

Nontraumatic ischemic spinal cord injury following vigorous exercise: a case report

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Abstract

Background: Spinal cord injuries (SCI), although rare in children, lead to significant morbidity and mortality.

Case presentation: This report describes the case of a 15-year-old male adolescent who developed SCI due to paradoxical embolism of a deep vein thrombus following strenuous exercise. He presented with bilateral upper limb weakness that progressed to severe tetraparesia, sensory deficits, and autonomic instability. Imaging revealed medullary ischemia with hypoperfusion of the anterior spinal artery. A family history of thromboembolism and obesity were noted as risk factors in the patient; however, thrombophilia screening was negative. Optimized fluid therapy was initiated for the medullary shock associated with dexamethasone and enoxaparin. As neurological recovery was incomplete, the patient was enrolled in an intensive rehabilitation program.

Conclusion: To the best of our knowledge, this is the first reported case of medullary ischemia due to a paradoxical embolism of a deep vein thrombus through a patent foramen ovale in an adolescent. This case highlights the need for a multidisciplinary approach to optimize long-term outcomes after medullary ischemia.

Keywords: Adolescent, Medullary shock, Spinal cord ischemia, Thromboembolism

INTRODUCTION

Spinal cord injuries are infrequent yet severe in the pediatric population and often result in substantial morbidity and mortality.^[1] Reports on pediatric spinal cord ischemia (SCI) are scarce, and traumatic vertebral injuries remain the predominant cause.^[2] Nevertheless, sudden interruption of spinal artery blood flow could also be due to other causes, such as infectious, cardiovascular malformations, thromboembolism, and vertebral, aortic, and cardiac surgeries.^[2]

Spinal cord injuries can affect the neurological pathways running through the spinal cord and compromise their regular functioning. Medullary shock consists of motor, sensitivity, and sensory deficits, encompassing paraplegia/quadruplegia and

bladder and/or bowel dysfunction.^[1] Its severity depends on the topographical location of the medullary insult and whether the section is complete or not.

Regardless of the underlying mechanism of the lesion, spinal cord injuries above the sixth dorsal vertebral level may lead to neurogenic shock.^[1] It is characterized by profound vasoplegia, hypotension, and bradycardia due to interruption of the descending sympathetic pathways with unopposed vagal tone.^[1] The anterior spinal artery is the most frequently affected vessel, leading to anterior spinal artery syndrome.^[3] Both pain and temperature sensation are commonly impaired, sparing the sense of vibration and proprioception.^[3]

This report details the case of an adolescent experiencing SCI secondary to embolism of a deep vein thrombus following minor trauma.

Case report

A 15-year-old male adolescent with obesity presented to the pediatric emergency department with diminished bilateral upper limb muscle strength and numbness after strenuous physical exercise. The exercise consisted of repeated movements of bilateral straight leg elevation in the supine position while keeping the knees extended. Examination revealed brachial diplegia without other motor abnormalities consistent with the “Man-in-the-Barrel Syndrome.” Additionally, both pain and temperature sensitivity were reduced in the lower limbs. The results of the remaining neurological examinations were normal. Laboratory test results at admission were unremarkable. Cranial and cervical computed tomography (CT) showed no abnormalities.

Over the initial 24 hours, the patient’s neurological status deteriorated, culminating in severe tetraparesia with distal predominance, thermo-algic hypoesthesia with T2 sensory level, reduced deep tendon reflexes with a flexor plantar response, left-sided Horner syndrome, and urinary retention.

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

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Figure 1. Cervical and thoracic T2-weighted magnetic resonance image shows extensive medullary lesion between C4 and T1 with T2 hypersignal (arrows), corresponding to acute medullary ischemia.

