Section: Neurology

Case Report

DISSEMINATED HYDATIDOSIS AND TRANSVERSE MYELITIS- AN UNDER RECOGNISED ETIOLOGY

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Abstract

Transverse myelitis is characterized by acute or subacute spinal cord dysfunction resulting in paresis, sensory loss with definite sensory level, and autonomic impairment below the level of the lesion. Etiologies can be broadly classified as Infectious, Para infectious, systemic autoimmune disorders, acquired demyelinating diseases, and drug/toxin induced. We present a case of acute onset symmetric paraparesis with definite sensory level along with bladder involvement but without any cranial nerve involvement. MRI spine revealed longitudinally extending acute transverse myelitis. Further evaluation for autoimmune causes was unremarkable. Imaging revealed multiple hydatid cysts in the liver as well as the lung. The patient's symptoms improved remarkably following the initiation of steroid anti-parasitic therapy. Parasitic infections including Schistosoma and toxoplasma are reported in patients with transverse myelitis. Here we are presenting a unique case of acute transverse myelitis with disseminated echinococcosis.

INTRODUCTION

Transverse myelitis refers to a pattern of extensive spinal cord injury, clinically manifesting as acute or subacute onset bilateral symmetric sensory and motor impairment. Patients usually have Upper motor neuron (UMN) signs below the level of the lesion and Lower motor neuron (LMN) signs at the level of the lesion with a clear sensory level. However, patients who may present in spinal shock can have predominantly LMN signs on examination. Contrast-enhanced MRI is one of the most important imaging modalities in the case of acute transverse myelitis.[1] The spinal cord is T2 hyperintense with variable contrast enhancement. Symptoms such as nonspecific fever and constitutional features are seen commonly preceding the development of neurological symptoms.[1] Transverse myelitis can be a presenting complaint in cases of autoimmune disorders of the CNS such as NMO, ADEM, and multiple sclerosis. Many postinfectious cases of acute transverse myelitis can be recognized in practice. Infectious causes include mycoplasma, Herpes simplex Virus, HBV, HCV, HIV, Mycobacteria, Spirochete, and Rickettsial infection.

Echinococcus granulosus, also known as dog tapeworm is the causative agent of hydatid cysts. The definitive host for this parasite is a dog and the intermediate host is sheep. A human being is the aberrant intermediate host and is infected by ingesting eggs in contaminated food, water, and soil or after direct contact with animals. The liver is the most common site for hydatid disease (75% of cases), followed by the lungs (15%), spleen (5%), and other organs such as muscle, bone, brain, and rarely spinal hydatid cyst.[2] The diagnosis of hydatid disease is based on the epidemiological background of patients, clinical features, or noninvasive screening procedures. Chest and abdominal X-ray, ultrasonography, and CT scan should identify cysts characteristics and the diagnosis is confirmed by detecting specific antibodies -an immunodiagnostic test.[2,3] Complications of echinococcosis are usually local due to either rupture or mass effects. However acute transverse myelitis with Echinococcus infection has not been reported in literature yet, hence we present a case of transverse myelitis related to disseminated hydatidosis.

CASE REPORT

A middle-aged male, resident of North India, driver by occupation, with no previous comorbidities or history of substance abuse presented with complaints of low to moderate grade, intermittent fever for two weeks duration, which was not associated with chills and rigor, diurnal variation, rash, cough, shortness of breath, chest pain, loose stool, abdomen pain. Two days after the onset of fever he developed gradual onset progressive tingling sensation in bilateral upper and lower limbs followed by band-like sensation around the chest at the level of the nipples and

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numbness over the abdomen and bilateral lower limbs. He also experienced burning sensation and alteration in hot and cold sensation in both upper and lower limbs. A few hours later he was unable to void urine despite the sensation of fullness and there was dribbling of a small volume of urine with effort. On the same day, this was followed by sudden onset progressive symmetric weakness of both proximal and distal muscle of bilateral lower limbs. There was no history of trauma, back pain, prior vaccination, blurring of vision, diplopia, dysphagia, or deviation of angle of mouth. There was no history of similar complaints in the past. There was no history suggestive of systemic autoimmune disorder like no history of dryness of mouth and eyes, Raynaud's phenomenon, oral ulcers, rash, photosensitivity, arthritis.

On examination, he was hemodynamically stable, with normal cardiovascular and respiratory examination. There were no palpable lymph nodes. He had a temperature of 101.3-degree F. Neurologic examination revealed a normal higher mental function, cranial nerve examination and deep tendon reflexes in upper limb with no spine deformity or spinal tenderness. Motor examination revealed hypotonia in bilateral lower limbs with a power of 2/5. Sensory examination showed hypesthesia to touch below the level of the nipple. The upper limb power was normal, and planters were bilaterally mute. There were no clinical signs of meningeal irritation.

