

9-1-1989

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Recommended Citation

Ladpli, Roongtam and Panichabhongse, Viratt (1989) "Cerebral schistosomiasis : the first case in Thailand," *Chulalongkorn Medical Journal*: Vol. 33: Iss. 9, Article 7.

DOI: 10.58837/CHULA.CMJ.33.9.7

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Cerebral schistosomiasis : the first case in Thailand.*

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Ladpli R, Panichabhongse V. Cerebral schistosomiasis : the first case in Thailand. Chula Med J 1989 Sep; 33(9): 681-688

Schistosomiasis, caused by the blood fluke schistosome, is a very rare disease in Thailand. While in 1959 a case of intestinal schistosomiasis was reported from Thailand this is our first case of cerebral schistosomiasis. A 36 year old Khmer female refugee, having stayed at a Border Camp for many years presented with a history of right temporal headache and seizures of left eyelids for two-months. Computed tomography revealed a right frontoparietal mass. The mass was totally removed and histopathological examination of the mass showed a granuloma with ova of schistosome. Post-operatively the patient recovered uneventfully and stopped complaining of headache and seizure. The pathogenesis and management of schistosomiasis as reviewed from the literatures are also discussed.

Reprint request : Ladpli R, Department of Surgery, Siriraj Hospital, Mahidol University.
Received for publication. May 22, 1989.

* This case had been presented earlier in annual meeting of Royal College of Surgeons of Thailand held in Grand Palace Hotel Pattaya on 5 July 1982 and in annual scientific meeting, Faculty of Medicine, Chulalongkorn University, on April 22, 1982.

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รุ่งธรรม ลัดพลี, วิรติ พาณิชย์พงษ์. สมอังกะเสบจากพยาธิใบไม้ในเลือด: รายแรกของประเทศไทย. จุฬาลงกรณ์เวชสาร 2532 กันยายน; 33(9): 681-688

Schistosomiasis เป็นโรคที่เกิดจากพยาธิใบไม้ในเลือด ซึ่งพบน้อยมากในประเทศไทย และพยาธิสภาพที่เคยรายงานในประเทศไทยก็พบที่ระบบทางเดินอาหารเท่านั้น รายงานผู้ป่วยรายนี้จึงเป็น *cerebral schistosomiasis* รายแรกในประเทศไทย ซึ่งพบเมื่อเดือนสิงหาคม 2524 ที่โรงพยาบาลกรุงเทพ

ผู้ป่วยรายนี้เป็นผู้อพยพมาจากประเทศเขมร ในระหว่างเจ็บป่วยพักอาศัยอยู่ในประเทศไทยเป็นเวลานาน มีอาการเกิดขึ้นเพียงสองเดือน ผู้ป่วยได้รับการตรวจด้วย *CT brain scan and cerebral angiography* พบว่ามีก้อนขยายที่สมองด้านขวา ได้รับการผ่าตัดเอาก้อนออกและมีอาการทุเลา การตรวจทางพยาธิสภาพพบว่ามี *ova Schistosoma* อยู่ในก้อน *granuloma* นั้น

ผู้รายงานจะได้เสนอรายงานผู้ป่วยโดยละเอียด พร้อมกับรวบรวมและวิจารณ์วารสารที่เกี่ยวข้องตลอดจน *pathogenesis* และการรักษาที่ถูกต้อง และได้เสนอรายงานนี้แล้วในการประชุมประจำปีครั้งที่ 7 ของราชวิทยาลัย ศัลยแพทย์แห่งประเทศไทย เมื่อวันที่ 5 กรกฎาคม 2525 ณ โรงแรมแกรนด์พาเลซ เมืองพัทยา ซึ่งเป็นรายเดียวกับที่ ศ.นพ. สำรวย ช่างโชติ รายงานในการประชุมวิชาการประจำปี 2525 เมื่อวันที่ 22 เมษายน ณ คณะแพทยศาสตร์ จุฬาลงกรณ์มหาวิทยาลัย.

