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INSIGHTS INTO CERVICAL INTRAMEDULLARY NEUROCYSTICERCOSIS: A RARE PRESENTATION

Neurosurgery

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KEYWORDS

INTRODUCTION

Cysticercosis is the most common parasitic disease of the central nervous system.The parasitic involvement of the spinal cord and its nerve roots is relatively rare and occurs in only 0.7 to 5.85% of patients with NCC(1).Subarachnoid location associated with CNS or systemic involvement is the common presentation. Isolated spinal intramedullary location without any systemic involvement is a highly rare presentation.

Case Report

A 35 yrs old female presented with insidious onset, diffuse, ill defined burning pain over trunk and all four extremities with frequency and urgency of micturition for eight months followed by spastic quadriparesis for five months. On examination, higher mental function and cranial nerves examination were normal. Power was 3/5 in upper limbs and 4/5 in lower limbs with spasticity in all the limbs. There was bilateral atrophy of supraspinatus, infraspinatus, deltoid and biceps, with no sensory deficits. All the deep tendon reflexes were exaggerated with presence of ankle clonus. Superficial reflexes were absent and plantar reflex was extensor bilaterally. Rest of the examination was normal.

On imaging C4-C6, intramedullary space occupying lesion was detected which was hypo intense on T1WI and hyper intense on T2WI. On contrast administration, ring enhancement was present [Fig. 1].Routine investigations were within normal limits.

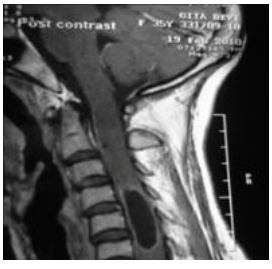


Fig. 1 : Pre op MRI

Treatment : Following GA, in prone position, C4-C6 laminectomy was performed. Dura was tense and non-pulsatile. After opening the dura, a midline myelotomy was made with the help of operating microscope and a total removal of the SOL was achieved.Per operatively, the tumor was cystic, white, glistening, smooth walled SOL of around 2 cm length and 1 cm diameter [Fig. 2]. Wound was closed in layers. Postoperative period was uneventful.

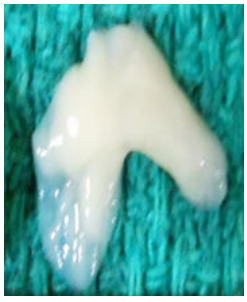


Fig. 2 : The specimen of cystic, white, glistening, smooth walled SOL

Histopathology of the lesion confirmed it to be the neurocysticercosis [Fig. 3].An intracranial and systemic lesion was searched for but not detected. Patient was discharged on the tenth postoperative day with marked neurologic improvement. Post operative MRI showed no residuum of the lesion [Fig. 4].

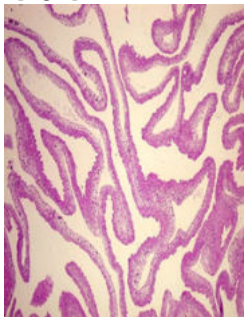


Fig. 4 : Histopathology slide

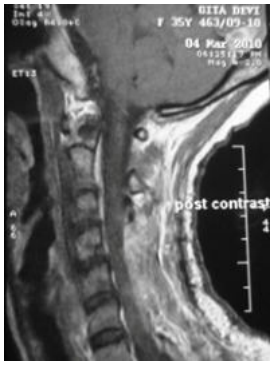
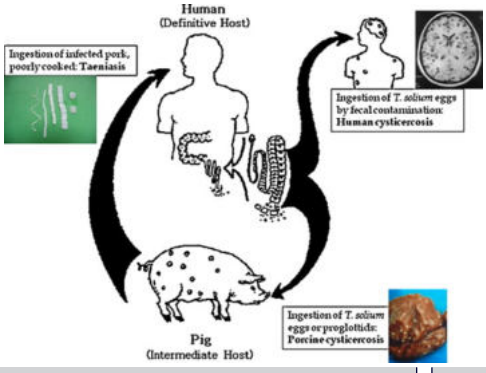


Fig. 4 : Post op MRI

DISCUSSION

Neurocysticercosis is caused by Cysticercuscellulosae, the metacestode state of Taenia solium. It results from direct ingestion of cysticercal eggs contaminated by human or porcine faeces.



