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Case Report

Rare presentation of acute anterior cord syndrome due to fibrocartilaginous embolism in a pediatric patient following minor trauma

Ali Alshehri[1], Rahaf Alanazi[2], Haifa Bin Dokhi[3], Hesham Alshalaan[4], Hanan Alqahtani[5], Nawaf Alhamied[3], Haya Aldabas[3], Khalid Althobaiti[1], Hamzah Alali[1], Naif Alharbi[2], Wael Alshaya[2]

Departments of [1]Pediatric Intensive Care Unit, [2]Neurosurgery, [3]General Pediatrics, [4]Pediatric Neuroradiology and [5]Intervention Radiology, King Abdullah Specialists Children Hospital, Riyadh, Saudi Arabia.

E-mail: Ali Alshehri - alshehrial5@mngha.med.sa; *Rahaf Alanazi - alanazirfd@gmail.com; Haifa Bin Dokhi - bindokhiha@ngha.med.sa; Hesham Alshalaan - shalanh@ngha.med.sa; Hanan Alqahtani - alqahtaniha3@ngha.med.sa; Nawaf Alhamied - nawaf.alhamied@gmail.com; Haya Aldabas - haya.aldabas@gmail.com; Khalid Althobaiti - k.albarrag@hotmail.com; Hamzah Alali - alaliha@mngha.med.sa; Naif Alharbi - alharbin189@hotmail.com; Wael Alshaya - waelalshaya@yahoo.com

***Corresponding author:**
Rahaf Alanazi,
Department of Neurosurgery,
King Abdullah Specialists
Children Hospital, Riyadh,
Saudi Arabia.

alanazirfd@gmail.com

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ABSTRACT

Background: Anterior spinal cord syndrome (ASCS) is an extremely rare condition defined as an infarction of the anterior two-thirds of the spinal cord. The type and timing of the clinical presentation, combined with the radiological findings, can provide a focused clinical picture of the severity and outcomes and direct the management plan. We present a rare case report of a pediatric patient with ASCS in acute settings due to a fibrocartilaginous embolism (FCE).

Case Description: We report a 10-year-old girl who was medically and surgically free. She presented with ASCS features 30 min after lifting her younger sister in her back. The clinical presentation consisted of bilateral lower limbs weakness 0/5 according to the medical research council's scale, weak anal tone, with pain and temperature significantly altered and absent up to T4 level. A diagnosis of anterior spinal infarction due to FCE was made after excluding the possible anterior cord syndrome etiologies. The management consisted of aspirin and extensive physiotherapy, and she significantly recovered over 1 month.

Conclusion: We report a rare case of acute presentation of a pediatric patient with ASCS due to FCE. The timing of a diagnosis affects clinical results. Correlating the radiological findings to the clinical presentation can narrow the differential diagnosis. The literature on the management of these cases is lacking. Animal studies reported a trial of medical therapy.

Keywords: Anterior cord syndrome, Case report, Fibrocartilaginous embolism, Pediatric, Spine trauma

INTRODUCTION

History and definition

Anterior spinal cord syndrome (ASCS) is a rare disorder that accounts for only 8% of all myelopathies and not more than 1% of all strokes. It was first described in 1909 by Spiller in a patient with anterior spinal artery (ASA) thrombosis who was noted to have an infarct at autopsy in the anterior part of the spinal cord, extending from C4 to T3. In 1966, Garland *et al.*[7] reported that it can result from

decreased perfusion pressure. Mosberg *et al*., 1954; Weisman and Adams, 1944 described a local interference with the spinal cord blood supply.[11,19] It is currently defined as an uncommon neurologic condition caused by ASA occlusion. The ASA supplies the anterior two-thirds of the spinal cord. As it runs along the entire length of the anterior surface of the spinal cord, occlusions can lead to reduced blood flow that eventually results in spinal cord infarction-resulting in neurological deficits manifest below the level of insult due to the spinal cord tract's anatomical distribution.[22] Fibrocartilaginous embolism (FCE) is an underrecognized cause of spinal cord infarction, accounting for 5.5% of cases.[9,13] The mechanism of FCE is unclear. However, the most common hypothesis is the rupture of the nucleus pulposus and the migration of the fibrocartilaginous into the vertebral vein or artery after trauma. The preceding event could be as minor as physical exercise or increased axial pressure by lifting a heavy object.[4,9,21]

