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Review article

# Fibrocartilaginous Embolism of the Spinal Cord in Children: A Case Report and Review of Literature

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**ABSTRACT**

Fibrocartilaginous embolism (FCE) is assumed to be caused by the migration of fibrocartilaginous nucleus pulposus components through retrograde embolization to the spinal cord artery. FCE is currently not well recognized among pediatricians due to its rarity. We present the case of a previously healthy 15-year-old soccer player who, after kicking a ball, developed progressive weakness in both legs and ischuria the next day. Magnetic resonance imaging revealed T2 hyperintensity in the anterior horn of the spinal cord at the Th12/L1 level with Schmorl's node at the level of L1/2. We also reviewed previous literature of FCE of the spinal cord in children (<18 years of age), and a total of 25 pediatric patients, including our case, were reviewed. The median age was 14 years and 64% of the reviewed patients were female. The most common trigger event was intense exercise or sports (52%). The neurological symptoms started within 1 day in most cases and time to symptom peak varied from a few hours to 2 weeks. The most common initial neurologic symptoms were weakness or plegia (100%), followed by paresthesia or numbness (44%). Affected areas of the spinal cord were distributed evenly from the cervical to thoracolumbar regions. Although steroids and anticoagulants were most commonly used, the prognosis was quite poor (mostly mild to severe sequelae and 3 patients died). Although FCE is a very rare condition, pediatricians should be aware of the characteristics and include FCE of the spinal cord in their differential diagnosis, especially for physically active patients.

**Keywords:** clinically isolated syndrome; infarction; ischuria; transverse myelitis; multiple sclerosis; myelopathy; plegia

## Introduction

Fibrocartilaginous embolism (FCE) of the spinal cord is a rare and underrecognized infarction. It is characterized by ischemic myelopathy, which is assumed to be caused by the migration of fibrocartilaginous nucleus pulposus components through retrograde embolization to the spinal artery[1]. Symptoms include rapidly progressive plegia, paresthesia, and bladder or bowel dysfunction typically following back or neck pain[2].

FCE is well described in veterinary literature; recently, it has also been increasingly recognized in humans[3-7]. However, it is currently not well recognized among pediatricians due to its rarity. Recognizing the clinical features that distinguish FCE from other causes of spinal cord disorders will be helpful in the proper management of these pediatric patients.

We present a case of a 15-year-old soccer player with ischemic myelopathy secondary to suspected FCE based on clinical and radiological findings. In addition, we also reviewed previous reports of FCE of the spinal cord in children younger than 18 years to elucidate the characteristics in pediatric patients. Comprehensive review of the literature includes epidemiology, clinical course, diagnosis, treatment, and prognosis of FCE among pediatric patients. We have obtained the patient's and his parents' consent to present this case.

## Case Description

A previously healthy 15-year-old soccer player presented with weakness of both lower extremities and ischuria. He was a junior high school soccer player who practiced for more than 2 hours 4 times a week. Two days before admission, he felt progressive heaviness on his right leg without back pain after kicking a soccer ball. A day before admission, in addition to right lower extremity weakness, left extremity weakness developed, and he started to feel tingling pain around both calves. Furthermore, he developed ischuria on the same day. He denied any trauma, infectious symptoms, or fever. He had no significant medical or family histories.

On admission, he was afebrile and other vital signs were normal. On neurologic examination, his mental status and cranial nerves were normal. He had no tenderness over his back, and he had 5/5 strength in his upper extremities. Although he described weakness of both lower extremities, he only demonstrated mild distal right leg weakness (ankle dorsiflexion and plantar flexion, 4/5). The right Achilles reflex was absent and all other reflexes were intact; there was no Babinski sign. Sensory and sphincter examinations were unremarkable but he did have ischuria. He was able to walk without assistance. However, his gait was unsteady; he could not walk on tiptoe.

Magnetic resonance imaging (MRI) studies of the brain, and cervical and thoracic spinal cord were unremarkable. However, a lumbar MRI showed a hyperintense lesion on T2-weighted images (T2WI) in the anterior horn of the spinal cord at the Th12/L1 level without enhancement and a disc collapse with Schmorl's node at the L1/2 level, resulting in a vertical protrusion of the intervertebral disc cartilage into the adjacent upper and lower vertebral bodies (Figure). Blood tests revealed no abnormalities and the cerebrospinal fluid (CSF) analyses were also normal except for slightly elevated protein and myelin basic protein (Table 1). The nerve conduction velocity (NCV) test was normal.

Our differential diagnosis on admission included transverse myelitis and clinically isolated syndrome (CIS), but the MRI findings were atypical. After a Foley catheter was inserted, he was given two days of high-dose steroids (1 g/day) that were discontinued due to elevated blood sugar. Steroids was tapered to 1 mg/kg for 2 days and then to 0.5 mg/kg for 2 days. Although his right leg weakness did not completely improve with steroid pulse, ischuria completely disappeared and the Foley catheter was removed 8 days after admission. A repeat MRI on the same day showed a new finding of enhanced lesion at T12/L1, and the previous high-intensity T2WI at T12/L1 was reduced (Figure). He was discharged 9 days after admission. Although the strength of his right leg remained at 4/5 at discharge, 3 weeks later his right muscle strength returned to 5/5, his gait was steady and he could walk on tiptoe. Although his likely diagnosis on admission was transverse myelitis or CIS, literature review led to the consideration of FCE of the spinal cord as the most likely final diagnosis.

