รายงานผู้ป่วย

Intramedullary cysticercosis: first case report in Thailand and literature review.

Sunan Prasathapong*

Prasathapong S. Intramedullary cysticercosis: first case report in Thailand and literature review. Chula Med J 1995 Jun; 39(6): 443-450

Intramedullary cysticercosis is rare. A female case is described in which the clinical features of spinal cord tumor, but there was cysticercus cyst within the thoracic cord (T_{10} - T_{11}) seen at surgery. Early surgical removal of the cyst resulted in marked improvement. There have been only 30 cases reported in liturature.

Key words : *Spinal cysticercosis, Intramedullary cyst, Spinal cord compression.*

Reprint request : Prasathapong S. Department of Neurology and Neurosurgery, Somdet Chaopraya Hospital, Klongsarn, Bangkok 10600, Thailand.

Received for publication. March 15, 1995.

*Department of Neurology and Neurosurgery, Somdet Chaopraya Hospital.

สุนันท์ ประสัตถพงศ์. ถุงตัวอ่อนพยาธิตืดหมูในเนื้อไขสันหลัง: รายงานผู้ป่วยแรกในประเทศไทยและทบทวนรายงาน. จุฬาลงกรณ์เวชสาร 2538 มิถุนายน; 39(6): 443-450

ถุงตัวอ่อนของพยาธิตืดหมูที่เกิดในเนื้อไขสันหลัง พบได้น้อยมากมีรายงานเท่าที่ทราบเพียง 30 รายเท่านั้น ได้รายงานผู้ป่วยหญิงหนึ่งรายมีอาการของการกดทับไขสันหลัง ซึ่งพบว่าเป็นถุงตัวอ่อนของพยาธิตืดหมูอยู่ในไขสันหลังระดับ T_{10} - T_{11} การผ่าตัดในระยะเริ่มแรกของอาการจะช่วยในการฟื้นของอาการได้เร็วขึ้น ซึ่งเป็นรายงานผู้ป่วยรายแรกของประเทศไทย

Human cysticercosis is the infestation by larvae of the pig tape worm "Taenia solium". In endemic areas the involvement of the central nervous system is not uncommon. Cysticercosis of the brain and meninges is far more common than of the spinal cord. Intramedullary cysts are extremely rare among the various forms of spinal cysticercosis.[1-3] Clinically, patients with spinal cysticercosis usually are presented with symptoms and signs suggestive of cord compression.[4,5] It is difficult to distinguish between intramedullary cysticercosis and intramedullary neoplasm even with using the magnetic resonance imaging (MRI) scan.

Case report

A 48 year-old Thai woman was admitted to the hospital on 15 July 1993 with progressive weakness of both lower limbs during the past month. About two weeks prior to admission she experienced numbness from groin down to toes bilaterally. About a week later she had pain in both thighs and an occasional cramping sensation in her right leg. There was no hesitancy of micturation and defecation. By the time of admission she was unable to stand or walk.

On examination she was found to be well nourished and in excellent general health. Her vital sings were normal. She was fully alert and conversant. Cranial nerves and sensorimotor functions in the arms were entirely normal. There was marked weakness of both lower limbs, more on the right side and with right foot drop. Deep tendon reflexes were hyperactive on both knee and ankle jerks with right ankle clonus. Impairment to both pin and touch was apparent below T_{12} on the right and below the groin on the left. The anal sphinctor tone was normal. Systemic examinations revealed unremarkable findings.

Laboratory data on admission included urine and feces exams which were normal. The white blood cell count was 10,500 (polymorphs 53%, lymphocytes 34%, monocytes 10%, eosinophils 3%) and the hematocrit 36. Serology for HIV was negative. Her chest X-ray and EKG were normal.

A magnetic resonance imaging (MRI) scan of the thoracic and lumbar regions was performed. At the level of T_{10} - T_{11} a low density lesion about 5 mm. in diameter was noted. The provisional diagnosis was intramedullary tumor (Fig 1.).

