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Corticosteroid-Responsive Longitudinally Extensive Transverse Myelitis in a Pediatric Patient and Its Consequences: A Case Report

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Abstract

Transverse myelitis is as a rare focal inflammation across the spinal cord manifesting as motor, sensory, and autonomic symptoms. The term “longitudinally extensive” indicates the involvement of 3 or more segments in the spinal cord and is commonly associated with poorer prognosis. A 12-year-old boy came to the emergency room with the complaints of weakness on both lower extremities, radiating pain, and difficulty in controlling stool. The symptoms were said to have been persisting for 30 days. T2-weighted images of the spine magnetic resonance imaging revealed transverse myelitis from T10 to T12. The patient immediately received intravenous corticosteroid for 5 days and is later tapered off intravenously for 9 days and then orally for 14 days. A slow but steady improvement was observed with some residual deficits during discharge. The patient was readmitted 5 days later for bronchopneumonia and suspicion for a steroid-induced immunosuppression was raised. This case report focuses on the treatment of longitudinally extensive transverse myelitis in pediatric patient presenting with significant disability and delayed admission resulting in poor prognosis and concerning side effect.

Keywords: Case report, longitudinally extensive transverse myelitis, corticosteroid, pediatric

Introduction

Transverse myelitis (TM) in children is a rare condition with worldwide prevalence as small as 1–2 cases per million per year ^[1]. According to the length and location of the lesion in the spinal cord, TM is classified into 3 types which are acute partial TM (APTM), acute complete TM (ACTM), and longitudinally extensive TM (LETM). APTM and ACTM involves 1–2 segments of the spinal cord with asymmetrical and symmetrical dysfunction, respectively. On the other hand, LETM is characterized with a spinal cord lesion involving three or more vertebral segments ^[2]. LETM is also more commonly reported on pediatric patients (66–85%) ^[2]. Symptoms of TM include rapidly progressive deficits in motor, sensory, and autonomic nervous system with acute or subacute onset ^[3]. All of which corresponds to the length and location of the lesion in spinal cord. MRI scans provide valuable finding regarding the extent and cause of myelopathy. TM is characterized with hyperintensity in T2-weighted images with the lesion affecting 2/3 of the cord diameter with central distribution ^[4].

While empiric corticosteroids are used as first line therapy, there is no consensus regarding the treatment of TM to date. Standard regiment includes 30 mg/kg up to 1000 mg of intravenous methylprednisolone daily for 3–7 days followed by oral regiment with the dose of 1 mg/kg daily tapered off within 3 to 4 weeks. Despite its effectivity in the treatment of TM on young patients, corticosteroids have been known for its potential side effects including increased risk of infections. This is especially found in patient receiving a course of 2 mg/kg prednisolone given daily for 2 weeks ^[5]. This paper aims to report a case of a pediatric patient with longitudinally extensive transverse myelitis receiving corticosteroid therapy with subsequent bronchopneumonia as a suspected immunosuppressive side effect.

Case report

A 12-year-old boy came to the emergency room carried by his father with chief complaint of weakness in both legs for the past 30 days. Initially, the patient felt heaviness upon moving

his legs. The symptoms further worsen in the span of a week and the patient faced difficulty during walking, resulting in being able to move only side-to-side while dragging his feet and leaning into a wall for support. The patient could no longer perform daily activities and had been absent from school for 2 weeks. The patient also complained of radiating pain in both of his legs originating from the lower back with the pain level of 7 out of 10. The patient denied history of tingling and numbness. The patient also complained of difficulty in controlling stool for the past 7 days. All symptoms forementioned persisted until the patient came to the ER with no episode of improvement. History of abdominal pain, change in stool color or consistency, urinary symptoms, headache, seizure, and vision problems were denied. The patient had not received any other medication aside from paracetamol tablets given by a general physician 4 days before admission. The parents later revealed that the patient experienced flu, dry cough, and mild fever 7 days before the weakness. Family history of similar symptoms or neurological diseases were denied as well as history for allergy and trauma. The patient is a junior high school student. The parents mentioned a decline in the patient's academic performance due to his absence from school. The patient's father is an active smoker but admitted to never smoke indoors. Antenatal history includes no abnormality during regular ANC visit to a midwife for 9 months of gestation. The patient was delivered vaginally with a birth weight of 3050 grams and no congenital abnormalities reported. History of delays in reaching developmental milestones were also denied. The patient received complete essential immunization from the government's program at local public health center.

The patient was fully alert with stable vital signs but weak general appearance. During neurological examination, the patient was cooperating and oriented. Cranial nerve examination was normal. Motor examination of the upper extremities showed normal tone, power, and reflexes. Meanwhile, the lower extremities showed bilateral power of 3/5 and hyperactive deep tendon reflexes. Pathological reflex examination revealed positive Babinski, Chaddock, Schaeffer, and Oppenheim reflex on both lower limbs. No sensory level was identified. Complete blood count showed leukocytosis ($15.49 \times 10^9/L$) while other blood laboratory tests were normal. Thoracolumbal MRI scans revealed spinal cord swelling extending from T10 to T12 with slight hyperintensity in T2WI and contrast enhancement. No compression was observed and a conclusion of longitudinally extensive transverse myelitis (LETM) was reached (Fig 1).