Schistosomiasis or Bilharziasis is a parasitic disease caused by the blood fluke schistosome of which the three main species (*Haematobium*, *Mansoni*, *Japonicum*) and one variation (*Maekongi*)^(1,2) have different geographic distribution (Figure 1.) with slight differences in their life cycles and clinical manifestations. (Figure

2,3). The important point to notice is that each species mentioned above may have ectopic lesion in the lung, spinal cord or very rarely the brain depending on the distribution of eggs to the different organs via blood circulation.^(3,4)

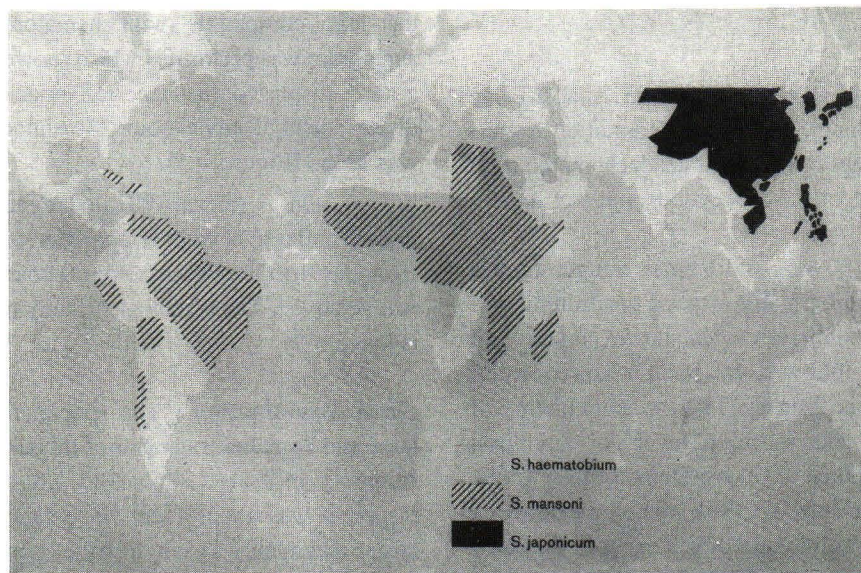


Figure 1 Geographic distribution of the schistosome.

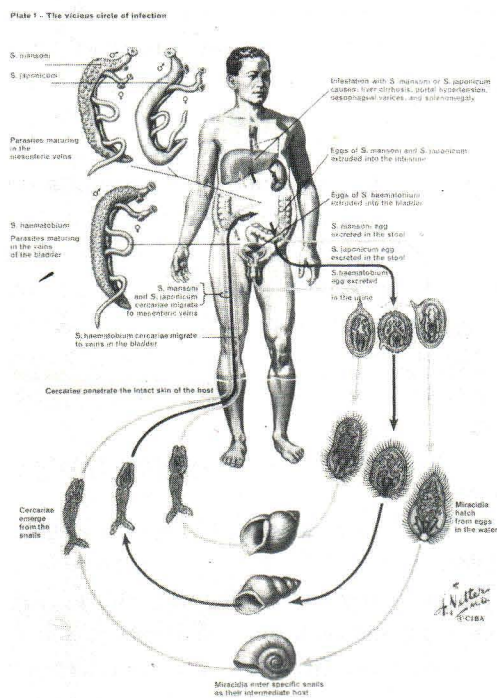


Figure 2 The vicious circle of infestation of schistosome.