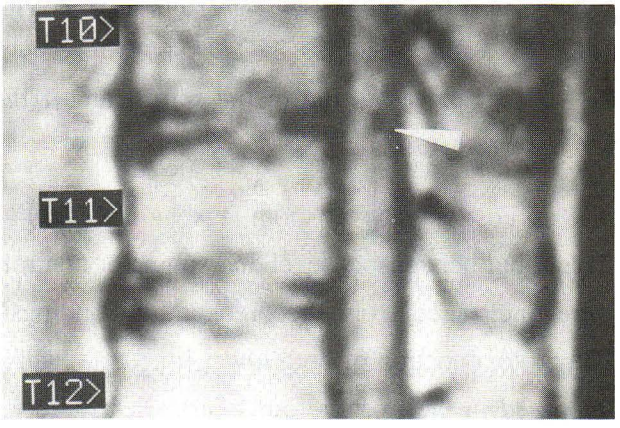

Figure 1. Intramedullary cystic lesion at T_{10} - T_{11} (arrow head) in MR imaging.

On 16 July, 1993 a laminectomy of T_9, T_{10} and T_{11} was performed. The dura appeared normal and pulsated. The spinal cord at T_{10} - T_{11} was slightly tense. A one cm. long midline myelotomy was done and a whitish cyst about 7 mm. in diameter found and totally removed. The contents of the cyst was clear fluid, and a whitish spot approximately 2 mm. in diameter was attached to one side of the cyst wall. The

cyst wall was a transparent membrane. The cavity left in the cord after removal of the cyst had a smooth wall. Histopathology proved it to be a cysticercus cyst (Fig. 2). A search was then made for other cysticercus cysts, muscle and skull radiograms and CT scan of the brain were negative.

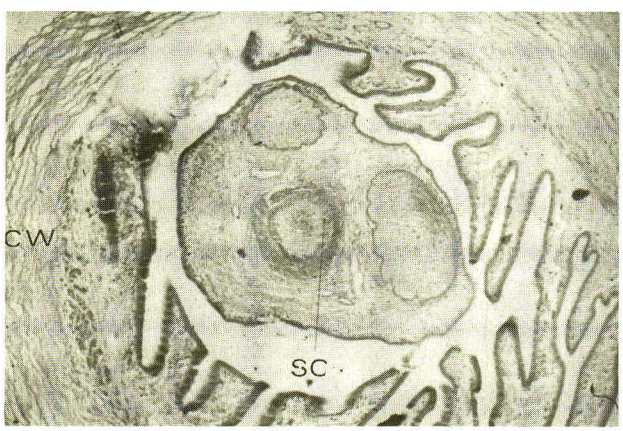

Figure 2. Photomicrograph showing three of the four suckers (SC), cyst wall (CW), and a portion of the spiral canal, HE x 45.

Post operatively, both legs were markedly flaccid but her sensory level and sphinctor control were unchanged from the preoperative findings. The motor function of both lower limbs gradually improved in the twenty-four hours after surgery, starting with the left leg and then the right. Her course in the hospital was uncomplicated. She was discharged on the seventh day after the operation and was able to ambulate with the assistance of a cane. The patient's right foot drop was improved. Decreased sensation to pin and touch were still present on the lateral aspect of the right leg and foot.

When examined two months later she had complete recovery of the motor and sensory functions of her left leg but the right side had numbness on the lateral aspect of the right leg and foot and slight weakness on dorsiflexion of the right foot. The patient resumed her normal activities.

Discussion

In clinical practice, the spinal forms of cysticercosis are rarely seen.[1-12] Queiroz, et al. indicated that the incidence of cysticercosis involving the spinal structure was only 5.85% in his review of the literature.[3] Hernandez-Absalon reviewed literature reports of 1632 patients with cysticercosis of the brain and found only six patients with a spinal form of cysticercosis.[6] The incidence in clinical series is expected to have a lower rate because spinal cysticercosis may mimic almost any other spinal disease and the severity of concurrent cerebral infestation.[3,4,7] Canelas and co-workers found 8 cases of spinal involvement among 296 patients with neurocysticercosis. This is a 2.7% incidence.[4]

The spinal forms have been classified anatomically and pathophysiologically as shown in table 1.[1-7,13] The leptomeningeal forms or growth in the subarachnoid space are more common than the intramedullary form by about six to eight times.[3,4] Only one subpial and two epidural cases have been reported.[1,3,5]

Table 1. Classification of spinal cysticercosis.