Given the neurological deterioration, cervical and thoracic magnetic resonance imaging (MRI) was performed. It revealed extensive, symmetric, anterior medullary involvement between C4 and T1 without signs of medullary expansion or other abnormalities (Fig. 1). Brain MRI was normal. Angiography revealed hypoperfusion of the anterior spinal artery with no evidence of vascular dissection or malformation (Fig. 2). A diagnosis of ischemic spinal cord injury was made, and the patient was referred to our pediatric intensive care unit (PICU). The medullary lesion was considered incomplete, and the neurological deficits were classified as grade C on the American Spinal Injury Association

impairment scale. The patient was administered dexamethasone (10 mg/m²/day) and acetyl-salicylic acid (ASA) (100 mg/day).

Patient had a positive family history for thromboembolism on the maternal side, as both his uncle and grandmother experienced acute ischemic stroke at a young age. Additionally, the patient's sister was administered anticoagulant and antiaggregation therapy during pregnancy because of previous spontaneous abortions of unknown cause.

A thorough etiological investigation was performed during hospitalization. Extensive thrombophilia screening was performed, which was negative for antiphospholipid syndrome, antinuclear antibody, antidouble stranded DNA antibody, antithrombin, protein C and S deficiency, factor V Leiden, and prothrombin G20210A mutation. Infectious, rheumatologic, and metabolic conditions were ruled out. Lower limb Doppler ultrasonography revealed a subacute occlusive thrombosis of an accessory vein of the posterior tibial vein (Fig. 3). Transcranial Doppler ultrasound revealed microemboli, which were suggestive of a right-left shunt. Transesophageal echocardiography revealed patent *foramen ovale*. Following these findings, ASA



Figure 2. Cranial and cervical angiography shows hypoperfusion of the anterior spinal artery (arrow) with no evidence of vascular dissection or malformation.

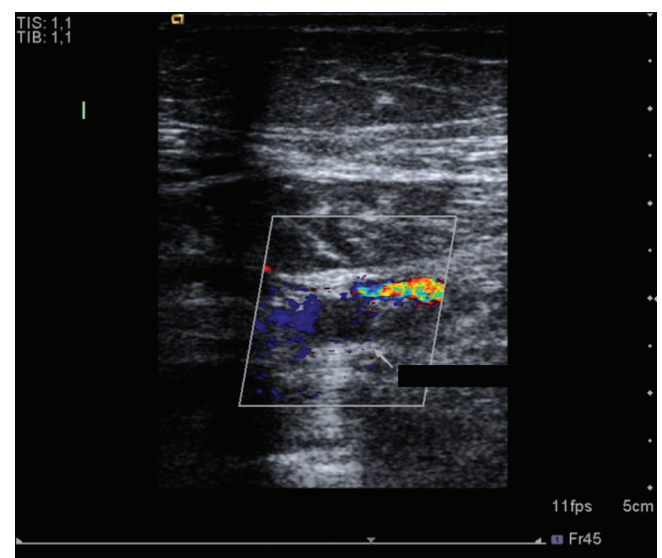


Figure 3. Left lower limb Doppler scan shows markedly reduced color flow in a non-compressible segment of an accessory vein of the posterior tibial vein (arrow), suggestive of subacute occlusive thrombosis.

was discontinued and anticoagulant therapy with enoxaparin was initiated, with regular monitoring of antifactor Xa activity.

On the third day, a dysautonomic state was observed with sinus bradycardia and hypotension. It persisted for approximately 48 hours and was successfully managed with an optimized crystalloid fluid intake of 2800 mL/day (1400 mL/m²/day), without the need for vasopressor support. During the hospital stay, a significant yet incomplete improvement of neurological deficits was observed. Motor function improvement was first noted in the lower limbs by the end of the first week, and 1 week later in the upper limbs. Additionally, deep tendon reflexes and urinary and fecal sphincter continence normalized during the second week. However, complete remission of symptoms was not observed upon hospital discharge. After discharge, the patient was enrolled in an intensive physical rehabilitation program that ultimately resolved the abovementioned neurological deficits. Following a cardiology consultation, the patent *foramen ovale* was percutaneously closed 2 months after discharge. After the procedure, antiaggregant therapy was resumed and maintained for 6 months without any thrombotic events. The enoxaparin therapy was maintained for 12 months without any new thromboembolic episodes.