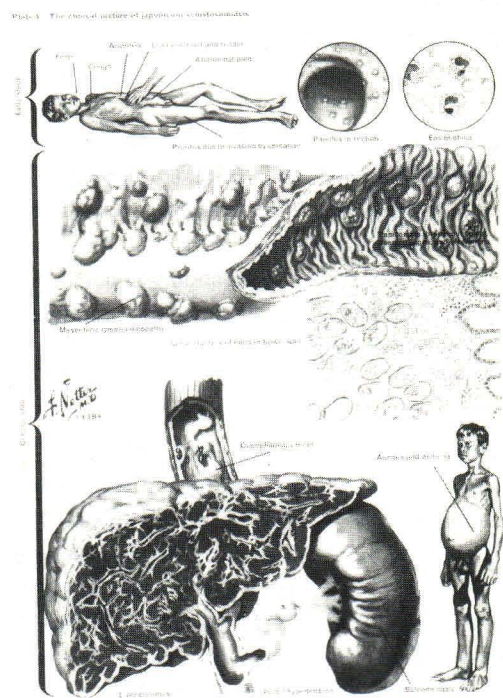


Figure 3 The clinical picture of japonicum schistosomiasis.

Schistosomiasis in Thailand is very rare; the first case was reported in 1959 and was of intestinal schistosomiasis.^(3,5) It is our intention to present the first in Thailand found with these particular manifestations of schistosomiasis although the patient was not a Thai native she had stayed in a refugee camp at the eastern border of Thailand for more than 2 years before she manifested with the parasitic infestation.

Case report

This patient was referred from Khao-I-Dang Medical Center to Bangkok General Hospital on August 10, 1981 with the chief complain of headache in the right temple and twitching of the left eyelids and left hand.

Present illness : A 36 year-old Khmer refugee female farmer who had been basically in good health until two months prior to the admission when she developed right temporal headache followed by focal seizure of left eyelids, left thumb and fingers. There was no history of unconsciousness, weakness, high fever, or any head trauma. She was admitted to Khao-I-Dang Refugee Camp Medical Center in on July 16, 1981 where an evaluation revealed no definite neurological deficit except mild ptosis of the left eyelid. Only relevant laboratory findings were elevation of blood eosinophils (11%) and of CSF Protein (Pandy positive 3+). During admission at Khao-I-Dang, medical treatment failed to improve her condition; although the seizures were controlled, the patient developed high fever and headache. The patient was then transferred to Bangkok General Hospital on August 10, 1981.

Physical examination : Blood pressure 110/80, pulse rate 80/min, respiration rate 20/min, temperature 37.0°C. Examination revealed a well-developed, well-nourished female with no acute distress. She was not anemic or jaundiced, and only slight dehydration was noted. Routine general and neurological examinations were within normal limits except bilateral papilledema and slight hyperreflexia on the left side.

Laboratory data : On admission, WBC was 5,750, PMN 76%, Lymph 18%, Eos 6%, hemoglobin 12.0 gm%, hematocrit 35%, FBS 80 mg%, BUN 10 mg%, creatinine 0.6 mg%. Urinalysis was normal. Blood group was "B". On 17 Aug 1981, WBC was 26,900, Hb 10.6 gm%, Hct 33.0%, PMN 79%, Lymph 17%, Mono 4%. Stool examination was positive for opisthorchis ova and Trichononas hominis. On 21 Aug 1981, WBC was 9,500, PMN 82%, Eos 1%, Lymph 14%, Mono 3%, Hb 11.8 gm%, Hct 36%. BUN 12 mg%, creatinine 0.8 mg%, sodium 137 mEq/L, potassium 5.0 mEq/L, chloride 93%

mEq/L, CO₂ CP 21.20 mEq/L. Chest X-ray revealed patchy infiltration of the posterior segment of left upper lobe but other lobes were normal. Skull X-ray was normal.

Hospital course : On 11 August 1981, the C.T. of brain revealed a right frontoparietal mass surrounded by edematous area which displaced the midline structure to the left, compatible with hematoma. Right carotid angiogram was performed which showed anterior cerebral artery displaced to the left side, and downward displacement of sylvial point. No tumor stain or aneurysm was seen. (Figure 4,5)

On 13 August 1981 under general anesthesia the patient underwent right temporoparietal craniotomy. An irregular firm tumor mass approximately 4.5×3×2 cm. was removed. The pathological study revealed as follows. (Figure 6-7)

Gross Examination : The specimen consisted of one large and 3 smaller fragments of rubbery tissue. The larger fragment measured 4.5×2.0×1.5 cm. Each smaller fragment was about 1 cm. in greatest dimension. The larger fragment was said to have been a bisected part of the total specimen and weighed 5 gm. Its external surface was nodular and circumscribed. Its cut surface was homogenous, smooth, and pale yellow. Representative parts were retained for microscopic study.