CASE PRESENTATION

History of presenting illness

A 10-year-old previously healthy girl presented with acute onset paraplegia and loss of bladder and bowel function, which began approximately 30 min after lifting her. She was playing with her cousin (trying to lift each other on their backs), which led to hyperextension of their back.

Physical examination

Initial examination on admission revealed pupils equal and reactive to light at 2 mm, cranial nerves were intact with everyday speech, language, and visual fields, full strength (5/5) in the upper limbs, complete motor paralysis (0/5) in the lower limbs, decreased sensation to pinprick and temperature "cold" at the level of T4 and below, but intact proprioception and vibrations, and weak anal tone.

Investigations

Urgent unenhanced thoracic and lumbar spine computed tomography (CT) showed L5 bilateral spondylolysis without associated spondylolisthesis. It also revealed a mild posterior disc bulge at the level of L4–L5 L5–S1 without associated neural foraminal narrowing or spinal canal stenosis. These findings did not explain her presentation. Thus, a cervical, thoracic, and lumbar spine magnetic resonance imaging (MRI) [Figure 1] demonstrated a long-segment abnormal signal involving the anterior segment of the spinal cord extending from T2 to T8 with corresponding restricted diffusion. The appearance and distribution suggest anterior cord syndrome (ACS) at the thoracic spine level. The patient was started on norepinephrine targeting a mean arterial pressure (MAP) of 80 and above for 3 days, then weaned off gradually. Laboratory work investigating the possible etiology was sent. Coagulopathy and risk of atherosclerosis were unlikely with a standard coagulation profile, D. dimer, and lipids profile. Moreover, lumbar puncture and cerebrospinal fluids analysis, microorganism meningitis polymerase chain reaction panel, and culture were all insignificant. Transthoracic echocardiography was also done, and it showed normal cardiac function and no evidence of aortic dissection. Therefore, a whole spine MRI [Figure 2] and 3D dynamic spinal magnetic resonance angiography were done on 3rd day of admission to look for vascular anomalies. There was a decrease in the vertebral body height at the posterior aspect of the level of T3, T4, T5, and T6. Schmorl's nodes were at the posterior aspect of disc spaces at the T2–T3, T3–T4, and T4–T5 levels. High T2-weighted signal intensity at the posterior capsular ligament at the disc spaces of T3–T4, T4–T5, and T5–T6 was detected.

Hospital course

No underlying abnormal arteriovenous malformation or aneurysm was observed. Spinal digital subtraction