## Method

### *Systematic review of the literature*

Literature in English was reviewed for cases reported as FCE of the spinal cord in pediatric patients. The PubMed database, up to January 29, 2019, was searched using the following search terms: "Fibrocartilaginous emboli" or "Fibrocartilagenous emboli" or "nucleus pulposus emboli". A total of 123 articles were found; the abstracts or manuscripts of these papers were considered carefully, resulting in 18 case reports, 1 letter to the editor, and 1 case report and review of the literature of interest. After studying these articles carefully, we identified 20 literatures that described a case of FCE of the spinal cord in pediatric patients, and a total of 25 children younger than 18 years of age, including our case, were reviewed[1-20].

## Result

***Characteristics of patients with FCE from the literature***

The characteristics of patients with FCE from the literature and our case are shown in Table 2. Twenty-five pediatric patients were reviewed. Only 3 cases were pathologically defined and the remaining 22 cases were suspected clinically by presentation, neurologic, and MRI findings. The median age at presentation was 14 years (interquartile range (IQR): 12–16). Of the 25 patients, there were more female patients than male (16 females vs 9 males). The trigger events included intense exercise or sports (n=13), trauma (n=6), motor vehicle accidents (n=1), riding a roller coaster (n=1), milking a cow (n=1), leaning forward (n=1), and unknown (n=2). The neurologic symptoms started within 2 hours in half of the cases (n=12), and within 1 day in most cases (n=18). The time to symptom peak varied from a few hours to a few weeks. Most cases showed pain (n=21), and back pain was the most common (n=17). Initial neurologic symptoms included weakness or plegia of the extremities (n=25), paresthesia or numbness (n=11), bladder or bowel incontinence (n=7), ischuria (n=4), and ventilatory failure (n=2). Abnormal findings of neurologic examination at presentation commonly included weakness of extremities ranging from complete plegia to mild weakness, diminished or compete loss of sensation, and bladder or bowel function problems. Affected spinal segments of the spine by MRI (n=23) were as follows (cervical (n=5), cervicothoracic (n=5), thoracic (n=6), thoracolumbar (n=5), and lumbar (n=2)). Two autopsy cases reported FCE in the spinal cord arteries from the lower medulla to T7 and in the lower medulla and C2-T7. Abnormal vertebral body or disc changes were reported in 15 cases and apparent Schmorl's node was reported in 6 cases. CSF was analyzed in 22 patients, and 13 were unremarkable. Elevated MBP, protein, and leukocytosis in CSF were observed in 3, 5, and 2 patients, respectively. Treatments were as follows: intravenous steroids (n=14), anticoagulant (n=7), IVIG (n=2), fluoxetine (n=1), plasma exchange (n=1), Vitamin B12 (n=1), cyclophosphamide (n=1), and 9 patients were not treated by any medicine. As for prognosis, only one patient fully recovered. Most patients (n=21) had mild to severe sequelae and 3 patients died.

## DISCUSSION

FCE is a very rare cause of acute ischemic neuropathy and is not often suspected on presentation. In our case, the diagnosis was delayed because of the rare and underrecognized disorder. According to our review, only 25 cases of FCE of the spinal cord in pediatric patients younger than 18 years of age have been reported in the available English literature from 1961[1-20.] In addition, only 3 cases were pathologically defined[17,19,20]. It is possible that FCE is more common among children than expected.

***Epidemiology of FCE of the spinal cord***

According to a previous study, FCE shows bimodal distribution, with peaks in adolescence and late middle age[1]. Some review articles about FCE among adults or adults as well as a few children have been reported and sex differences differ depending on reports (male dominancy[21], and female dominancy[19,22,23]). According to our review, the median age of FCE of spinal cord among children was 14 years (IQR: 12-16) and the youngest was 8 months old. Our article showed female dominancy (64%) among children with FCE of the spinal cord. Although vertebral body damage by osteoporosis or cartilage degeneration, often found in postmenopausal women, is a possible risk factor for the occurrence of FCE of the spinal cord, it is rare in female children. Therefore, the reason for female dominance of FCE is unclear.

***Pathophysiology of FCE of the spinal cord***

The definitive pathophysiology of FCE remains unclear. However, some mechanisms have been proposed[1,2,23]. The most accepted hypothesis is that forceful herniation of the intervertebral disc nucleus pulposus material into the intradiscal or vertebral body vessels secondary to minor trauma or some axial loading forces induces the prolapse of cartilaginous material into the spinal artery[1]. The nucleus pulposus may also extend into the vertebral body by Schmorl's node[2,23] or probably abnormal vertebral body or disc changes[9-12,15-17,19]. Schmorl's nodes, focal masses of fibrocartilage within the vertebral body, are commonly found in adults but are rare among children[24,25]. However, our review showed that 6 cases (24%) had apparent Schmorl's node. In addition, abnormal vertebral body or disc changes were reported in 15 cases. Therefore, these abnormalities will also be helpful to diagnose FCE of the spinal cord among children. It is well known that the common blood supply to the spinal cord and nucleus pulposus, existing in infancy to childhood, generally becomes avascular after adolescence. The persistence of this vasculature has been postulated to increase the incidence of FCE around adolescence[23]. From our review, the median age of FCE of spinal cord among children was 14 years. Therefore, this result is compatible with the physiology of the vasculature of the spinal cord and nucleus pulposus. In the present case, we hypothesized that our patient developed FCE by minor trauma (kicking a soccer ball), which resulted in increased intervertebral pressure, which, in turn, caused the migration of fibrocartilaginous material (Schmorl's node may be involved) into this vasculature.