Empiric intravenous methylprednisolone was immediately given with the dose of 1000 mg daily for 5 days followed by a period of taper until after the patient was discharged. The patient also received physical therapy as soon as the first day of hospitalization. The patient was released from the hospital on the 14th day of care with the mRS score of 3 – 4. The patient received outpatient treatment that includes oral methylprednisolone and weekly monitoring. Gradual improvements in sensory, motor, and autonomic function were observed. However, residual deficit in muscle strength persists and are refraining the patient from regular daily activities.

The patient was later readmitted to our hospital 5 days after being home. The patient suffered from fever, productive cough, and dyspnea for the past 2 days. It was later revealed

that the caretaker of the patient was experiencing the same symptoms 2 days before the patient. Physical examination revealed chest retraction with rales breathing sound. Complete blood count revealed neutrophilic leukocytosis and chest X-ray was positive for bronchopneumonia (Fig 2). The patient was given intravenous cephalosporin for 5 days with ongoing corticosteroid regiment maintained. The symptoms subsided at the end of the antibiotic course and the patient was sent home.

Discussion

The treatment of TM requires immediate corticosteroid administration without any delay, even because of waiting for test results. Delay in treatment of TM is associated with partial recovery, worsening progressivity, and risk of respiratory decompensation resulting in admission to the ICU. A study in Turkey reveals that the average time from onset to admission that is associated with better functional outcomes in pediatric patients is as soon as 4 days [3]. Nevertheless, delay in treatment of TM remains a considerable problem yet to conquer. The broad differential diagnosis and possible overlap with non-inflammatory myelopathies often withhold physician to start therapy. Two of the most prevalent causes are late diagnosis and misdiagnosis, which are common in acute and subacute myelopathies. At initial presentation, TM can often be misdiagnosed as Guillain-Barre Syndrome (GBS), stroke, systemic infection, syring, vitamin deficiencies, and psychiatric disorders. A case report in India presented a 13-year-old patient that was only diagnosed with LETM after being treated in other health facility for 20 days. The patient presented with fever, difficulty in walking, and loss of bowel and bladder sensation. The patient received pulse injection of methylprednisolone for 5 days and 2 doses of intravenous immunoglobulin on the second and third day of admission. Despite the therapy, there was no significant improvement reported and therefore the patient was eventually discharged after 1 month of hospitalization with persisting paraparesis and impaired bowel and bladder function even after 1 year of discharge [6].

Another cause worth to notice is the low awareness for TM in the community, mainly due to its rarity, causing patients to neglect their symptoms. A case report from India presented a 14-year-old patient complaining of weakness on both upper limbs for 4 days. Unfortunately, the patient was only brought to the hospital 1 month later when the weakness already spread to the lower limbs causing the patient unable to walk. The patient was immediately started on pulse injection methylprednisolone and responded well to the therapy. However, the patient suffered from persisting paresthesia on both upper limbs last reported on 8 weeks clinical follow-up [7].

Our patient was brought to the hospital 30 days after the onset of the weakness. The patient's presentation also checked all 3 predictors for severe residual disability in TM, including: delayed corticosteroid treatment, significant disability at presentation, and extensive spinal cord involvement [2]. LETM specifically was also associated with poorer prognosis and higher chance of relapse. Nevertheless, aggressive treatment with corticosteroid as well as physical therapy was given to our patient and despite the slow improvements, no worsening progressivity was observed. The patient in this report received a total of 14 days of intravenous corticosteroid continued with another 14

days orally. The patient reported the pain reduced around day 5 of treatment. However, motor improvements had just been apparent in the seventh day in which the patient started to be able to sit and walk independently for a limited distance. Around the same period of time, symptoms related to fecal incontinence also began to improve. The recovery was considered slow, but not in a refractory manner in which plasmapheresis is suggested to be supplemented. Therefore, the corticosteroid therapy was maintained along with watchful monitoring. The patient was able to walk 2 meters independently at the ninth day of treatment and later 5 meters in the eleventh day. On the fourteenth day of treatment, symptoms regarding sensory and autonomic system were mostly healed and the weakness was a lot less conspicuous. The patient regained his ability to perform daily activities with assistance in some aspects due to residual disability.