It is an endemic condition to Brazil, Peru, Mexico, Korea, India, South America, Tropical African, and Southeast Asian countries.Spinal NCC can be extraspinal or intraspinal.The parasitic involvement of the spinal cord and its nerve roots is relatively rare. The newer studies point to decreasing incidence of NCC with improving medical facilities and better hygiene practices. Global incidences are now in range of 0.7 to 3%,(2) It is usually associated with cerebral cysticercosis, but may also present as an isolated lesion. Subarachnoid location of spinal cysticercosis occurs most frequently in approximately 80% of cases(3). Spinal NCC can be divided in extradural, intradural subarachnoid (leptomeningeal), and intramedullary (IM) (parenchymal) forms. The thoracic spine is most commonly involved. Spinal distribution of cysticerci occurred as follows: 34% in the cervical; 44.5% in the thoracic; 15.5% in the lumbar; and 6% in the sacral region [2]. . Among these, IM presentation is usually the rarest. And likewise, Dhar et al, in 2021(5) , published a case report with review of literature which revealed only 33 articles (case reports and series) pertaining to the IM-NCC, the earliest being published in 1996.It translates to three articles being published from 1996–2000, followed by nine articles being in print from 2001–2010, and finally 21 articles from 2011–2020. However, among them ,there were only 6 cases of isolated cervical intramedullary NCC .2 case reports have been published from India in 2017, 2 from USA, 1 each from Brazil and Gautemala. Ours case could be 3rd from Indian subcontinent and possibly 7th overall(**table 1**). Hence, increase in data availability will help to make more evidence-based treatment guidelines possible.

Table 1. showing the relevant isolated cervical intramedullary NCC cases published(pubmed indexed) till date.*[adapted from study by Dhar et al, 2021(5)]*

sl. No.	Authors	Country	year	age(yr)	gender	s.c level	symptoms
1	Jobanputra et al (6)	USA	2020	44	F	C5-C6	Motor
2	Yadav et al (7)	INDIA	2017	8	M	C5-C6	Motor,Pain
3	Ranja (8)	INDIA	2017	6	M	C4-C6	Pain
4	Salazar Noguera (9)	GAUTEMALA	2015	43	M	C7	Motor
5	Lambertucci (10)	BRAZIL	2011	23	M	C3-C5	Motor
6	Sheehan (11)	USA	2002	16	F	C1-C2	Motor

Route of dissemination for subarachnoid location is mainly CSF, while for intramedullary location, it is mainly hematogenous.Presentation depends on the location and size of the lesion, and the inflammatory response or arachnoid scarring generated by cyst breakdown. The important differential diagnoses are arachnoid cysts, dermoid cysts, hydatid cysts, tuberculosis, sarcoidosis and forms of subarachnoid neoplasia, demyelination and vascular pathologies.

Serology with the enzyme-linked immunoelectrotransfer blot assay (EIBT) in the serum and CSF provide relatively reliable result in the endemic areas and is 100% specific and 98% sensitive.Cerebrospinal fluid (CSF) findings typically include moderate lymphocytic pleocytosis, variable eosinophilic pleocytosis, elevated protein and low/normal glucose levels.

The patients with severe neurologic symptoms caused by spinal cysticercosis are the best candidates for surgery, as it alleviates the compression and provides tissue for confirmation of diagnosis. Mortality rate [15%] and morbidity rate [85%] are high among surgical candidates(12). Postoperative anticysticercal treatment is generally recommended because cysticercosis is considered to be a generalized disease with focal symptoms. Albendazole (15 mg/kg/day) and praziquantel (50 mg/kg/day) regimens have been used for both 8- and 30-day periods. Steroids must be added to the antihelminthic regimen because the inflammatory response around the cyst after treatment may deteriorate the spinal cord functions.

The therapeutic options for spinal NCC depend on the neurological status. Due to the limited size of the spinal canal, the mass effect and cord oedema from the cyst are not well tolerated and clinically the consequences could be devastating. Medical therapy is considered in patients with stable and non-progressive symptoms when the diagnosis is made by CSF, serum immunoelectric antibody assay, or concomitant intracranial lesions. However, severe symptoms, progressive deterioration, missed or uncertain diagnosis, and failed medical therapy should be treated with surgery which not only helps confirm the diagnosis but also prevents permanent damage.

CONCLUSION

Solitary intraspinal NCC at intramedullary location is among the most rare lesions , more so in cervical spine . Possibility of this lesion should always be kept in mind in a SOL of spinal cord in a patient from the endemic regions in order to make a correct choice between operative and conservative management. Medical management may be advocated by some, but in cases like this where neurological deficits occur, surgery is the mainstay of treatment followed by medical therapy to prevent recurrence.

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