***Symptoms of FCE of the spinal cord***

FCE patients typically present with weakness that progresses to paralysis over a period of minutes to hours, often with sudden back pain, sensory problems, or bladder dysfunction after a suggestive history of a minor traumatic events[21-23]. Although the clinical course usually involves back or neck pain at onset[22], some cases, including our case, did not experience such pain[9,10,15]. The time to symptom peak reported by previous articles including adults and few children varied from a few hours to 48 hours[21-23,26]. The characteristic finding of difference between spinal cord infarction and inflammatory cord disease is a rapid course of symptoms to equilibrium state, mostly over hours[23]. Our patient developed sudden progressive right lower extremity weakness after kicking a soccer ball. He then developed left extremity weakness and felt tingling pain around both calves. Furthermore, he developed urinary incontinence later. Our review also showed that the neurological symptoms occurred within 2 hours from the event in half of the cases, and within 1 day in most cases. The time to symptom peak varied from a few hours to 2 weeks. These results are the same as those of previous articles on adults and a few children[21-23]. This temporal progression, like that in the present case, may be important in differentiating FCE from other mechanisms of arterial embolism[23]. Minor injuries and/or Valsalva maneuver have been often reported as the trigger of FCE[21-23]. Similarly, the trigger event in our review showed that only 6 patients (24%) had an apparent trauma event, and half of the patients (n=13) had intense exercise or sports. Therefore, progressive weakness, paresthesia, or bladder or bowel dysfunction after these events are highly indicative of FCE of the spinal cord.

***Diagnosis of FCE of the spinal cord***

FCE is often misdiagnosed with transverse myelitis, like in our patient[3]. Although a definitive diagnosis of FCE can only be made after an autopsy, diagnosis by clinical and radiological features is considered possible[1-18]. The number of clinically diagnosed cases has been exceeding the number of autopsy-proven cases, and we found only 3 pediatric cases of FCE of the spinal cord that were pathologically defined[17,19,20]. The MRI findings of expansion of the spinal cord with increased signal on T2WI involving the spinal cord without early contrast enhancement in association with a narrowed disc or Schmorl's nodes strongly suggest the diagnosis of FCE[1,2,23]. Schmorl's nodes have been often found in the vertebral bodies in approximately 40% patients with FCE[20,26] but AbdelRazek et al.[23] reported that degenerative disc diseases and Schmorl's node were observed in only 32% and 17% of FCE patients, respectively. DWI of MRI is also reported as good modality because abnormalities can be found even within a few hours in patients with spinal cord ischemia[12]. However, MRI sometimes reveal normal results

in the early stage of FCE[2-4,13]. Therefore, in such a case, repeated MRI of T2WI or enhanced MRI will be needed later. Subacute MRI often shows "pencil-like" T2WI hyperintensity in the spinal cord[21-23]. CSF analysis is usually unremarkable. However, Mateen et al.[21] reported elevated CSF protein in 6 out of 8 patients. In our patient, the clinical course was characteristic, marked by a progressive neurologic deficit after physiological effort, increased T2WI signal intensity in MRI without early contrast enhancement, and the lesion was found in the anterior cord and was asymmetric (more on the right), corresponding with his right weakness, and the presence of Schmorl's node was highly suggestive of FCE. The differential diagnosis for progressive weakness is broad, including transverse myelitis, acute polyneuropathy, early multiple sclerosis, or neuromyelitis optica. Acute transverse myelitis usually involves multiple spinal cord levels on MRI and typically involves at least two-thirds of the cross section of the spinal cord[27], whereas in our patient, only a small portion of the anterior spinal cord was involved. In the present case, the patient had no prodromal illness or trauma, and had a negative thrombotic evaluation and aquaporin-4 immunoglobulin G (IgG) in the blood as well as lymphocytic pleocytosis, elevated protein level or IgG index, oligoclonal bands in the CSF, and a normal NCV test. Therefore, the clinical history in combination with these results and MRI features were highly suggestive of FCE, and the abovementioned differential diagnoses were excluded. Our study also showed that 23 patients were performed MRI and all cases showed T2WI hyperintensity in the spinal cord. Therefore, T2WI of MRI will be helpful tool to diagnose FCE of the spinal cord. The commonly affected sites are reported to be the cervical[1,22,23] or thoracic regions[21,23,26]. However, our review of the literature of FCE in children showed that the affected area was distributed evenly from the cervical to thoracolumbar regions.

***Treatment and prognosis of FCE of the spinal cord***

There is no specific treatment for FCE of the spinal cord. It varies from emergent surgery to steroid or anticoagulant therapy or blood pressure control, and supportive care and rehabilitative physical therapy are also important in long-term treatment[21-23]. However, there have been no report of significant clinical improvement after treatment. The prognosis of FCE of the spinal cord varies from poor to significant neurologic improvement due to the involved area in the spinal cord and the extent of spinal cord ischemia[21-23]. Involvement of the cervical spinal cord frequently shows an unfavorable progression due to respiratory failure[1,26]. In our case, we started steroid pulse therapy for unknown cause of myelitis and started rehabilitative physical therapy. His lower extremity weakness improved daily, and finally, his ischuria recovered. After 3 weeks

from onset, difficulty in ambulating on tiptoe has not improved. Although the degree of recovery of spinal cord ischemia is generally poor, our patient showed relatively good neurologic improvement, which may because the involved spinal cord area was the lumbosacral part and the extent of spinal cord ischemia was small compared with previous reports[2,5,23]. Our review showed half of the cases were treated by steroids and/or anticoagulant. However, most cases had finally mild to severe sequela. Therefore, the effect of these treatments on FCE are unknown. However, one completely recovered patient was treated by steroid and 3 patients who died were not treated by medicine. Further research will be necessary.