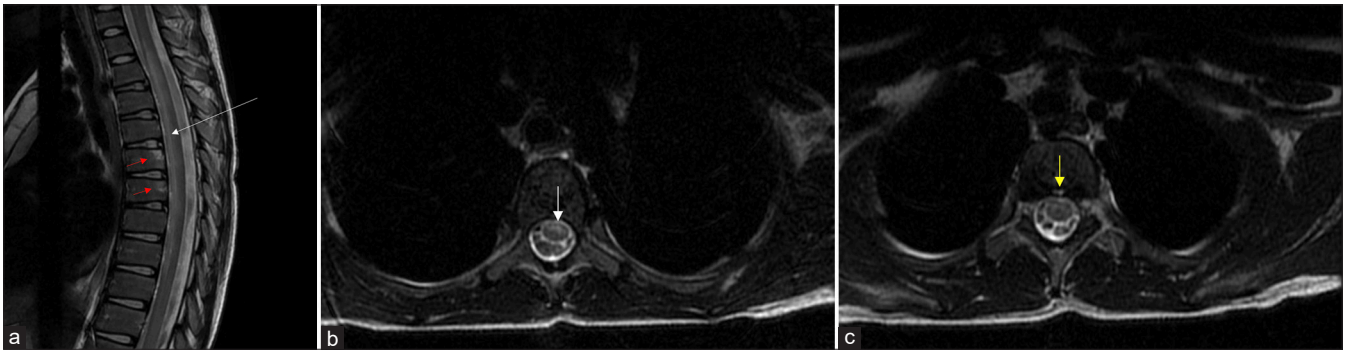

Figure 1: (a) SAG T2 SE images of the cervicothoracic spine demonstrate a slit-like high signal along the anterior horn of the spinal cord (white arrow). There is altered disc signal intensity at the T2/3 and T3/4 levels (red arrow). There is a subtle signal at the disc capsule. (b) AX T2 SE images of the cervicothoracic spine demonstrate a high signal along the anterior horn of the spinal cord (white arrow). There is a subtle signal at the disc capsule. (c) AX T2 SE images of the cervicothoracic spine demonstrate a high signal along the anterior horn of the spinal cord. There is a subtle signal at the disc capsule (yellow arrow). SAG: Sagittal,SE: Spin echo, AX: axial, T2 SE: T2-Weighted