Microscopic Examination : The specimen was brain tissue containing numerous granulomas with severe desmoplastic reaction and gliosis. These granulomas were comprised of schistosome eggs with wrinkle and distorted shells, histiocytes, multinucleated giant cells of foreign body type, lymphocytes, plasma cells, granulation tissue, and necrotic foci. The histiocytes and foamy cells frequently had foamy cytoplasm, suggesting the presence of cytoplasmic lipid content and accounted for the pale yellow appearance of the cut surface of the specimen. The size of the eggs were about 30×60 micron. The unusually small size of the eggs was likely related to tissue compression.

Diagnosis was Schistosomiasis of brain, right temporal lobe.

Post-operatively, the patient's condition was uneventful without any complaint of headache or seizure.

On 4 September 1981 the patient underwent sigmoidoscopy and multiple inflammatory necrotic surface nodules were seen. The biopsy showed schistosomiasis. No investigation was done to rule out pulmonary schistosomiasis as suspected.

After the operation medication for parasites was started. Fugacar 1 tab. tid. pc. was given for 3 days for

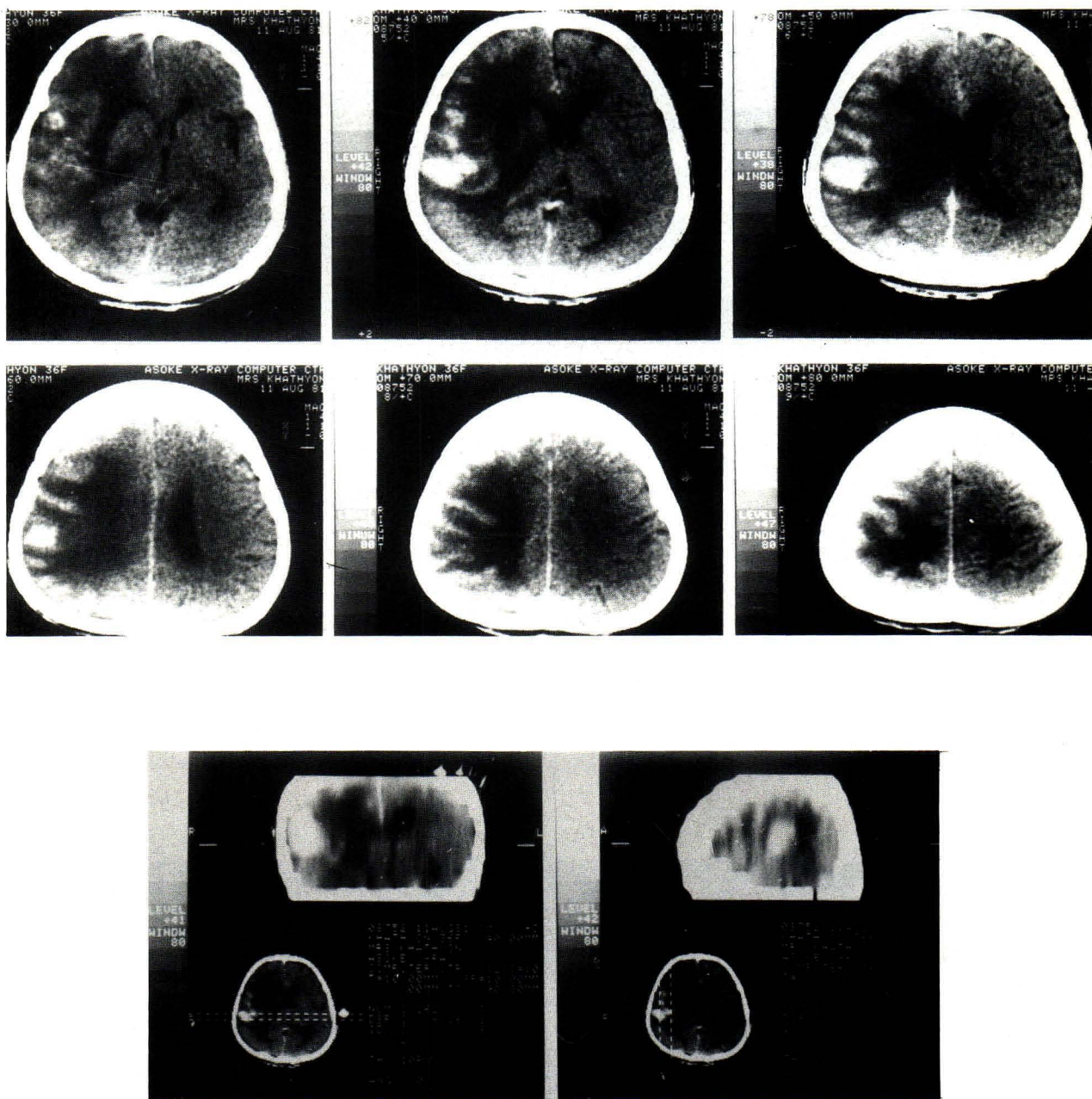


Figure 4,5 C.T. brain scan of the patient.

opisthorchis and potassium antimony tartrate was planned for schistosomiasis at the Khao-I-Dang Medical Center of the refugee camp.

She was discharged on 4 October 1981 and final-

ly departed to her country after three months, with no follow-up despite requests to the medical coordinator of International Red Cross at Khao-I-Dang refugee camp.



Figure 6 The schistosome mass in right frontoparietal region.

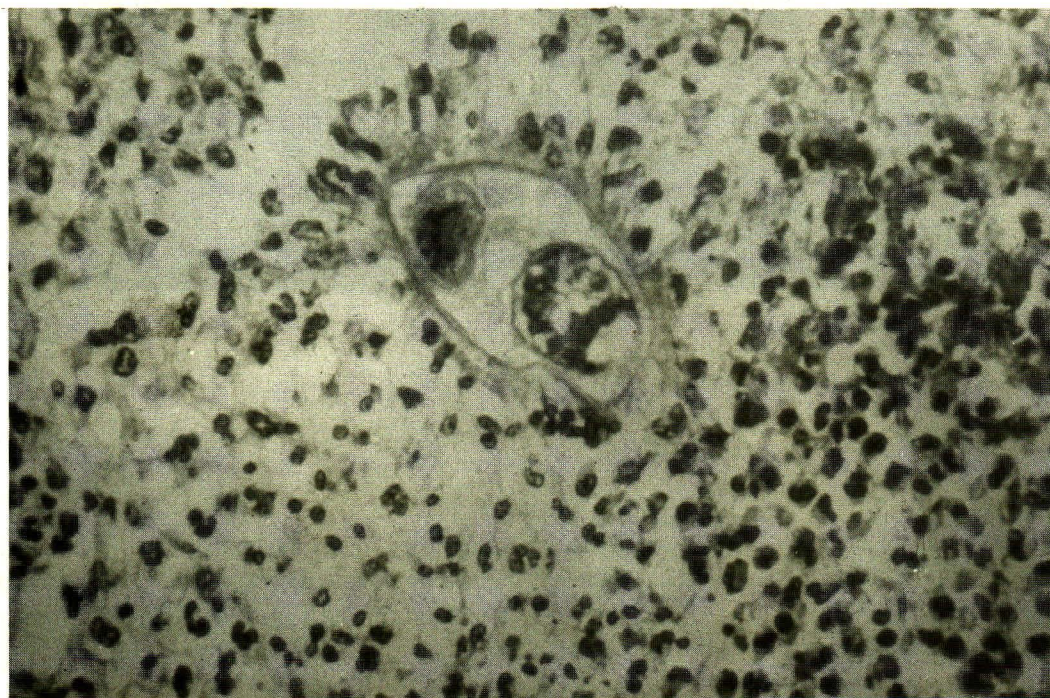


Figure 7 Microscopic examination of the mass showed granutoma and egg of the schistosoma japonicum.

