American Scientific Research Journal for Engineering, Technology, and Sciences (ASRJETS)
ISSN (Print) 2313-4410, ISSN (Online) 2313-4402

http://asrjetsjournal.org/

Unusual Cases of Spinal Cord Infarct Secondary to Fibrocartiliginous Embolism in Minor Injuries or Exercises; Review of Case Reports and Literature

Yasir Rehman*

MD, MSc (Neuroscience), PhD Candidate, Health Research Methodology, McMaster University, Ontario, Canada
Email: drrehmany@yahoo.ca

Abstract

Spinal cord infarction, secondary to fibrocartiliginous embolism (FCE), are reported after a major traumatic injury, or surgeries that relate to the spinal cord and the back of the upper body, but are rarely reported with the activities that are usually minor. Spinal cord infarct, secondary to FCE in minor trauma or activity is quite common, and bears potentially devastating consequences. It is postulated that an acute increase in intra discal pressure embolizes disc debris retrogradely to the spinal artery, leading to spinal cord infarction. The main objective of this literature review is to present important cases of spinal cord infarction, secondary to FCE, during routine activities. Using the Ovid Medline, our search gave us eighteen published articles on FCE induced spinal cord infarcts in minor or routine activities. A clinical diagnosis of FCE would be desirable for many important reasons, as no single non-invasive investigation has been proven effective. This literature review is done to acquire a detailed knowledge about pathogenesis, clinical features, investigations, treatments and prognosis for FCE.

Keywords: Fibrocartilaginous Embolism; Spinal Cord Infarction; Intervertebral Disc; Magnetic Resonance Imaging.

---

* Corresponding author.

1. Introduction

Fibro-cartilaginous embolism (FCE) is an unusual cause of the spinal cord and brain stem ischemia, mostly diagnosed at autopsy [1]. The ischemic myelopathy, secondary to FCE, has been firmly documented following sport injuries, falls, accidents, neoplasm, infections, aortic surgery or atherosclerosis of the aorta and spinal arteries [2, 3]. Patient with hypertension, obesity, diabetic mellitus, coagulopathies, episodic hypotension, and degenerative disease of the spine, are at an increased risk of developing a spinal cord infarction [4,5].

FCE causes spinal cord infarction and bears potentially devastating consequences and often considered as a diagnosis of exclusion [6]. Previously, References [7,8,9], did the literature reviews; however, they mostly focused on major traumas, or in certain cases, the mechanisms of injury was not reported. This is the first review that focuses on the FCE related to the routine physical or recreational activities. The main objective of this literature review is to focus and develop a better clinical understanding about FCE induced spinal cord infarct. In majority of the cases, patient did not have predisposing factors. A better understanding about FCE- spinal cord infarct pathophysiology and clinical presentations in similar clinical situations may help to identify and contribute to our understanding of this disorder and its natural history.

2. Data synthesis and acquisition

As the first case of FCE was reported in 1962, therefore we searched literature from 1961 to 2016 using the Ovid medline data base. The search term used were Fibrocartiligenous embolism, spinal cord infarction, case report, case series, rare disease, trauma, epidemiology, pathogenesis and diagnosis. The main focus was on the studies that reported FCE in minor traumatic events or FCE resulting during the routine activities. Articles that reported FCE due to herniated disc surgery, FCE during the heavy exercise such as weight lifting, surfing, falls and any pre-existing spinal or vertebral column pathology were excluded. Based on the inclusion and exclusion criteria, the database yielded 18 case reports.

3. Summary of the included cases

Table 1 gives the brief summary of the cases.

Reference [10] reported a 16-year-old girl that was admitted into the hospital after doing a “flic-flac” sport injury. In hospital admission, she had pain in her cervical spine but non-neurological deficits or signs of amnesia. Plain radiography showed no injury to the cervical or thoracic spines. She progressively developed quadriplegia with sensory and motor deficits in her arms and legs with dermatomal pattern. High-dose methylprednisolone bolus was administered followed by infusion. MRI showed minimal edema and swelling of the upper cervical spinal cord. One year of long and intensive rehabilitation led to complete recovery without any continuing neurological or motor deficits.