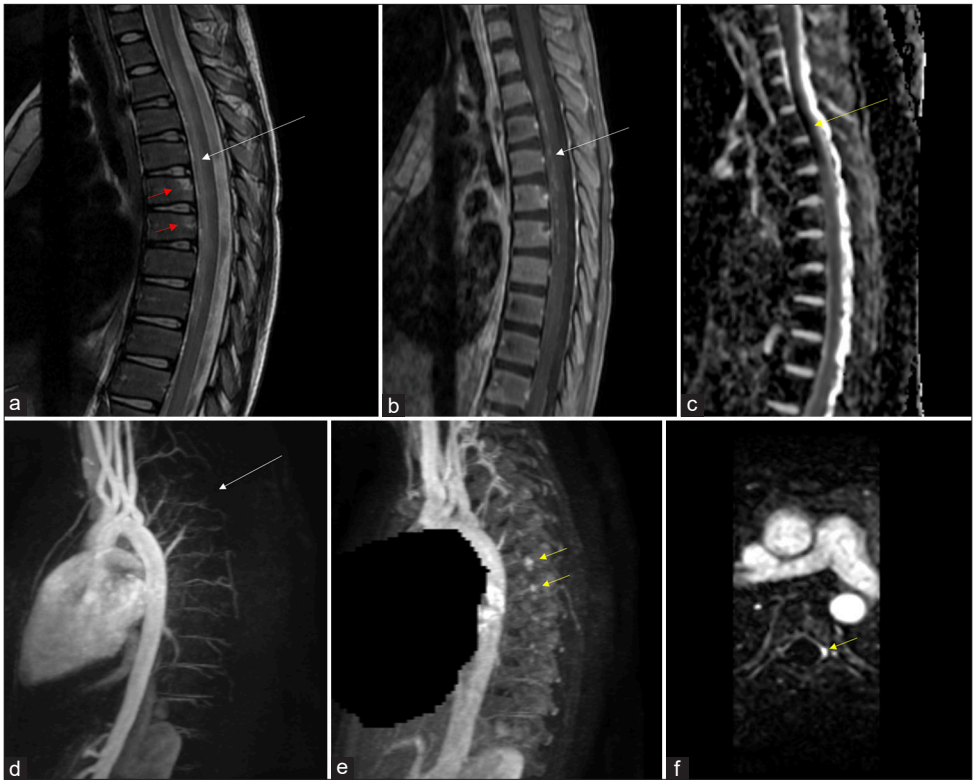

Figure 2: (a) SAG T2 SE images of the cervicothoracic spine demonstrate worsening cord anterior horn edema (white arrow). The vertebral altered signal is more evident suggestive of contusion (red arrow). (b) SAG T1 SE post gadolinium administration images of the cervicothoracic spine demonstrate heterogeneous anterior cord enhancement consistent with post ischemic re-perfusion (white arrow), (c) SAG ADC images of the cervicothoracic spine demonstrate restricted diffusion of the anterior cord consistent with ischemia (yellow arrow), (d) time-resolved – magnetic resonance angiography (MRA) sequences TRICKS 3T GE system in arterial phase demonstrate non-visualization of normal anterior spinal artery (ASA) in expected anatomic location (white arrow), (e) time-resolved-MRA sequences TRICKS 3T GE system in early venous phase with reconstructed axial multiplanar reformation (MPR) demonstrate non-visualization of normal ASA in expected anatomic location and contrast pool at left neural foramina consistent with congested venous plexus (yellow arrow), and (f) time-resolved-MRA sequences TRICKS 3T GE system in early venous phase with reconstructed axial MPR demonstrate non-visualization of normal anterior spinal artery (ASA) in expected anatomic location and contrast pool at left neural foramina consistent with congested venous plexus (yellow arrow). SAG: Sagittal, T2 SE: T2-Weighted spin echo, ADC: Apparent diffusion coefficient, MRA: Magnetic Resonance Angiography, TRICKS: Time-Resolved imaging of contrast kinetics, GE: Gradient echo

angiography [Figure 3] was performed for further confirmation. It showed no evidence of arteriovenous shunting or significant vascular injury in dissection and/or avulsion. Anterior spine artery could not be detected at the thoracic level. After excluding the common causes of ACS along with the typical history of sudden onset pain and rapidly progressive paraplegia following trivial trauma, a diagnosis of spinal infarction due to FCE was made. The patient was started on antiplatelet medication along with extensive physiotherapy and discharged on aspirin with close physiotherapy sessions along with outpatient follow-up in the clinic.

Follow-up after 1 month

At a follow-up after 3 weeks, the patient surprisingly ambulated with lower limb power 4 out of 5.

DISCUSSION

ASCS Incidence and prevalence

ASCS is an infrequent neurologic condition, accounting for approximately 1.2% of all vascular neurological disorders. In the review by Foo and Rossier,[6] out of 60 collected cases of ASCS, 26 were male, and 34 were female, indicating a slightly