Side effects associated with corticosteroids remain a constant source of concern. The immunosuppressive nature of steroid is needed in treating autoimmunity in TM but is also causing the patient to be more susceptible to infection. The immunosuppressive property of corticosteroids is dependent to dosage, duration, and timing within the phases of inflammation [8]. Administration of oral corticosteroid (OCS) in high doses as much as an equivalent of 10 mg prednisone daily is associated with increased risk of infectious complications, ranging from hospitalizations due to pneumonia to bacterial sepsis [9]. Corticosteroids inhibit

neutrophil adhesion and activation while at the same time reducing lymphocytes, eosinophils, basophils, and macrophages count [10]. While some of these findings can be observed in our patients' complete blood count (CBC) during readmission for bronchopneumonia 5 days later, it remains unclear whether it is solely due to corticosteroid therapy or aggravated by bacterial infection (Table 1). Nevertheless, it is advised for pediatrician to be more vigilant through the monitoring of clinical signs and CBC. Another important measure to approach is to also educate the patient's caregiver regarding the regimen and its possible side effects. This is due to their outlook being a factor in directing treatment and determining compliance [5].

Conclusion

Longitudinally extensive transverse myelitis (LETM) is a rare disease affecting motor, sensory, and autonomic nervous system. Its rarity plays a part in low awareness of its existence in the community resulting in late admission. Delayed treatment of LETM is associated with significant residual disability. Watchful monitoring has to be maintained during the course of corticosteroid therapy in case of possible side effects.

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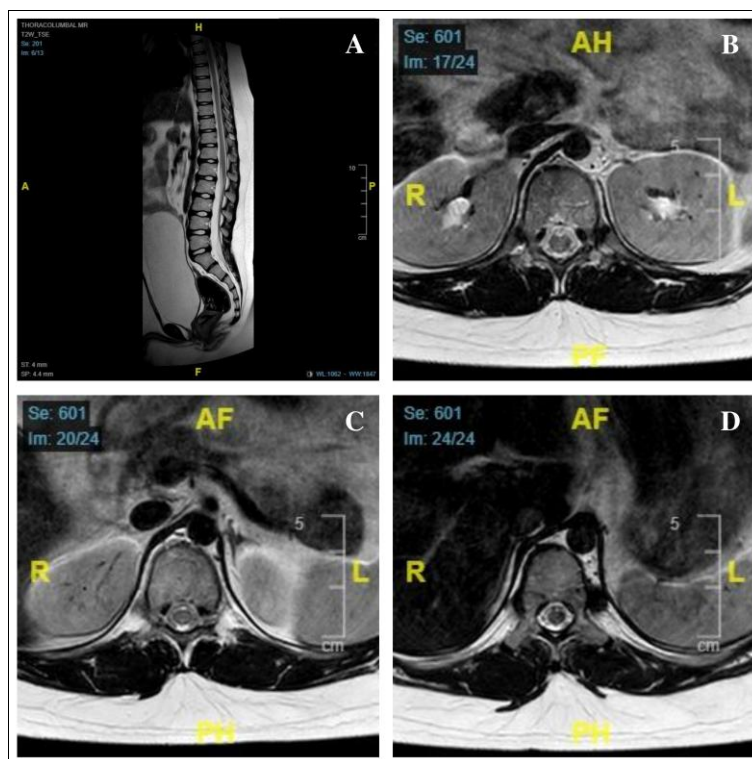


Fig 1: (A) Spinal cord swelling and slight hyperintensity in sagittal T2WI thoracolumbal spine MRI with axial view of T10 (B), T11 (C) and T12 (D)

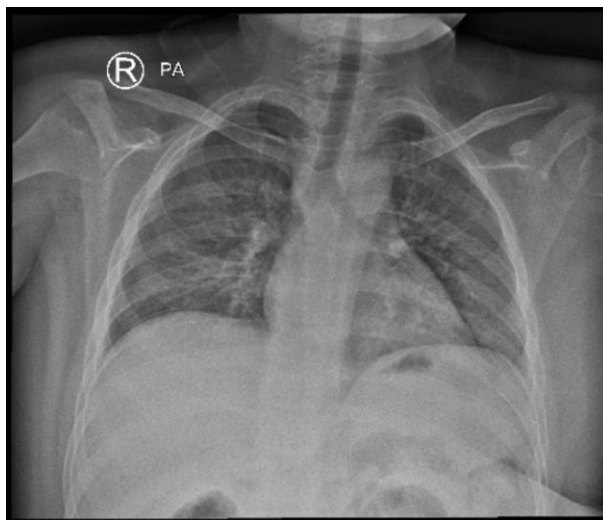


Fig 2: CXR showing infiltrate in right paracardial with increased bronchovascular pattern

Table 1: White blood cell count on second admission for bronchopneumonia

Parameter	Laboratory value	Unit	Reference range
Leukocyte	21.04	$10^9/L$	4.0–13.0
Eosinophil	0.00	$10^9/L$	0.00–0.81
Basophil	0.02	$10^9/L$	0.00–0.21
Neutrophil	16.79	$10^9/L$	1.50–8.00
Lymphocyte	0.83	$10^9/L$	1.10–4.50
Monocyte	1.00	$10^9/L$	0.2–1.00

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