A. Anatomic classification
1. Intramedullary cysticercosis
2. Extramedullary cysticercosis
a. Leptomeningeal form
b. Epidural form
B. Pathophysiologic classification
1. Primary spinal cysticercosis
a. Isolated spinal infestation
b. Spinal infestation in the case of multiple cysticercosis
2. Secondary spinal cysticercosis
a. Direct spinal extension of massive intracranial cysticercosis
b. Cervical pachymeningitis with cord degeneration accompanying posterior-fossa cysticercosis

To our knowledge, only 30 cases of intramedullary cysticercosis have been recorded, including the present case (Table 2). Twenty-one cases were indentified at operation and the other nine were indentified at postmortem.[4,5] Most of these intramedullary cysts were found in the thoracic region.

Trelles and Trelles pointed out that the leptomeningeal form occurs primarily in the cervical region and is the result of propagation along subarachnoid pathways from racemose cysticercosis of the basal cysterns.[3,4,14] Most authors belived that hematogenous dissemination of the oncosphere produced the intramedullary cysticercosis. Queiroz. et al. reported that the spinal cord blood flow is roughly 100 times less than that of the brain and peculiarities of the cord tissue are contributory factors for the rarity of intramedullary cysts.[1,3-5]

In cases of spinal cysticercosis, the neurological symptoms and signs might appear to be due to three possible mechanisms :[1,3,7]

1. Inflamatory reaction caused by the metabolites of the parasite or the degenerated larval remains.

2. A mass effect of intramedullary or extramedullary cysts.

3. Degeneration due to pachyleptomeningitis or vascular insufficiency.[7]

Almost all reported cases were seen to have the signs and symptoms of progressive cord compression. Outstanding symptoms were progressive spastic paraplegia, foot drop, sensory disturbance and urologic disturbance. Venkataramana, et al. reported a case of intramedullary cysticercosis in which there was exaggeration of the symptoms and neurological deficits during pregnancy but which subsided after delivery.[11]

Diagnosis of intramedullary cysticercosis is very difficult prior to operation. The clue to the diagnosis of parasitic disease in our case was eosinophilia, but this is an inconsistant finding in

Table 2. Publishes cases of intramedullary cysticercosis.

Case	Author	Year	Sex	Age	Country	Site of cysticercosis
1.	Walton	1881	?	?	U.S.A	C_3 - C_4
2.	Pichler	1900	?	?	-	T_{11}
3.	Walbraun	1917	?	?	-	Low cervical
4.	Knapp	1919	?	?	U.S.A	T_5 - T_6
5.	Kimpton	1920	?	?	U.S.A	-
6.	Salles	1934	M	12	Brazil	Mid thoracic
7.	Barini	1954	M	56	Brazil	T_{10} - T_{11}
8.	Lima Costa	1957	?	?	Brazil	-
9.	Trelles	1958	F	32	Peru	C_3-C_4, L_4
10.	Rocca	1959	M	39	Peru	T_7
11.	Cabieses case 1	1959	M	42	Peru	T_4
12.	Cabieses case 3	1959	M	58	Peru	C_5 - C_6
13.	Dixon	1961	?	?	England	Cervical
14.	Granes	1963	M	28	Brazil	T_6
15.	Figueiredo	1963	M	20	Brazil	Sacral cord (T_{12})
16.	Portugal	1964	M	40	Brazil	C_7
17.	Hesketh	1965	M	22	Singapore	Upper dorsal (T_3)
18.	Testa	1965	M	40	Italy	T_8
19.	Singh	1966	M	40	India	T_7 - T_8
20.	Trelles (cases 2)	1970	M	31	Peru	T_9
21.	Metha	1971	M	23	India	T_{11}
22.	Antoniuk	1974	F	-	Brazil	T_{10}
23.	Queiroz	1975	F	60	Brazil	T_8
24.	Natarajan	1976	M	22	India	T_4
25.	Garza-Mercado	1976	M	22	Mexico	Sacral cord (T_{12})
26.	Akiguchi	1979	M	24	Japan	Sacral cord (T_{12})
27.	Holtzman	1986	M	18	U.S.A.	T_4
28.	Venkataramana (Case 1)	1989	F	22	India	T_2
29.	Venkataramana (Case 2)	1989	M	45	India	C_4
30.	Present case	1995	F	48	Thailand	T_{10} - T_{11}

cysticercosis. Serologic studies, which sometimes may be helpful, were negative in our case.[1,13] Myelographics in the intramedullary forms are reported to be non-specific revealing partial or total block.[2,3] CT scans and MRI are helpful for locating the intramedullary cyst.