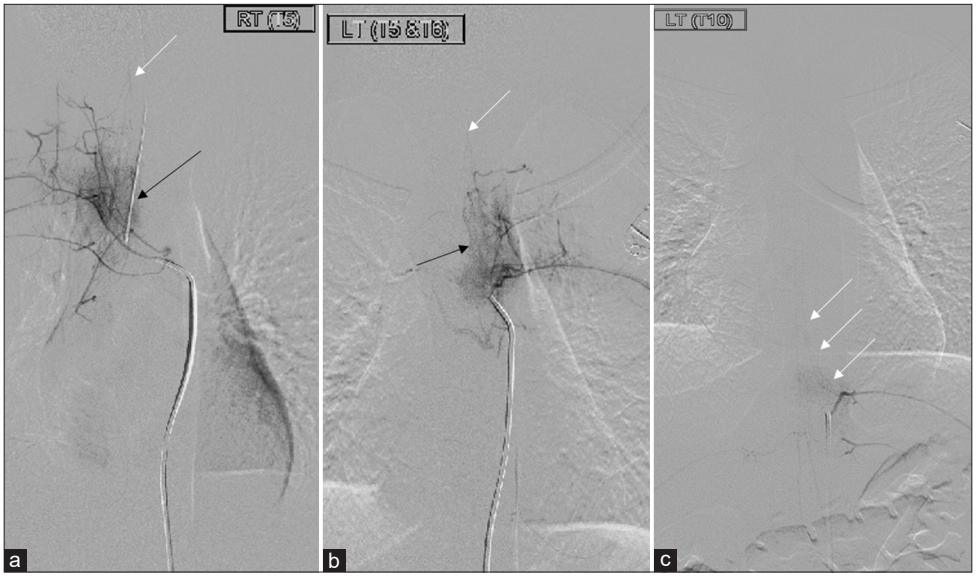

Figure 3: (a) Selected digital subtracted spinal angiogram images at T5–T6 level showed vertebral body hyperemia related to the trauma (black arrows). Neither vascular injury nor shunting was noted. A faint flow was seen at the upper thoracic T1 could represent the spinal artery (white arrows). However, at the targeted area T5–T6, there was absent flow (b) Selected digital subtracted spinal angiogram images at T5–T6 level showed vertebral body hyperemia related to the trauma (black arrows). Neither vascular injury nor shunting was noted. A faint flow was seen at the upper thoracic T1 could represent the spinal artery (white arrows). However, at the targeted area T5–T6, there was absent flow. (c) The artery of Adamkiewicz is seen at the left T10 level (white arrows).

higher prevalence in females for ASAS specifically, with age groups ranging from 5 years old due to infection to 76 with abdominal aortic neurectomy.[22] In a study by Salvador de la Barrera *et al*., spinal cord infarction primarily affected older individuals, with the average age of the patients being 59.3 years. The gender distribution showed a higher prevalence in males, with 66.7% of the cases (24 out of 36 patients) being men and 33.3% being women. The most frequent etiology was idiopathic, accounting for 36.1% of the cases. Other causes included complications from aortic surgery (25%), systemic atherosclerosis (19.4%), and acute perfusion deficits (11.1%), such as hypovolemic shock or cardiac arrest.[16]

ASCS characteristics

It is generally described as motor deficits below the level of injury and impairment of pain and temperature while preserving vibration and proprioception sensation. By understanding the anatomy, the clinical presentation can point to the area of injury.

ASA origin

The ASA forms from the bilateral vertebral arteries at the foramen magnum and runs as an uninterrupted artery within the anterior median sulcus of the spinal cord to the conus medullaris. Radicular arteries enter the spinal canal through the intervertebral foramen and primarily supply the nerve roots; however, some anastomoses contribute to the ASA. The largest of these radicular arteries is the artery of Adamkiewicz, also known as arteria radicularis magna, which most commonly arises from a left intercostal artery between segments T9 and T12 but can vary anatomically. The ASA branches into small sulcal and penetrating arteries that enter the body of the spinal cord.[10]

ASA branches and supply

The ASA supplies blood to the spinal cord's bilateral anterior and lateral horns and the bilateral spinothalamic and corticospinal tracts. The anterior horns and corticospinal tracts control the somatic motor system from the neck to the feet. The lateral horns, spanning levels T1 to L2 of the spinal cord, comprise the neuronal cell bodies of the sympathetic nervous system. The spinothalamic tracts relay pain, temperature, and sensory information.[20]

ASCS evaluation

History and physical examinations are essential for timing diagnosis. When the patient presents with symptoms and signs similar to those described above, ruling out vascular causes could be the top priority for establishing

the differential diagnosis. CT angiography can be done in patients with abdominal pain and hypotension to rule out aortic dissection; CT spine can be used when suspecting spine trauma or vertebral artery injury. MRI is the gold standard with the following sequencing: T2-weighted (T2W) sagittal, T2W axial, short-tau inversion recovery sagittal, diffuse weighted images (DWI), and contrast-enhanced T1W sagittal sequences.[20]

ASCS management

The management is mainly supportive, limiting underlying causes and preventing future complications. Steroids can be given in cases of edema. Some studies have described the benefit of lumbar drain, increasing MAP from 60 to 90 or 100 mmHg.[17]