Routine hematologic testing showed an elevated total leukocyte counts of 21,780 cells/mm3 and platelets within normal limits. Liver and kidney function tests were also within normal limits. Fever workup for tropical diseases and routine cultures were negative. His cerebrospinal fluid (CSF) analysis showed elevated protein of 102mg/dl, sugar 73mg/dl with 10 cells/ul, and 50% monomorphs. The gram stain of the fluid did not reveal any organisms and culture was sterile. A contrast enhanced MRI brain and whole spine was obtained which was suggestive of longitudinally extensive transverse myelitis in the cervical and dorsal cord extending from C2 to D9 vertebral level with normal brain imaging. [Figure 1] In view of the MRI findings a suspicion of autoimmune disorders was kept and further work up was done including CSF for Aquaporin 4, Anti MOG, and antinuclear antibodies which came to be negative.

A chest x ray was obtained which revealed a solitary round lesion in the left lung. [Figure 2] and USG abdomen showed hepatomegaly with 2 cystic lesions in the liver with few internal septations and daughter cysts inside. [Figure 3] Further a contrast enhanced CT thorax and abdomen was done which confirmed a well-defined non-enhancing cystic lesion of size 9.3x5.5x9.3 cm in the subhepatic region and lesion of 7x7.4x7.6 cm in seg VI and VII of the right lobe of the liver. [Figure 4] Another well-defined fluid field cystic lesion with multiple non-enhancing septa was noticed in the left upper lobe of the lung. [Figure 5] Hydatid cyst serology IgG was positive (Echinococcus IgG, serum- 1.89 (positive); Bio ref level <0.9). Review of history revealed the patient had pet dogs which could have been a likely source of hydatid infection.

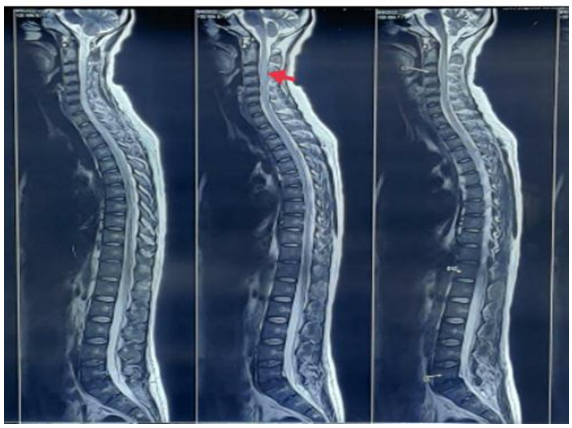

Figure 1: MRI study on presentation: T2 MR sequence of sagittal section of spinal cord showing long segment T2 hyperintense intramedullary signal in cervical and dorsal cord extending from C2 to D9 vertebral level involving central part of cord as shown by arrow likely: showing demyelination or longitudinally extensive transverse myelitis.

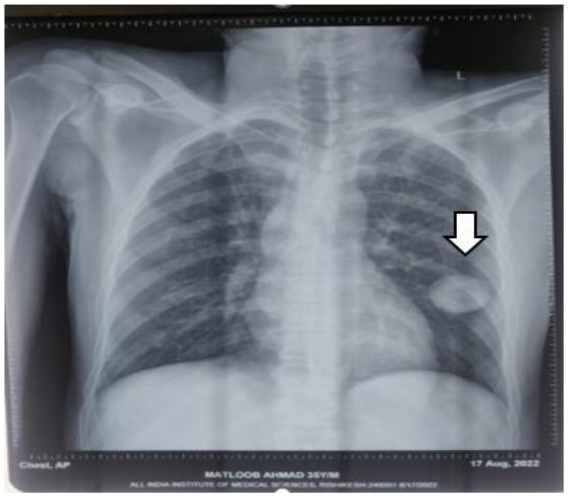

Figure 2: Chest X-ray PA view erect showing rounded opacity in left lung field depicted by arrow.

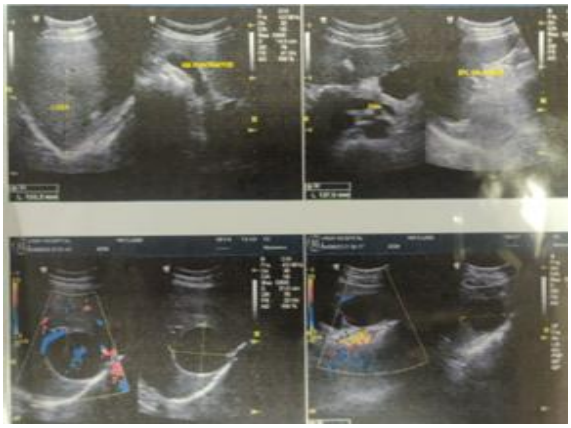

Figure 3: On USG correlation it was showing few internal septations and daughter cyst within.

