



Postoperative Acute Ischemic Spinal Cord Infarction without Vertebral Fracture in Children

CASE REPORT

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ABSTRACT

Anterior spinal artery infarction is extremely rare in children; it is caused by the hypoperfusion of the anterior spinal artery, leading to ischemia. Patients typically present with acute paraparesis or quadriparesis, depending on the level of spinal cord involvement. In general, it is clinically diagnosed, and neuroimaging is used to confirm the diagnosis and exclude other conditions. There are only a few reports in the pediatric literature characterizing the etiology, diagnosis, treatment, and prognosis of spinal cord infarction. It is associated with moderate-to-severe disabilities and a high mortality rate. Here we report an unusual case wherein a 12-month-old male developed spinal cord infarction following surgery to repair the coarctation of the aorta. Spinal magnetic resonance imaging showed bilateral hyperintensity corresponding to the anterior horns of grey matter in the conus.

Keywords: Spinal cord infarction, myelopathy, magnetic resonance imaging

INTRODUCTION

Spinal cord infarction (SCI) is a rare but often devastating disorder caused by a wide array of pathological conditions (1, 2). There are a few studies on SCI that report heterogeneous etiologies, including spontaneous and traumatic vertebral artery dissection, hypotension, atherosclerosis of the vertebral arteries with severe stenosis, infrarenal abdominal aneurysm repair, epidural anesthesia administration, and vasculitis. SCI is most frequently caused by surgical procedures and pathologies that affect the aorta (1). In addition, SCI is often located in the anterior spinal artery (ASA) territory. The most common clinical presentation of SCI is the ASA syndrome (1, 3). Consistent with its neuroanatomical function, an ASA infarct typically presents as loss of motor and pain/temperature sensations, with relative sparing of proprioception and vibratory sensation below the level of the lesion (1). Magnetic resonance imaging (MRI) is required to diagnose SCI. MRI can also provide confirmatory evidence of SCI and information regarding its underlying etiology (1, 3).

CASE REPORT

A 12-month-old male was hospitalized for the surgical repair of the coarctation of the aorta. Vital signs and routine bloods test results prior to anesthesia administration were normal. General anesthesia was administered. The coarcted segment was opened and repaired with patch plasty during an uncomplicated surgery. Then, the patient was transferred to the intensive care unit. The patient's vital signs were postoperatively stable, and he was conscious and active. There were no pathological, physical, or neurological signs; however, postoperatively at 6–8 h, the patient was confused. He was also tachycardic and hypotensive.

Neurological examination showed that he was quite hypoactive and exhibited flaccid paraplegia (0/5 power in the legs) and areflexia in the lower limbs. Moreover, cutaneous abdominal reflexes in all regions were negative. The anal sphincter was flaccid. Based on these findings, acute flaccid paralysis was suspected, and the patient was referred for spinal MRI (Siemens Magnetom Aera 1.5-Tesla MR scanner). T2-weighted sagittal and axial MRI of the spinal cord showed bilateral hyperintensity corresponding to the anterior horns of grey matter in the conus (Figure 1). He was subsequently diagnosed with SCI. As soon as the infarction was detected, the patient was subcutaneously administered low molecular weight heparin (Clexan, Aventis Intercontinental, Fransa) 1 mg/kg/day (in 2 doses) for 10 days. On postoperative day 10, his condition had not improved. During clinical follow-up, he was in deep coma, and it was difficult to assess his neurological function. His pupils were fixed and dilated and bilateral pupillary light response and all brain stem reflexes were negative. Transcranial Doppler ultrasonography showed that the intracranial arteries were bilaterally insonated. After postoperative day 10, the patient died. We were allowed by the patient's parent to use the patient information.

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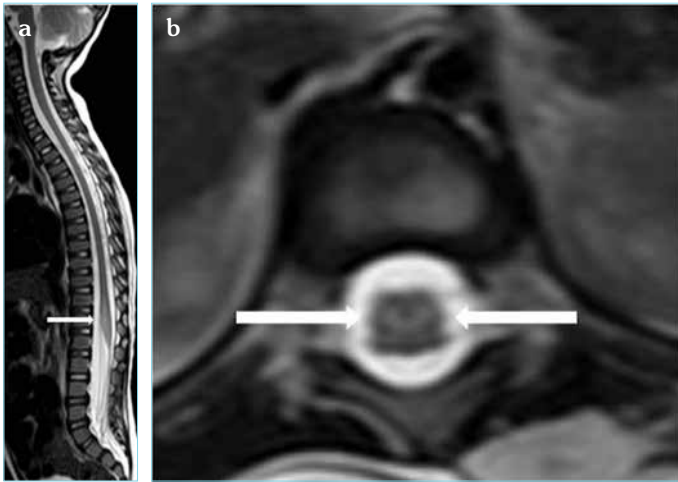