Reference [11] presented a 58-year-old man with an acute, but progressive onset quadriparesis following the neck rotation. The initial CT of the brain was normal. At the ER, his muscle strength in all limbs partially improved. Three hours later patient developed acute urinary retention and received urinary catheterization.

Table 1: Summary of the cases with FCE and spinal cord infarcts

Reference	G/Age	Mechanism of injury	Site of injury	S/S	Radiological finding	Rx	Outcome
Beck 2000[12]	F/ 16	Flic flac dancing	Hyperextension of C -spine	Progressive quadriplegic		methyl prednisone	complete resolution
Chen 2010[5]	M/ 58	Exercise- neck near vertical angle around 360°	C-spine	sudden onset of weakness over left upper extremity, quadriparesis and urine incontinence	Mild indentation of the thecal sac. high intensity edema of "H" gray matters of C3 spinal cord	aspirin and IV dexamethasone	improved significantly
Cuello 2014	M/ 50	Bending down to untie his dog	C3- T1	Sudden onset back pain, quadriparesis,	Hyperintensity from C3 to T1, consistent with early sub acute cord infarct.	Dexamethasone and analgesics	Partial recovery
Davis 2000	F/ 6	Came out of car	T5-T10	Progressive leg weakness and diminished pain, and proprioception loss, loss of anal tone and urinary incontinence	multiseptated syrinx between T5 and T6 to T10, increased signal extending from T10 to conus medullaris consistent with the area supplied by the anterior spinal artery	Dexamethsaone	Minimal/ Partial recovery
Dickerman 2006[13]	M/14	sit-ups	central cord syndrome (C-spine)	Quadriparesis, proprioception	negative for any mass effect	corticosteroids and rigid collar	Complete resolution
Han 2004[18]	M/16	Lifting Exercises, squat exercise	T7-T8	Dull lumbar back pain, weak walk, low sphincter tone. Left leg flaccidity, hemiparesis, absent quadriceps and achilles reflexes, absent sensation to	T7 to T8 spinal cord level consistent with ischemia	high-dose steroids	neurologic improvement

				pinprick bilaterally below the T6 Anteriorly and T8 level posterior.			
Ho 1980[10]	F/22	Dancing	C3 to T1 and was soft and dusky.C5-6 on histopathology	Excruciating pain in the back of her neck with radiation down the spine. No weakness, paraesthesia but becomes stupors	Nil	dexamethasone therapy	Died on nine days after admission.
Kase 1989[20]	F/ 23	Working in office	C2-3, medulla oblongata and brain stem	Sudden onset back pain, progressive neurological deficits, brain stem signs,	Initial CT normal, Autopsy→ C2-3 and medulla and brain stem infarct; FCE in ASA and Basilar artery	ventilator	Respiratory arrest and death.
Millichip 2007	M/ 63	Bending over	C6 to T3	Acute onset numbness, weakness hand and legs bilaterally, absent anal tone, diminished reflexes in legs bilaterally, reduces pain and proprioception	T2 signal in the anterior cord from C6 to T3 with narrowing of the right intervertebral foramina and cord compression at C5–6	Antiplatelet therapy with aspirin and clopidogrel was continued for prevention of recurrent stroke	No recovery with discharge to spinal rehabilitation
Moorehouse 1992[11]	F/63	Straining at stool	T10-11	Thoraco-lumbar pain, progressive weakness bilateral lower extremity weakness and reduced ankle jerks. Pain and temperature appreciation were	Autopsy showed softening in the T 10-L 1 region	Nil	Fatal cardio respiratory arrest.; Death