***Limitation***

Because FCE of the spinal cord can be definitively diagnosed only by autopsy, some previously reported cases of spinal cord infarction might not have been caused by FCE. In addition, as we only included articles in English, we might have missed some important articles written in other languages.

## Conclusion

In conclusion, although FCE of the spinal cord can only be diagnosed by histopathology, we finally diagnosed our patient with FCE according to clinical presentation, laboratory data (which failed to reveal any other plausible diagnoses), and MRI findings. Although FCE is a very rare condition, physicians should be aware of the characteristics of FCE and should include FCE of the spinal cord in their differential diagnosis, especially for physically active young pediatric patients.

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## Conflict of Interests

This study was supported by a Grants-in-Aid for Scientific Research (KAKENHI) from the Ministry of Education, Culture, Sports, Science and Technology of Japan (subject ID: 18K089181 8 to Hiroaki Nagase).

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**Figure 1.**

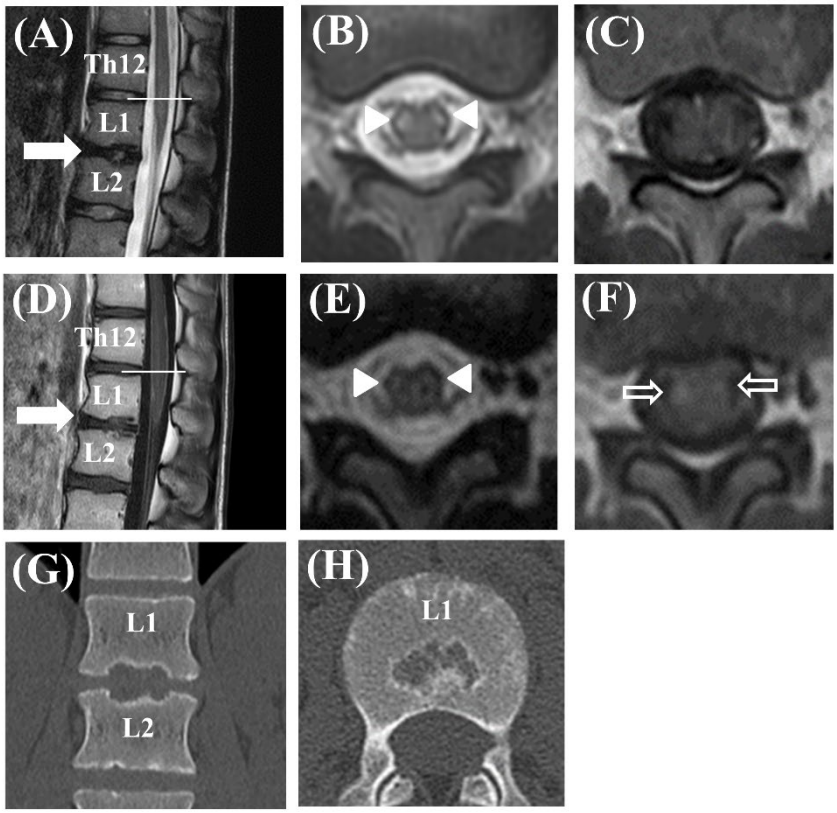


**Figure legends**

**Figure 1.** Sagittal (A), axial T2-weighted (B), and contrast-enhanced (C) images of the lumbar spinal cord on day 1. The sagittal and contrast-enhanced images of Th12/L1 showed no abnormalities. The axial T2-weighted image showed an increased signal bilaterally at the Th12/L1 level in the anterior horn of spinal cord (B) (arrowhead). The sagittal image revealed no abnormality on day 13 (D). However, a high-intensity area of the axial T2-weighted image at Th12/L1 appeared diminished (E) (arrowhead), and new findings were noted on a contrast-enhanced image at Th12/L1 (F) (the right side is more strongly enhanced than the left side; empty white arrow). In addition, L1/L2 disc collapse and Schmorl's node (white arrow) (A and D) were observed, along with adjacent vertebral end-plate destruction on computed tomography (G and H).