Figure 1. a, b. T2-weighted sagittal (a) and axial (b) images of the spinal cord show bilateral hyperintensities corresponding to the anterior horns of grey matter in the conus

DISCUSSION

Three major vessels arising from the vertebral arteries in the neck supply the spinal cord: 1 ASA and 2 posterior spinal arteries (4). The principal vascular supply to the spinal cord is by ASA, which supplies the anterior two third of the spinal cord. ASA is a single artery formed by the union of the branches of each vertebral artery. It receives most of its blood from 6–9 radicular arteries (4, 5). The most prominent thoracic radicular artery is the artery of Adamkiewicz, which is also known as the artery of lumbar enlargement. The artery of Adamkiewicz contributes to ASA between the T9 and T12 level in 75% of individuals, but it can be located above or below this level (4).

Surgical repair of thoracic and thoracoabdominal aortic aneurysms is the most common cause of SCI (1, 2, 5). The spinal cord ischemia rate following thoracic aortic surgery is reported to be as high as 29%, but it is commonly reported to be 10%–11%. Both open surgery and endovascular repair are associated with spinal cord ischemia. Coarctation of the aorta and its surgical repair is a less common cause of SCI (4). SCI following thoracic aortic surgery can be clinically apparent immediately following surgery or after a period of normal neurologic functioning (6). Delayed spinal cord ischemia has been reported as late as postoperatively at 27 days (7).

On postoperative day 1, the presented patient developed flaccid paralysis. Many factors can play a role in this complication, including systemic hypotension before, during, or after the procedure, aortic cross clamping resulting in decreased arterial perfusion and increased spinal canal pressure, and occlusion of important feeding arteries such as the artery of Adamkiewicz or other intercostal arteries owing to ligation, resection, or embolization. Episodes of systemic hypotension in many cases appear to be temporally associated with delayed onset of ischemia (6, 7). During surgery, the presented patient was not hypotensive or tachycardic. Between postoperative 6 and 8 h, the patient became hypotensive and tachycardic and developed flaccid paralysis. Therefore, SCI was suspected. T2-weighted spinal MRI

showed bilateral hyperintense involvement in the anterior horns of grey matter in the conus. The length of the involvement was 3 cm and extended through the level of T11-L1-consistent with ASA infarction.

Treatment of SCI is primarily supportive, ideally with intensive multidisciplinary rehabilitation. Given the rarity of diagnosis, there is no consensus concerning optimal treatment approaches. Thrombolytic therapy has been reported to be successful in only a few published case reports of SCI (4). A significant barrier to thrombolytic treatment is initial diagnostic uncertainty that can delay diagnosis beyond the treatment window, including the need to exclude aortic dissection and vascular malformations, which contraindicate the use of thrombolytic agents. Thrombolytic therapy for spinal cord ischemia remains to be investigated at present. Systemic corticosteroids have not been studied in acute ischemic injury to the spinal cord and are not recommended for acute ischemic stroke involving the brain; however, in rare cases wherein it is unclear if a patient has an ischemic or demyelinating spinal cord lesion, it might be reasonable to use corticosteroids, pending a firm diagnosis (8).

The long-term prognosis of SCI is poorly described in children. Small-scale case series have attempted to define the range of outcomes following SCI. The case fatality rate varies depending on the case mix included in the series but is often between 10% and 20% (1, 9). Patients presenting with cardiac arrest and acute aortic rupture or dissection and those with high-level cervical lesions are at the greatest risk of mortality. Among survivors, most have some improvement in functional deficits. Independent gait is achieved in 11%–46% of patients, whereas 20%–57% patients remain wheelchair bound. Poor prognostic factors for recovery include severe impairment at presentation, female sex, advanced age, and lack of improvement within the first 24 h (1, 3, 9, 10). Patients with residual deficits after SCI must often contend with various other complications, including bladder, bowel, and sexual dysfunctions; spasticity; and chronic pain.

CONCLUSION

Spinal cord infarction is a rare disorder caused by a wide variety of pathologies and is associated with moderate-to-severe disability and a high mortality rate, regardless of patient age. Additional research is needed to identify and evaluate strategies for the prevention of spinal cord ischemia associated with aortic surgery.

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