				reduced below T10			
Nance 2007	F/ 14	In school	C1-C7	Sudden onset of breathing difficulty, and arms and leg movement, loss gag reflex, facial muscle weakness, absent cough reflex, absent tendon reflexes in all limbs	T2 hyper intensity in the lower medulla and anterior cord from C1-C7 and at T3.follow up MRI showed signals from C2 to C5 and at T3 consistent with anterior spinal artery infarct.	steroids, plasmapharesis, and IVIG,	Minimal/ partial recovery
Redmond 1983[14]	M/ 15	Dancing	C5	Neck pain, tingling, weakness, flaccid quadriparesis	Myelogram showed mild enlargement 55% of the interpedicular distance at the C5 level.	dexamethasone	residual weakness of the left arm
Reisner 2013 [15]	F/ 8	Dancing at ballet	C2-T1	paraesthesia and numbness in her hands, unable to walk	T2 hyperintensity at C2–T1 central cord, diminished T2 hyperintensity in the C3–4 disc space	intravenous Solu-Medrol	minimal recovery in legs (ASIA Grade C)
Spengos 2006 [16]	F/ 30	Ballet dancer	Cervical spine	proximal paresis of both arms	Spinal cord infarction in the anterior spinal territory at the C3–C6 spinal cord level.	corticosteroids	Complete resolution
Thone 2007	M/ 45	physical exercise	C5- T10	Progressive gait disturbance, leg numbness, reduced strength in the legs bilaterally, diminished reflexes bilaterally, urinary retention and faecal incontinence, reduced sensory	Hyperinetse signal extending from C6 to conus, vertebral body infarctions C5–7/Th8–10	aspirin (100 mg/day) and intensive physiotherapy	no improvement

				levels below Th10, reduced pain, temperature sensation and proprioception.			
Toro 1994[19]	F/16	Milking a cow	Sacral region	Acute low back pain, progressive leg weakness, flaccid paraplegia and sensory loss at L1, areflexia	Fibro-cartiiganous emboli causing infarct at lumbro-sacral region		Death
Tosi 1996[17]	F/16	Handstand	Sacral	sudden back pain radiating down to her upper thighs, Touch, pain, and temperature appreciation were reduced on the left leg below the knee	enlargement of the distal cord and conus medullaris Five months later pronounced atrophy of the lower spinal cord.		Complete paraplegia
Yousef 1998[6]	F/14	Bending forward	Thoracic	sudden back pain and progressive bilateral leg weakness leading to complete paraplegia	Enlargement of the thoracic cord between T5-7	Chiropractic manipulations	Death

Neurological examination was normal, except for the variable motor quadriparesis. The deep tendon reflexes were absent over four limbs. Sensory examination unfolded normal pinprick, light touch and vibration test. Cervical spine MRI showed mild indentation of thecal sacs, edema of "H" gray matters at C3.The patient was given four doses of aspirin, IV dexamethasone and started to show muscle power, which improved gradually, and on day 3, the sphincter function began to resume. The follow-up C-spine MRI correlated to the clinical symptoms of the patient with old C3 ischemic infarction with residual irregular longitudinal hyper-dense lesion and absence of oedematous changes.

Cuello J and his colleagues 2014 [12] reported a case of 50-year-old man with sudden sharp, stabbing right shoulder pain, when risingup form a bending position to untie his dog. On examination patient had distal bilateral arm and leg paresis. Sensory examination was significant for reduced sensory finding distal to T5, intergluteal folds and genitalia. Patient's past history was significant for Parkinson disease controlled with carbidopa/ levodopa. Both MRI's showed T2 hyperintensity, consistent with C3 to T1 early sub acute cord infarct. Brain imaging, Echo EKG, CSF analysis, culture and serology were normal. Patient was admitted in neurology ICU and adminstered dexamethasone 10 mg and analgesics. Patient did not exhibit neurological improvement and was was discharged to spinal cord rehabilitation service for further management.

Reference [13] reported a case of a 6 years old girl with a 1 week history of episodic pain, who actively participated in "cart wheel" gynamstics. On the morning of the presentation, patien came out of her car and while standing, her legs began to get weaker and she fell onto the ground. Neurological examination was significant for the loss of pain and temperature sensation to T-11, loss of anal tone and urinary incontinence. Patient was administered dexamethsaone. The MRI showed multiseptated syrinx between T5 and T6 to T10. Second MRI two days latter showed increased signal extending from T10 to conus medullaris consistent with the area supplied by the anterior spinal artery. Two months after the discharge patient had regained minimal strength in her left quadriceps muscle only.