Discussion

Spinal fractures account for a small percentage (1%–5%) of all fractures in children.^[4] The cervical spine is the most commonly affected segment, especially in younger children, because of the relatively high weight of the head and weakness of the cervical muscles.^[4] Cervical spine injuries display a bimodal age distribution, with the first peak occurring at preschool age and a later peak at adolescence.^[5] Cervical spine sports-related injuries are more frequent in adolescents, accounting for 25% of all cervical injuries. Male adolescents are particularly prone to these injuries, as they adhere more commonly to high-kinetic-energy contact sports.^[5,6]

Strenuous concentric muscle exercise, involving bilateral straight leg elevation, was the apparent trigger event in our patient, leading to embolism of a previously formed thrombus in the lower limb to the spinal vasculature. Vigorous physical exercises involving the Valsalva maneuver may lead to neurovascular complications, such as dissection of the cerebral and cervical arteries and intracranial aneurism rupture.^[7] It is thought to be secondary to supraphysiological increase in blood pressure, with a marked increase in cerebral blood flow and subsequent vascular injury.^[7]

Paradoxical embolism occurs when a thrombus crosses an intracardiac shunt into systemic circulation, leading to local hypoperfusion and ischemia.^[8] The transient pressure elevation in the right atria during the Valsalva maneuver may induce a reversal of the left-to-right intracardiac shunt and embolism to the systemic circulation, with disastrous consequences.^[8,9] To the best of our knowledge, this is the first report of spinal cord infarction secondary to paradoxical embolism in a pediatric patient.

The literature on SCI in children is scarce; however, severe systemic hypotension, thromboembolic phenomena, local vascular malformations, trauma, and vasculopathies have been proposed as potential risk factors.^[9] Thrombotic conditions have been reported to cause spinal cord infarctions in the pediatric population.^[10] These conditions consist of genetic or acquired deficiencies in coagulation. A family history of recurrent thrombotic episodes led to the investigation of suspected thrombophilia in

our patient; however, all results were negative. Obesity may have played a determining role on the deep vein thrombus formation.^[11] Obesity is a significant risk factor for thromboembolic events in adults. However, this association has not been well-established in children and adolescents.^[12] Obesity in adolescents may impose a higher risk of deep vein thrombosis, especially due to its association with a sedentary lifestyle and prolonged periods of venous stasis.^[11]

Imaging played a decisive role in the diagnosis in this case. The medullary lesion was not apparent on CT because its sensitivity to spinal cord infarction is globally low. Currently, state-of-the-art imaging equipment have superlative diagnostic capacity. However, a non-negligible proportion of cases do not display any radiological findings. This condition was originally described in the 1970s and is known as spinal cord injury without radiographic abnormality.^[13] It is common in children and may be caused by spinal cord injury secondary to vascular injury, spinal hyperflexion, or hyperextension.^[13] MRI is the preferred imaging modality for assessing medullary ischemia.^[14] Over the first few hours after a spinal insult, the usual MRI findings are restricted diffusion on weighted imaging, hyperintense signals on T2 and short tau inversion recovery, and isointensity on T1.^[14]

As the spinal injury was located above T6 level, the patient developed autonomic instability consistent with neurogenic shock, which was managed accordingly. The symptoms include profound vasoplegia, bradycardia, and hypotension. Optimal fluid resuscitation and vasoactive therapy are the cornerstones of neurogenic shock treatment for preventing secondary medullary ischemia.^[1,9] Management guidelines focus on maintaining an adequate blood pressure level to ensure spinal cord perfusion.^[15] The general recommendation for adults is to maintain a mean arterial pressure above 85–90 mmHg for 7 days after injury.^[15] There are no evidence-based pediatric guidelines; however, this target is considered adequate for adolescents.^[15] Hemodynamic instability often requires premature vasopressor support involving alpha- and beta-agonists to manage hypotension and bradycardia, respectively.^[16] Norepinephrine is considered a first-line treatment option. However, no randomized trials have been published to differentiate between the available vasopressors.^[15]