# Tables

**Table 1. Laboratory data of the patient**

| | Reference range | Value | | Reference range | Value |
|---|---|---|---|---|---|
| **CBC** | | | **Antibody** | | |
| WBC ( /μL) | (3,900-9,800) | 6,900 | Toreponema antibody | (n.d.) | n.d. |
| Hb (g/dL) | (12.6-16.5) | 12.2 | Anti-nuclear (times) | (≦40) | < 40 |
| Plt ($\times 10^4$/μL) | (17-41) | 22 | Anti-DNA (IU/mL) | (≦6) | ≦2.0 |
| | | | Anti-DS-DNA IgG (IU/mL) | (≦12) | < 10 |
| **Chemistry** | | | anti-CL.B2GP1(U/mL) | (≦3.5) | ≦1.2 |
| AST (U/L) | (14-30) | 23 | anti-aquaporin4 (U/mL) | (≦3) | < 1.5 |
| ALT (U/L) | (9-35) | 11 | MOG antibody | (n.d.) | n.d. |
| Na (mEq/L) | (138-144) | 139 | C3 (mg/dL) | (73-138) | 93 |
| K (mEq/L) | (3.7-4.7) | 3.9 | C4 (mg/dL) | (11-31) | 25 |
| Cl (mEq/L) | (102-109) | 104 | CH50 (/mL) | (25-48) | 30.3 |
| BUN (mg/dL) | (6.8-18.8) | 10.4 | | | |
| CRE (mg/dL) | (0.48-0.93) | 0.80 | **CSF** | | |
| GLU (mg/dL) | (73-109) | 85 | Cells (/uL) | (0-5) | 1 |
| TP (g/dL) | (6.3-7.8) | 7 | Glu (mg/dL) | (47-69) | 57 |
| CK (U/L) | (50-275) | 210 | Protein (mg/dL) | (8.8-27.1) | 31 |
| TSH (μU/mL) | (0.55-4.78) | 1.586 | MBP (pg/mL) | (≦102) | ≦40 |
| Free T4 (ng/dL) | (0.95-1.74) | 1.19 | Oligoclonal band | (n.d.) | n.d. |
| ESR (mm) 1h | (2-10) | 4 | IgG index | (≦0.73) | 0.38 |
| 2h | (4-20) | 10 | | | |
| Protein C activity (%) | (64-146) | 90 | **Others** | | |
| Protein S activity (%) | (67-164) | 76 | T-SPOT | (n.d.) | n.d. |
| | | | Mycobacterium culture | (n.d.) | n.d. |
| **Coagulation** | | | | | |
| PT (%) | (70-130) | 96 | | | |
| PT (INR) | (0.9-1.1) | 1.02 | | | |
| APTT (sec) | (26.6-40.3) | 27.4 | | | |
| AT-III | (83-118) | 111 | | | |
| D-D | (≦0.9) | 1 | | | |

Abbreviation: CBC, complete blood cell counts; WBC, white blood cells; Hb, Hemoglobin; Plt, platelet; AST, aspartate aminotransferase; ALT, alanine aminotransferase; BUN, blood urea nitrogen; CRE, creatinine; GLU, glucose; TP, total protein; CK, creatine kinase; TSH; thyroid-stimulating hormone; ESR, erythrocyte sedimentation rate; PT, prothrombin time; INR, international normalized ratio; APTT, activated partial thromboplastin time; AT, Antithrombin; D-D, D-dimer; MOG, myelin-oligodendrocyte glycoprotein; CSF, cerebrospinal fluid; MBP, myelin basic protein; n.d., not detected;

**Table 2. Comprehensive review of our case and previous articles reporting fibrocartilaginous embolism in children younger than 18 years of age**