Reference [14] presented a 14-year-old boy with acute onset progressive paresis of the upper extremities and gait difficulty following sit-ups. The plain radiographs, CT and MRI within 24 hours were normal. On day three, the strength in his arms and legs decreased. Lower extremities were spastic with difficulty to walk secondary to reduced proprioception. The patient was placed on Decadron (dexamethasone) 4 mg every 6 h with rapid taper over five days. The patient made significant improvement over the next 24 h with 3/5 strength in biceps and deltoids and 4/5 in remaining upper extremities and gait improved. Patient was discharged on Decadron, and was placed in a rigid collar. A three month repeat MRI was normal and the patient status improved to normal.

Reference [15] presented a case of a 16-year-old high-school student who woke up with dull lumbar pain with paraesthesia radiating down both posterior thighs and lateral aspects of his calves. The night prior to the pain, he had performed lifting exercises, including a squat exercise. He had bilateral leg flaccid paresis, absent quadriceps, Achilles reflexes and plantar responses. The sensory examination was remarkable for absent sensation to pinprick bilaterally distal to T6 level anteriorly and T8 level posteriorly. The MRI 2-weeks post admission showed increased intensity at the T7 & T8 spinal cord consistent with ischemia. Four weeks post, he had some improvement on strength and was able to appreciate pin pricks. Eight months post admission, he

ambulated independently, without assistive devices, but knee and ankle reflexes remained absent.

Reference [8] presented a case of a 22 year old woman who developed excruciating pain in her neck radiating down to her spine while dancing. Initially, patient had no limb weakness, but in the next 15 minutes she became breathless, and stuprous. At the admission desk, she was comatose and was unable to breathe without a respirator. Her pupils were in mid position, unresponsive to light stimulation, with absent oculocephalic responses. She was areflexic and had flaccid quadriplegia. A CT scan revealed severe brain edema and marked compression of the ventricular system. The patient died despite of dexamethasone treatment and resuscitation. Autopsy showed swollen cord from C3 to T1 and was soft and dusky consistent with FCE.

Reference [7] presented a case of 60 year old woman with sudden onset lumbosacral pain while shopping and walking. The pain radiated to lower extremities bilaterally and collapsed in the shopping mall. On examination, she had hyperaesthesia distal to T7 and abdominal reflexes could not be elicited. The musculature of the lower limbs was hypotonic, and the bilateral loss of reflexes in the region of L-2 to S-2 were found. The paraplegia continued, fever and urinary incontinence persisted, and her blood pressure gradually decreased. The patient died on the 18th day of her illness in spite of antibiotic and symptomatic therapy.

Reference [16] reported a case of a 23 years old female working in a bank who complained of sudden occipital headache with progressive neurological deficits. On her way to the hospital, she developed bilateral arm anf leg weakness, followed by a respiratory arrest. She was put on mechanical ventilation. On examination patient demonstrated brain stem deficits bilateral facial weakness, full visual field loss, extra ocular movements, discrete up beating nystagmus in upward gaze, and respiratory paralysis. Patient also demonstrated quadriparesis, pain distal to C3, absent joint position sense. Patient died on the 12th admission day. Autopsy showed FCE in the anterior spinal artery, C2-3, medial medulla and brain stem infarct.

Reference [17] reported a case of 63-year-old diabetic and hypertensive patient, who developed acute onset chest pain, numbness, and progressive weakness while bending over. Over the next few hours, patient developed urinary retention. Neurological examination was significant for reduced bilateral hand and legs strength, absent anal tone, diminished reflexes in arms and legs bilaterally. Pain, temperature and proprioception and vibration sensations were reduced below the C7 and T4 respectively. MRI of the spine showed abnormal T2 signal in the anterior cord from C6 to T3 with narrowing of the right intervertebral foramina and cord compression at C5–6. Despite of the antiplatelet therapy with aspirin and clopidogrel, neurological deficits persisted unchanged and the patient was transferred to the rehabilitation center.

Reference [9] reported a case of a 63-year-old woman, who developed sudden onset severe thoraco-lumbar pain while straining at stool. The pain radiated down both lower extremities in a sciatica distribution and was accompanied by severe weakness. Neurological examination revealed an alert patient with grade 3/5 bilateral lower extremity weakness and reduced ankle jerks with unobtainable plantar responses. Pain and temperature appreciation were reduced to T10. Anal sphincter tone was reduced. Patient died of respiratory compromise. Autopsy showed anterior spinal artery FCE in the immediate vicinity of the area of infarction. Additionally, a small intramedullary vessels contained fragments of cartilage.

