic lesion of 8.7×7.4 cm in the right lobe of liver. Patient was referred to the Postgraduate Institute of Medical Education and Research, Chandigarh, which is a tertiary care centre in North India. A repeat USG confirmed the presence of liver abscess and pigtail catheter drain was inserted into the abscess cavity through which frank pus drained out. Patient was started on ceftriaxone, amikacin, cloxacillin and metronidazole. A provisional diagnosis of pyogenic liver abscess was made and the pus aspirate was sent for bacterial culture and sensitivity which was reported to be sterile. Patient had hypoalbuminaemia (albumin—1.7), which was treated with albumin infusion. On the third day of admission, the patient had signs and symptoms of pleural effusion for which patient was given nasopharyngeal continuous positive airway pressure. Haemogram showed evidence of ongoing inflammation with leucocytosis of 26.4×10^9 , and predominant polymorphonuclear cells and marginally raised eosinophils (4%). Amoebic and hydatid serology were negative. Intervention radiology done at this stage showed a semiorganised abscess of 6×5 cm dimensions in the right lobe of liver. The pig tail drain was continued in situ and saline flushes were done

Figure 1 Unstained larva of *Spirometra*

to aspirate out the thick pus. While doing the saline flush the treating resident noted a long thread-like structure coming out of the drain which was sent for parasitological evaluation. The worm was subjected for fixation and staining. The worm had flat, long and thread like (approximately 35–40 cm long and 1.5–1.7 mm broad)

whitish unsegmented wrinkled body with a groove like mouth part with no suckers or hooklets and on further microscopic evaluation, longitudinal muscular striations were noted (figures 1 and 2). The worm was identified as larva of *Spirometra* spp., and treating unit was advised to rule out possible involvement of other sites like eye and central nervous system (CNS). However, no abnormality was noted in either eye or CNS. Patient was also evaluated for immunosuppression, and a screening nitro blue reduction test was negative. At the end of second week, patient was afebrile and pig tail discharge was also minimal. A repeat USG showed a 5×4 cm partially organised residual abscess. Metronidazole therapy was stopped after 15 days but other antimicrobial agents like ceftriaxone-cloxacillin were continued for one more week to cover for superadded bacterial infection. The computerised axial tomography abdomen showed no evidence of intrahepatic or intraluminal residual worm. Finally, the USG done at the end of third week showed 3×2 cm organised collection. Patient was discharged after the pig tail catheter was removed and daily wound care of catheter site advised. Six months after, the patient was examined in outpatient department and was growing well with no signs or symptoms of the disease.

INVESTIGATIONS

Sparganosis in humans is usually diagnosed following surgical removal of the larvae. However, species identification requires feeding the spargana to a proper definitive host and recovering the adult parasite for morphological study. The antisparganum ELISA having good sensitivity (100%) and specificity (96%) may also be used.⁵ CT and MRI scans are especially useful for diagnosis of visceral sparganosis. In our patient, USG of the abdomen revealed abscess cavity in the liver. Thus detailed clinical history, haematological evaluation, radiological tests and

Figure 2 Stained larva of *Spirometra*

serological tests may be used to make a diagnosis of sparganosis. However, availability of serological test kits may be problem in many parts. Surrogate markers like eosinophilia can be a soft pointer towards parasitic infections in general but may not be there in all the cases

DIFFERENTIAL DIAGNOSIS

In endemic countries other infections like tuberculosis, cysticercosis, amoebiasis and paragonimiasis that cause space occupying lesion should be considered in differential diagnosis.

TREATMENT

Surgical removal of sparganum larvae is often the best treatment.¹ Medical therapy includes antiparasitic drugs like metronidazole and praziquantel. However, medical therapy used alone is shown to have limited success.

OUTCOME AND FOLLOW-UP

In our patient a course of metronidazole and removal of the worm led to recovery.

DISCUSSION

Sparganosis is endemic or potentially endemic in 48 countries but the majority of cases occur in Southeast Asia and Eastern Africa. A higher prevalence of the disease occurs in Asian countries, including South Korea, Japan, Thailand and China. Reports from Indian subcontinent are scanty and most of the cases had involvement of brain, few had renal and eye involvement.³⁻⁴ Here we report the first case of hepatic sparganosis from India. In a thorough English literature review we did not find reports of similar cases in the past.

Learning points

- ▶ Sparganosis in general and hepatic sparganosis in particular is very rare.
- ▶ Hitting upon the definite diagnosis is important for the proper management of the patient.
- ▶ The diagnosis can be reached upon by keeping a high index of suspicion in endemic areas followed by either recovery of the parasite, or by indirect tests like antibody detection. Surrogate markers like eosinophilia can be a soft pointer towards parasitic infections in general but may not be there in all the cases.

Competing interests None.

Patient consent Obtained.

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