| Cases | Author | Publication (y) | Age | Sex (M/F) | Triger event | Time from trigger event to neurologic sympyoms/ Time to symptom peak | Initial neurologic symptoms | Pain | Abnormal findings of neurologic examination on admission | MRI findings (days after trigger event) | Presence of abnormal vertebral body or disc changes | Treatment | Lunber puncure | Suspected case/ Pathologically definite case | Prognosis at latest follow-up |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **Present case** | Yamaguchi H, et al | | 15y | M | Kicked a ball during soccer practice | soon after the event/2d | Weakness of both lower extremities and ischuria | Tingling pain both calve | Mild distal weakness in right lower extremity<br>Absentright Achilles DTR<br>Umbulate difficulty without assistance and unable to bear any weight on his both legs<br>Ischuria | T2WI hyperintensity in the anterior horn of the spinal cord at the Th12/L1 (d2) and enhanced lesion at T12/L1 (d8) | Schmorl's node (L1-L2) | iv high-dose steroids | Yes/elevated MBP and mildly elevated protein | Suspected | Steady gait after 3 wks; however, umbulation difficulty on tiptoe |
| **1** | Shah Sv, et al[3] | 2018 | 12y | F | Unknown | - | Right-sided weakness, and paresthesia | Headache<br>Neck | Moderate weakness in the right lower extremity<br>Complete monoplesia in the right upper extremity<br>Diminished sensation to light touch in the right forearm and upper arm | Normal (2d after admission)<br>T2WI hyperintensity within the C2-C3 intervertebral disc posteriorly with a small focal protrusion (several d after admission) | No | Fluoxetine<br>Aspirin<br>IVIG<br>iv steroids | Yes/unremarkable | Suspected | After several wks, able to walk with ankle foot orthotic for the right lower extremity |
| **2** | Nagata K, et al[4] | 2017 | 8y | F | Hit back and neck with hyperflexion during a handstand | <2h/2h | Weakness in lower extremities | Back (minor) | Total paraplesia<br>Complete loss of pain, temperture sensation and a sense of vibration below T11 region<br>Bradycardia | Normal (d1)<br>T2WI hyperintensity with the spinal cord T11-cranial level (d2)<br>T2WI hyperintensity with longutudinal extension of the region between C6-T11 (d2)<br>DWI hyperintensity below C6 (1wk)<br>Minor bleeding at T11 vertebral region and spinal cord edema below C6 (T2 star)(2 wks) | No | None | Yes/unremarkable | Suspected | After 1y, complete paraplegia with sensory loss below T11 |
| **3** | Jones DD, et al[5] | 2016 | 14y | M | Run sprints and made quick turns at the gym | <1d/1d | Weakness of lower extremities and urinary incontinece | Lower back | Complete paraplesia in lower extremities with the exception of severe weakness in left extensor hallicus longus and dorsiflexion of left ankle<br>Diminished bilateral sensation at L2-S1 region<br>Absent patellar and Achilles DTR<br>Absent volitional contraction of sphincter | T2WI hyperintensity over two-thirds of the spinal cord at T12-L1 with DWI restricted diffusion (d1) | Schmorl's node (Th12-L1) | Heparin<br>iv steroids | No | Suspected | After 1 mo, bladder catheterization and able to walk with a front-wheeled walker |
| **4** | Eid R, et al[6] | 2016 | 12y | F | Rode a roller coaster | <several h/several h | Left-sided weakness | Neck | Mild weaknesses in left sternocleidomastoid and trapezius and right deltoid<br>Severe weakness in left upper extremity of deltoid<br>Complete plesia in biceps, triceps, wrist flexion, wrist extension, and grip<br>Mild weakness in hip flexion, foot dorsiflexion, and foot plantar flexion<br>Moderate weakness in left lower extremity<br>Mild slowing of coordination in the right upper extremity | T2WI hyperintensity along the midline portion of C2-C3 and swelling of the spinal cord between C2-C5 (d1) | No | iv high-dose steroids<br>Enoxaparin | Yes/unremarkable | Suspected | After 1 y, left hemiparesis, spasticity of both upper and lower extremities, and limited motion of the right upper extremity with greater limitation in the right shoulder |
| **5** | Nelson JA, et al[7] | 2016 | 17y | F | Intense tennis practice | <1d/5d | Left leg monoplegia and paresthesia | Back and left hip<br>Bilateral lumber | Flaccid paralysis of the left lower extremity<br>Mild weakness in righ lower extremity<br>Decreased temperature sensation in the right lower extremity<br>Highper DTR in right knee<br>Urinary incontinence | A small disc extrusion with superior migration at T10-T11 with T2WI hyperintensity of the anterior spinal cord between T9-T11 (d3 and 5) | Schmorl's node (T11-T12) | Aspirin<br>iv high-dose steroids<br>Vitamin B12 | Yes/elevated MBP | Suspected | After 18 mo, decreased pain, temperature, and pressure sensation and nearly baseline strength of right leg and left leg, and return to modified athletic activity |
| **6** | Rengarajan B, et al[8] | 2016 | 14y/10y | M/F | Case1: Hammered fence posts with a sledgehammer<br>Case2: Competed in a long jump event | Case1: 15min/several h<br>Case2: <2h/a few h | Case1: Weakness of lower extremities, paraesthesia, and bowel and bladder incontinence<br>Case2: Weakness of lower extremities, paraesthesia, and bowel and bladder incontinence | Back (center)/ Back and buttock | Case1: Mild weakness in right hip flexors and moderate weakness in distal muscles of the right lower extremity<br>Complete plegia of left hip, knee, or ankle<br>Absent DTR in the patella or ankle<br>Decreased tone in both lower extremities<br>Absent sensation of pin prick, light touch, temperature, and vibration below the T5 region<br>Absent proprioception in lower extremities<br>Case2: Moderate weakness in bilateral hip extensors, knee flexors and extensors<br>No movement at the ankle and toes<br>Absent DTR in patella and ankle<br>Positive Babinski's reflexes<br>Absent sensation to fine touch, pin prick, cold, vibration, or proprioception below L3 region | Case1: Long pencil-like segment of T2WI hyperintensity and restricted DWI in the anterior portion from C7/T1 to T6/T7 (d1)<br>More apparent T2WI hyperintensity at same level (d10)<br>Case2: T2WI hyperintensity and restricted DWI in the conus medullaris at the level of T12-L1 (d1)<br>T1WI slightly hyperintensity at the anterior spinal artery at the T12-L1 and normal enhanced-MRI (d1) | No/No | Case1: iv high-dose steroids<br>Enoxaparin<br>Warfarin/<br>Case2: iv high-dose steroids<br>Aspirin | Yes/unremarckable/<br>Yes/unremarckable | Suspected/<br>Suspected | Case1: After 13mo, able to walk, bike, and run normally. However, persistent right lower limb temperature hypoesthesia<br>Case2: After 11mo, able to walk independently with a left ankle foot orthosis, and decreased sensation below knee |