Discussion

Parasitic infestations of the nervous system can be classified presently as follow^(3,4)

1. Cerebral malaria
2. Amebiasis of brain and meninges
3. Toxoplasmosis
4. Trypanosomiasis
5. Trichinosis
6. Eosinophilic meningitis or
Eosinophilic meningoencephalitis
 - A. *Angiostrongylus eosinophilic*
Meningoencephalitis
 - B. *Gnathostoma eosinophilic*
Radiculomyeloencephalitis
7. Cerebral cysticercosis
8. Sparganosis
9. Paragonimiasis
10. Schistosomiasis or bilhaziasis

Concerning the three main species of schistosoma that can involve the C.N.S., schistosoma Japonicum which causes Asiatic intestinal schistosomiasis, can produce ectopic lesion in the brain more than in the spinal cord. The two species, schistosoma mansonii which causes intestinal schistosomiasis and schistosoma haematobium which causes urinary schistosomiasis produces ectopic lesions in the spinal cord more than in the brain. About the pathogenesis of ectopic lesions in the lungs, spinal cord or brain it has been postulated that one of three following mechanisms may play the role.^(3,4)

- I. Through the vertebral venous plexus (By Batson 1940)
- II. Anastomotic channels between the portal and systemic circulation in the pelvis.
- III. Through the posterior mediastinal veins via porta to systemic circulation from hepatic fibrosis.

Clinical manifestations of schistosomiasis are mainly fever and symptoms of the affected organs such as hematuria (causes by *S. Haematobium*), diarrhoea (causes by *S. Mansonii* and *S. Japonicum*) and neurological symptoms and signs may be caused by the direct effect of laying eggs in the brain or egg embolism. The toxin and foreign protein of the eggs would result in cerebro spinal meningitis. Eggs granuloma would be formed and produce space occupying lesion affecting the brain or spinal cord. The indirect effect of schistosomiasis is hepatocerebral disease resulting from liver failure.^(6,7)

Treatment involves both medical and surgical methods.^(1,8)

I. Medical treatment

Trivalent antimony agent (Fuadin)
Niridazole for *S. Masoni*
s. *Hematobium*
Praziquantel for *S. Japonicum*

II. Surgical treatment

mass in brain or cord

surgical treatment for portal hypertension

Schistosoma Mekongi (variation of *Japonicum*)^(2,9) which is mostly seen on the banks of river Mekong and Khong island on southern strip of Laos. Therefore Thailand may get an epidemic of schistosomiasis as there are refugees from Laos residing in Thailand. It has also been seen that intermediate host is present in the Mool river in Thailand.

Conclusion

We have reported one of the unusual cerebral manifestations of schistosomiasis which is most likely to be the first case of cerebral schistosomiasis in Thailand. The patient, a 36 year old Khmer refugee female farmer who was infected with intestinal schistosomiasis, later developed right temporal headache followed by focal seizure of the left eyelids and left fingers. After she was transferred to Bangkok General Hospital, C.T. brain scan and right carotid angiogram were performed followed by right temporoparietal craniotomy for total removal of the cerebral mass which was confirmed as cerebral schistosomiasis. The patient returned to the camp without neurological deficit for medical treatment of schistosomiasis and departed to her country 3 months after discharge.

Acknowledgement

Authors wish to thank and are highly indepted to

- A. Professor doctor. Samruay Shuangshoti, Faculty of Medicine, Chulalongkorn University for confirmation of neuropathological diagnosis and references.
- B. Professor doctor Anond Pradatsundarasar ex-head Department of Parasitology, Faculty of Medicine, Chulalongkorn University for his advice and references.
- C. Doctor Pongsak Viddayakorn director of Bangkok General Hospital for allowing this case to report.

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