We administered a high-dose dexamethasone regimen, as per the local protocol, after a neurosurgery consultation. The therapeutic regimen was initiated at 20 mg/day for 5 days, followed by a 10-day tapering phase. Although there is no high-quality evidence comparing the efficacy of dexamethasone and methylprednisolone in SCI, the former is widely used in neurosurgical practice. The use of corticosteroids in spinal cord injuries is controversial, as the evidence is heterogeneous and conflicting.^[17] In adults, a short course of high-dose corticosteroids, such as methylprednisolone, may be considered within 8 h in spinal cord injuries.^[16] In addition, its use is associated with improved neurological outcomes owing to its neuroprotective effects.^[18,19] Evidence of its effects among children is even less robust, and its use is merely based on an extrapolation of the effects reported in adults. Some studies have suggested that timely administration of corticosteroids may also be considered in children, particularly in cases of cervical hyperextension.^[16]

Both antiaggregant and anticoagulant therapies were initiated early in the disease course in the present case. Following the identification of the deep vein thrombus, ASA therapy was suspended, and the enoxaparin dose was increased from preventive to therapeutic dosing. Anticoagulant therapy was maintained for a year without further thrombotic events. Recent guidelines

recommend maintaining anticoagulant therapy for 6–12 months for unprovoked events.^[20] Enoxaparin and warfarin are currently considered the gold standards for children, and clinical trials supporting the use of direct oral anticoagulants are still in progress.^[21,22] Venous thromboembolism management guidelines do not recommend antiplatelet therapy as a first-line option, reserving its use for secondary prophylaxis in children with significant risk factors.^[21] Regarding spinal cord infarction combined anti-aggregant and anticoagulant therapy may enhance patient outcomes in thrombotic disorders.^[23]

Rehabilitation after spinal cord injury is complex and requires a multidisciplinary team. Our patient enrolled in an intensive program with remarkable improvement. Considering the neuroplasticity in younger patients, rehabilitation is of pivotal importance in diminishing possible neurological sequelae.^[24] Rehabilitation should begin upon PICU admission to prevent sarcopenia, joint contractures, and other immobilization-induced complications.^[24] Additionally, elimination management should be prioritized as both urinary and gastrointestinal motility are commonly affected.^[24] Rehabilitation should also focus on mental health and social reintegration.^[16] Ensuring return to school, integration in community activities, and, ultimately, achieving an optimized quality of life are critical in pediatric cases.^[25] Depression, anxiety, and poor school performance are the most common mental health conditions, particularly in adolescents, and may contribute to poor quality of life in children with SCI.^[16,25]

Conclusion

In conclusion, pediatric SCI is rare and presents intricate challenges. This case highlights the complexity of the diagnosis and management of spinal cord injuries. To the best of our knowledge, this is the first report of medullary ischemia following paradoxical embolization of a deep vein thrombus through a patent *foramen ovale*. Acute management of spinal cord injuries in an intensive care setting remains controversial, particularly regarding corticosteroid use. Secondary neurological deficits may be long-lasting and significantly impact the quality of life of the affected children and their families. Thus, a holistic approach to rehabilitation is crucial for obtaining the best outcomes.

Conflict of interest

The authors declare no conflict of interest.

Author contributions

Sousa A and Branco J participated in conception and design, provision of study materials and patients, data collection and assembly. All authors contributed to data analysis and interpretation, manuscript writing, and final approval of the manuscript.

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Ethical approval of studies and informed consent

All procedures performed in this study were in accordance with the Helsinki Declaration (as revised in 2013) and with the ethical standards of the institutional research committee, and the study is exempt from ethical approval. Written informed consent was obtained from the patient for the publication of this case report.

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