Table 2. *Continued*

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|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 7 | Bamal S. et al[9] | 2014 | 16y | F | Intense exercise | soon after the event/2d | Umbulate difficulty and paresthesia of lower extremities<br>Tingling of the right leg | None | Mild weakness in the lower extremities in the bilateral iliopsoas and quadriceps<br>Hightper DTR in the bilateral patella and positive Babinski's reflex in the right<br>Diminished proprioception, temperature, and pinprick distal to T12<br>Dysmetriabilateral heel-shin-knee testing<br>Wide-based gait and positive Romberg sign | Several T2WI hyperintensity in the dorsal spinal cord at T11-T12 (d4)<br>restricted DWI at T11-T12 (d5) | Intervertebral disc desiccation and bulge at T11/T12 | Anticoagulation | Yes/elevated MBP | Suspected | After 2mo, mild sensory changes in the right extremity |
| 8 | Reinner A. et al[10] | 2013 | 8y/8m/12y | F/F/F | Case1: Danced at ballet class<br>Case2: Fell off of a changing table<br>Case3: Ground-level fall | Case1: soon after the event/ 2d<br>Case2: 2h/2h<br>Case3: soon after the event/ <1d | Case1: Paresthesia and numbness in hands and difficulty bending, and umbulation difficulty<br>Case2: Paraplegia in upper extremities<br>Case3: Complete quadriplegia and numbness over entire body | Neck/ None/ None/ | Case1: Complete quadriplegia, diminished sensation to pain and temperature<br>Case2: Paraplesia in her upper extremities<br>Case3: Complete quadriplegia and diminished sensation over her entire body<br>ventilatorary failure | Case1: T2WI hyperintensity in the C2-T1, and diminished T2WI hyperintensity in the C3-C4 (d2)<br>Case2: Central spinal cord signal changes at C2-T1 (d1)<br>Case3: T2WI hyperintensity and restricted DWI at C1-C4 (d1) | Case1: No<br>Case2: T2WI hypointensity in multiple intervertebral discs (C2-C7)<br>Case3: Diminished T2WI signal in C3-C4 disc space | iv steroids/ iv high-dose steroids/ iv high-dose steroids | Yes/unremarkable<br>No/<br>Yes/unremarkable | Suspected/ Suspected/ Suspected | Case1: After 3mo, improved to an ASIA Grade D, ambulatory without assistance<br>Case2: After 19mo, ASIA Grade C<br>Case3: After 12mo, ASIA Grade D with residual weakness on the left side, and able to walk independently |
| 9 | Holland[11] | 2012 | 17y | F | Unknown | - | Quadriparesis, ventilatorary failure and weakness in upper extremities | Back (between sholder blades) | Moderate weakness of deltoid and biceps, significant weakness of triceps and the wrists, and paraplegia<br>Numbness in both legs coming up to the trunk<br>Reduced DTR in the arms and absent in the legs<br>Ischuria | T2WI and enhanced T1WI hyperintensity in the spinal cord at C5-7 (d7 after admission) | Small annular tear at C5-6 | IVIG<br>plasma exchange<br>iv cyclophosphamide | Yes/unremarkable | Suspected | After several mo. weaned from the ventilator and regained some strength in the upper extremities. However, remained paraplegic and confined to a wheelchair |
| 10 | Manara. et al[12] | 2010 | 13y/12y | M/F | Case1: Payed basketball, intense stretching, squanting and lifting exercises<br>Case2: Fell backward, banging her back against a door | Case1: several d/several d<br>Case2: a few h/a few h | Case1: Paraplegia, paresthesia in lower extremities<br>Case2: Leg weakness, bowel and bladder incontinence | Case1: Back (interscapular) spreading to the epigastric region<br>Case2: Back | Case1: Flaccid areflexic paraplegia with absent abdominal reflexes and hypoesthesia below T7<br>Bladder disfunction<br>Case2: Flaccid areflexic paraplegia, bilateral mild hypoesthesia below T10 | Case1: Slight intramedullary T2WI hyperintensity at the T7-T8 (several d)<br>Intramedullary T2WI hyperintensity in the anterior spinal artery region at T4-T5 and T7 (several d-d2)<br>Medullar T2WI hyperintensity in the anterior spinal artery territory (several d-d5)<br>Case2: T2WI hypointensity of the L2-L3 disc (d1)<br><br>T2WI and DWI hyperintensity "owl's eyes-like" within the spinal conus (d3) | Case1: Thiner intervertebral disc and T2WI hypointensity at T4-T5<br>Case2: Schmorl's node (L1-L2, L2-L3) | Case1: iv high-dose steroids<br>Case2: iv high-dose steroids | Case1: Yes/unremarkable<br>Case2: Yes/mild leukocytosis | suspected | Case1: After 1y, symptom-free<br>Case2: After 2y, weakness in left lower extremity |
| 11 | Tan K. et al[13] | 2009 | 16y | M | Elbowed in the face during gym class and wrestling | 2d/2d | Weakness of lower extremities | Lower back | Complete paraplegia, sensory loss below T10<br>Urinary and bowel incontinence | Normal (d1)<br>T2WI hyperintensity at T11-L2 (19mo) | Schmorl's node (L1-L2) | None | Yes/mildly elevated protein | Suspected | After 26mo, asymmetric spastic parparesis and reduced lower extremity sensation |
| 12 | Raghavan A, et al[14] | 2004 | 14y | M | Trivial forced flexion of neck while playing with his dog | 10d/10d | Weakness of all extremities | Back (interscapular) | Flaccid quadriplegia<br>Absent DTR and abdominal cremasteric reflexes<br>Absent pain and temperature sensation below C1<br>Urinary incontinence | T2WI hyperintensity with linear lesion at C1-T1 (d1) | No | None | No | Suspected | After 6wks, recovered antigravity strength of finger and wrist extension and flexion on the right, with a flicker of finger flexion on the left |