ASCS outcomes and prognosis

The reported etiologies consist of idiopathic with the best motor recovery rates, with 92.9% of these patients regaining some motor function. ASCS caused by post-infectious myelopathy or vaccination had an 88.9% recovery rate, while cases due to ASA occlusion and aortic lesions had lower recovery rates of 33.3% and 20%, respectively. The data highlight the variable outcomes based on the underlying cause of the syndrome.[22]

Mortality during the hospital stay was 22.2%, and most survivors had significant long-term disabilities, with 57.1% requiring a wheelchair.[16]

Fibrocartilaginous embolism (FCS) history

Although FCE has been widely reported among animals, especially dogs, it is a rare cause of human anterior cord infarction.[5] The first FCE case reported was in 1961 for a 15-year-old boy who had paraplegia after a minor trauma while playing basketball in school, followed by death 3 h from the event.[13] According to a study that included a sample of 168 ACS cases, only nine were secondary to FCE. The most common etiologies reported were vascular anomalies or postoperative complications.[1] FCE has a bimodal pattern with a peak incidence at 22 and 60 years of age.[9] The underlying pathological mechanism of FCE varies among the two age groups. At a younger age, FCE is attributed to the herniation of intervertebral disk nucleus pulposus components that migrate into the spinal arteries, causing embolism. However, among the older age group, it is likely due to degenerative changes in the cartilage and osteoporosis.[9]

FCS incidence and prevalence

Since 1961, only 25 cases of ACS secondary to FCE have been reported in children aged <18 years old, with female predominance (16 female vs. nine male).[21] According to the latter study, the mean age at the presentation time was 14 years, unlike our case, which was 10 years old. Even though intense exercise and lifting heavy weights are considered the most common triggers, it has been reported that half of the patients completed their exercises generally without any symptoms.[5] Minor trauma and Valsalva maneuver have also been reported as the trigger of FCE.[6] Similarly, trivial trauma was the only trigger in our patient's case as she was playing with children, trying to lift each other on their backs, proceeding with the symptom's onset.

Diagnosing FCS as a cause of ASCS

Such cases are infrequently experienced in pediatric emergency departments. Therefore, confirming the diagnosis is challenging as it resembles other conditions with ascending paralysis, such as Guillain–Barre syndrome, transverse myelitis, and other neurological diseases.[14] In our case, she presented with sudden back pain after 30 min of minor trauma while she was playing, which progressed to acute weakness and a tingling sensation that prevented her from walking within hours. In consistency with the literature, our case had developed paraplegia and hypotonia and the absence of reflexes in the lower limbs, along with decreased pain and temperature sensation below the level of T4.[2] Intact proprioception and vibration sensation below the mentioned level were suggestive of ACS. Similar to our case, bowel and bladder incontinence or both are widely described in the literature on presentation time.[3,12] In our patients, CT showed the presence of Schmorl's nodes, which has been reported as a characteristic of FCE.[8,18] Schmorl's nodes represent a herniation of the nucleus pulposus through the bony end plate and cartilage into the adjacent vertebrae. It is considered a common incidental finding in adults that does not cause pain or symptoms, but it is rare in the pediatric population.[15] These nodes are caused by either direct trauma to vertebra intervertebral disc degeneration or metabolic or infectious processes. Moreover, it is hypothesized that it could be due to ischemic necrosis or abnormal vascular development, such as AV malformation, which has been ruled out in our case.

CONCLUSION

This case report highlights the rare presentation of ASCS due to FCS in a pediatric patient following hyperextension of the back. Our management plan was initiated with a spinal cord injury protocol that includes inotropic support targeting a MAP of 80 and above to prevent further spinal cord injury. She was also started on antiplatelet therapy and extensive physiotherapy. Favorable outcomes were achieved during 1 month of follow-up. Limited data from human studies are published in the literature. Further studies are needed to

support the appropriate and timing of medical management for these cases.

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