The only method of treatment employed so far in cases of intramedullary cysticercosis is surgical intervention. Holtzman, et al. in their review of the literature indicated that in 1920[4] A.R. Kimpton was the first to publish a report on removal of an intramedullary cysticercus cyst. They also found 20 other cases of intramedullary cysticercus cysts which were operated upon. The total operative mortality rate was 15% and lasting morbidity from the illness was 85%. Among these, 12 patients were reported to have recovered sufficiently to be able to walk with or without some form of assistance. This poor outcome was thought to be due to delays in surgery and the poor neurological status of the patients before surgery.[4,9,12] Most of the patients came to surgical attention only after a definite spastic paraplegia had been present for months to years. The revesibility of neurological deficits in such instances is impeded by the parenchymal gliosis resulting from chronic exposure to the toxic waste products of the larva.[1,4] Sometimes, gliosis is associated with inflammatory granulation tissue around the cyst.[11] Therefore, even if mechanical decompression is achieved, complete restoration of the spinal cord function may not be expected to occur.[4,9] The mechanical distortion of the cord plays a significant pathophysiologic role and this is demonstrated by the clinical recoveries observed after removal of the cysticercus cyst.[4]

Isoquinolinepyrazine derivative Praziquantel has been used to treat cerebral cysticercosis, its use for spinal cysticercosis has not been reported yet. This is clearly due to the fact that a definite diagnosis in such cases can be made only after surgery. As seen in our case, a near complete recovery of neurological dificit was a significant result of early surgical removal of the cyst. The cysticercus cyst should be removable in toto without rupturing during surgery because the cyst is well encapsulated and non-adherent to cord tissue. In endemic areas of cysticercosis, cysticercus cysts should be considered an important differential diagnosis in all cases of cord compression.[11]

Although there have been many reports of neurocysticercosis in Thailand, none have reported intramedullary cysticercosis. This is the first case report of intramedullary cysticercosis in Thailand and the thirtieth case in the world literature.

I wish to thank Dr.Winai Tumweerapong who was responsible for the pathological study.

References

1. Akiguchi I, Fujiwara T, Matsuyama H, Muranaka H, Kameyama M, Intramedullary spinal cysticercosis. Neurology 1979 Nov; 29(11): 1531-4
2. Kim KS, Weinberg PE. Spinal cysticercosis. Surg Neurol 1985 Jul; 24(1):80-2
3. Queiroz L de S, Filho AP, Callegaro D, Faria LLD. Intramedullary Cysticercosis. Case report, literature review and comments on pathogenesis. J Neurol Sci 1975; 26(1): 61-70
4. Holtzman RNN, Hughes JEO, Sachdev RX, Jarenwattananon A. Intramedullary cys-

ticersis. Surg Neurol 1986 Aug; 26(2): 187-91

5. Hesketh KT. Cysticercosis of the dorsal cord. J Neurol Neurosurg Psychiatry 1965 Oct; 28(5):445-8
6. Garza-Mercado R. Intramedullary cysticercosis. Surg Neurol 1976 Jun; 5(6):331-2
7. Kahn P. Cysticercosis of the central nervous system with amyotrophic lateral sclerosis : case report and review of the literature. J Neurol Neurosurg Psychiatry 1972 Feb; 35(1):81-7
8. Cabieses F, Vallenas M, Landa R. Cysticercosis of the spinal cord. J Neurosurgery 1959 May; 16(3):337-41
9. Venkataramana NK, Jain VK, Das BS, Rao TV. Intramedullary cysticercosis. Clin Neurol Neurosurg 1989; 91(4):337-41
10. Roy RN, Bhattacharya MB, Chartterjee BP, Pal NC. Spinal cysticercosis. Surg Neurol 1976 Aug; 6(2):129-31
11. Natarajan M, Ramasubramanian KR, Muthu AK. Intramedullary cysticercosis of the spinal cord. Surg Neurol 1976 Sep; 6(3): 157-8
12. Singh A, Aggarwal ND, Malhotra KC, Puri DS. Spinal cysticercosis with paraplegia. Br Med J 1966 Sep 17; 2(5515):684-5
13. Firemark HM. Spinal cysticercosis. Arch Neurol 1978 Apr; 35(4):250-1
14. Trelles JO, Trelles L. Cysticercosis of the nervous system. In : Vinken PJ, Bruyn GW, eds. Handbook of Clinical Neurology. Vol 35. Amsterdam : North-Holland Publishing, 1978: 291-320