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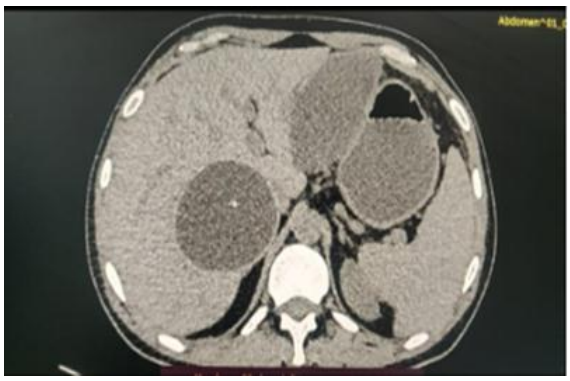

Figure 4: CECT abdomen showing enlarged liver of size 16.3 cm. Well-defined non-enhancing cystic lesion 7x7.4x7.6 cm in seg VI and VII without any septations. Another well-defined cystic lesion 9.3x5.5x9.3 cm is seen in the left subhepatic space.

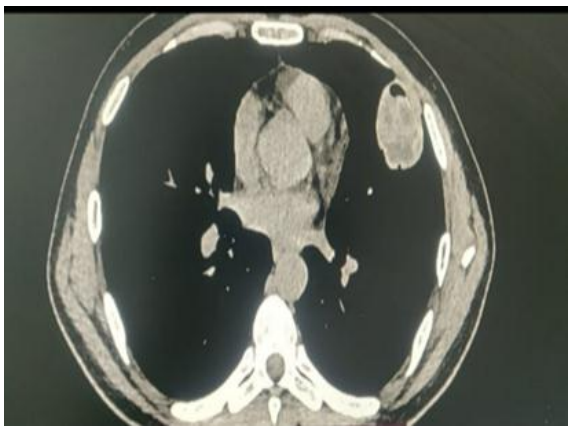

Figure 5: CECT chest showing the well-defined multiloculated cystic lesion in the left upper lobe of the lung likely ruptured hydatid.

Table 1: Represents power in hip, knee, Ankle, shoulder, elbow, wrist joints on presentation, on discharge (after 15 days) and follow up (after 30 days)

Power	Presentation	Discharge (after 15 days)	Follow Up (after 30 days)
Hip flexion and Extension	3/5	3/5	+4/5
Knee flexion and Extension	3/5	+4/5	5/5
Ankle planter flexion and dorsiflexion	-4/5	5/5	5/5
Shoulder flexion and extension	5/5	5/5	5/5
Elbow flexion and extension	5/5	5/5	5/5
Wrist flexion and extension	5/5	5/5	5/5

The patient was started on oral Albendazole therapy along with intravenous methylprednisolone pulse followed by oral steroids in tapering doses. The patient showed significant clinical improvement within 7 days and power improved with the 4 weeks of steroid therapy. [Table 1]

DISCUSSION

Patient can present with transverse myelitis due to numerous etiologies. The Longitudinally extensive transverse myelitis on MRI shows 3 or more contagious spinal segment involvement. It is characteristically seen in neuromyelitis optica. However, they are many more diseases including systemic lupus erythematosus that can cause similar clinical and radiological pictures.[4] Other demyelinating conditions include multiple sclerosis, postinfectious myelitis, sarcoidosis, etc. Among infections and postinfectious causes, viruses that are commonly responsible are herpes, retrovirus, hepatitis B and C, and Polioviruses. Others are bacterial, mycobacterial myelitis as well as parasitic infections which are reported previously are schistosomiasis, toxoplasmosis, and cysticercosis.[5] Myelitis from helminth is far less common than parenchymal and meningeal involvement.[6] In this case, we excluded the more common causes such as autoimmune causes of acute transverse myelitis. Oligoclonal bands are positive in serum as well as CSF, indicating a rise in the immunoglobulins level in CSF in response to systemic infection. Immunoglobulins against echinococcus are also positive in the serum which could be the likely cause of positive Oligoclonal bands (OCB). Matching OCB present in serum and CSF suggestive of systemic immune reaction. No case of transverse myelitis has been previously reported with Echinococcus infection. Echinococcus granulosus also called as dog tapeworm is the causative agent of hydatid cysts. The most common organ involved is the liver, and other organs are the lung, spleen, muscle, bone, brain rarely spinal hydatid cyst. A spinal hydatid cyst is a rare but well-known entity that presents as compressive myelopathy.[7]

CONCLUSION

The etiologies of Acute transverse Myelitis are protean and varied ranging from Infectious, Para infectious, systemic autoimmune disorders, acquired demyelinating diseases, and drug/toxin-induced. Here we present a unique case of acute transverse myelitis with disseminated echinococcosis which is a rare etiology. Recognising etiology and timely management can be helpful in clinical improvement of Acute Transverse Myelitis.

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