**Table 2.** ***Continued***

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|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **13** | Han JJ, et al[2] | 2004 | 16y | M | Lifting exercises | 1d/2d | Weakness in lower extremities, paraparesis and ischuria | Back (lumber) | Flaccid in left lower extremity, and severe weakness in right lower extremity<br>Absent sensation to pinprick bilaterally below T6 level anteriorly and T8 level posteriorly<br>Absent perianal pinprick sensation<br>Trace DTR in the upper extremities and absent quadriceps and Achilles DTR<br>Low sphincter tone | Normal (d1)<br>T2WI hyperintensity at the T7-T8, localized within the left central and anterior spinal cord (2wks) | No | None | Yes/unremarkable | Suspected | After 8mo, normal strength in the upper extremities, abdominals, and right lower extremity<br>Normal strength in left lower extremity except for mild weakness of knee flexion reduced pinprick and light touch on the right (T8-S5) |
| **14** | Beer, et al[15] | 2002 | 7y | F | Car accident | <1d/1d | Paraplegia in lower extremities, paraparesis, ischuria, bowel incontinence | Chest (blunt trauma) | Flaccid paraplegia with a complete sensory loss below the T12 region, ischuria and bowel incontinence<br>Absent Knee and ankle DTR | T2WI hyperintensity of the spinal cord from T4 down to the lumbar region (d3) | Narrowed intervertebral disc at T6/T7 | None | No | Suspected | After 6mo, able to walk with ortheses<br>Slight activity in all muscle groups of the lower extremities<br>Marked spasticity of the lower extremities<br>Impaired perception of touch, position sense and vibration below L3 |
| **15** | Davis GA, et al[16] | 2000 | 6y | F | Performed cartwheel | a few w/a few w | Paraplegia in lower extremities and bladder incontinence | Lower back | Flaccid paraplegia of lower extremities<br>Absent DTR in lower extremities<br>Absent pain and temperature sensation to T11 | Multiseptated syrinx between T5 and T6-T10 (d1)<br>T2WI hyperintensity in the anterior spinal cord from T10 to the conus medullaris (d2) | Narrowed intervertebral disc at L1/L2 | iv steroids | Yes/unremarkable | Suspected | After 2mo, regained minimal strength in left quadriceps muscle, while the remainder of deficit not improved |
| **16** | Yousef, et al[17] | 1998 | 14y | F | Leaning forward to pick up an object | Soon after the event/1d | Weakness and numbness in lower extremities and ischuria | Upper back | Flaccid paralysis of the lower extremities<br>Decreased pain and light touch below level T6<br>Absent DTR in lower extremities | T8-T9 levels without evidence of spinal cord lesion (d1-2) | Irregularities of the end plates at T7-T8 and T8-T9 | None | Yes/unremarkable | Defined | Death on the 3rd day |
| **17** | Tosi, et al[1] | 1996 | 16y | F | Handstand | 30min/1d | Left foot weakness | Back radiating to lower extremities | Absent flexion and extension of the left foot and weak on the right<br>Absent ankle DTR<br>Reduced sensation of touch, pain, and temperature on the left extremity below the knee | T2WI hyperintensity within the cord from T11-L1 (d3) and in the mid-dorsal region (d10)<br>Pronounced atrophy of the epiconus at T11-T12 (T1WI) (5mo) | Schmorl's node (T12) | None | yes/elevated protein | Suspected | Not described |
| **18** | McLean, et al[18] | 1995 | 17y | M | Weight lifting | 12h/12h | Paresthesia in the lower extremities, paraplegia, and urinary incontinence | Burning pain in thighs, lower back, and lower extremity | Flaccid paralysis and absent sensation to all modalities at T12<br>Present priapism | T2WI hyperintensity at T7 (d3)<br>Atrophy of the cord at the T11-12 (T1WI) (3mo) | No | iv high-dose steroids | Yes/unremarkable<br>Yes/slight leukocytosis (Day5)<br>Yes/elevated protein (3wks) | Suspected | After 24 mo, no neurological improvement |
| **19** | Toro, et al [19] | 1994 | 16y | F | Milking a cow | 15min/<1d | Paraplegia in lower extremities | Lower back | Flaccid paraplegia with completely sensory loss from the L1 down and Absent DTR in the lower extremities<br>Ischuria<br>Loss of perineal sensation | Not performed | Old fracture at L1-L2 vertebral body | None | Yes/elevated protein | Defined | Death after 6wks |
| **20** | Naiman, et al[20] | 1961 | 15y | M | Basketball practice | 40min/2h | Weakness of all extremities and ventilatorary failure | Epigastrium, back, neck and shoulders | Complete flaccid paralysis and Absent DTR in all extremities<br>Respiratory paralysis<br>Sensory loss extending up to T3-T4 | Not performed | No | None | No | Defined | Death after 2h |

Abbreviation: ASIA; American Spinal Injury Association, d; day, DTR; deep tendon reflex, DWI; diffusion weighted image, F; female, h; hour, iv; intravenous, IVIG; intravenous immunoglobulin, mo; month, min; minute, M; male, w: week, T1WI; T1 weighted image,T2WI; T2 weighted image, wk; week, y; year,-; not applicable

Weakness is categorized as follows (manual muscle test (MMT): 0/5 complete plesia, 1/5-2/5 severe, 3/5 moderate, 4/5 mild, 5/5 normal)

ASIA Impairment Scale is categorized as follows (Grade A; Complete lack of motor and sensory function below the level of injury, Grade B; Some sensation below the level of the injury, Grade C; some muscle movement is spared below the level of injury, Grade D; most of the muscles that are spared below the level of injury, Grade E; all neurologic function has returned to normal)