Reference [3] a cases of a 14-year-old female with sudden onset breathing difficulty, bilateral arms and leg weakness, painful burning sensation radiating down to her neck and back. Patient had the loss gag reflex thus was intubated. On examination patient had facial muscle weakness, absent cough reflex, absent tendon reflexes in all limbs with minimal withdrawal to touch. Patient was treated with the steroids, plasmapheresis, and IVIG, with noy clinical improvement. The first MRI showed hyperintensities at the lower medulla and anterior cord from C1-C7 and at T3, whereas second MRI showed hyper intensity in the anterior cord from C2 to C5 and at T3 consistent with spinal cord infarction in the anterior spinal artery territory. At 18 months, patient gained minimal movement of her head, right hand, and decreased pinprick sensation and patient in the C3 dermatome and below and remained incontinent.

Reference [18] presented the case of a 15 year old boy dancing to the disco, when he suddenly developed neck pain, tingling with gradual quadriparesis, reduced reflexes in all limbs and bilateral extensor plantar responses. He had lost pain and temperature sensation from C4-L1 on the right, and from C4-T1 on the left side. Cervical spine myelogram showed mild enlargement of the lower cervical cord at C5. He was treated with dexamethasone, and over the next two weeks the strength in his leg returned. Eight weeks latter, she continued to had severe weakness in her left arm. The patient had moderate wasting of the intrinsic muscles in both hands, and moderate weakness of the finger extensors. Reflexes were normal in the right arm, but the left biceps jerk remained absent with an inverted supinator jerk.

Reference [19] presented a case of a 8-year-old girl dancing to ballet when she developed sudden posterior neck pain, paraesthesia and numbness in her hands. She had difficulty breathing and walking. The next day, she had significantly reduced strength in her arms and legs; however the patient had intact sensation to light touch and vibration, and diminished sensation to pain and temperature. MRI showed T2 hyperintensity in the C2–T1 central cord and diminished T2 hyperintensity in the C3–4 disc space. She was admitted into the ICU and intravenous Solu-Medrol was started. On day 15 she regained minimal movement in her lower extremities. At her 3-month follow-up, the patient's condition had improved and she was ambulatory without assistance.

Reference [20] presented a case of a30-year old female ballet dancer with sudden onset neck pain radiating to both her shoulders. She had progressive severe weakness and numbness of both arms, as well as inability to empty her bladder and bowel voluntarily. On the next day, weakness in her arms partly remitted but numbness progressed to her trunk and legs. The MRI scans of the cervical spine showed hyper-intensities on T2 weighted images at the C3–C6 levels of the spinal cord. Her symptoms began to reduce with corticosteroid treatment. A repeated MRI revealed reduced signal intensity on the corresponding segments with apparent diffusion coefficient (ADC)-mapping sequences, establishing the ischemic nature of the lesion.

Reference [21] reported a case of a 45 year old man with sudden belt like chest pain that started after the physical exercise and frequent valsulva maneuvers. Patient developed progressive gait disturbance and leg numbness. On examination, patient had reduced strength in the legs bilaterally, diminished reflexes bilaterally, absent babinski and abdominal reflexes and urinary retention and faecal incontinence. Sensory examination was significant for reduced sensory levels below Th10, reduced pain, temperature sensation and proprioception. Patient condition worsened despite of treatment with I/V steroid and on day 5 patient developed complete

paraplegia and sensory below T4, urinary and bowel incontinence. Initial MRI showed demyelinating or ischemic changes from T7-9, whereas follow up MRI showed hyperinetse signal extending from C6 to conus, and vertebral body infarctions at (C5–7/Th8–10). Secondary prevention with aspirin (100 mg/day) and intensive physiotherapy was started; however, 8 weeks later no improvement was accomplished.

Reference [22] reported a case of a 16 year old girl who developed sudden onset paraplegia and an acute sharp, low back pain radiating down to the thighs while she was milking a cow. On examination, the patient had flaccid paraplegia with complete sensory loss distal to L1 and areflexia in the lower limbs. Patient had urinary retention with loss of perineal sensation. Patient was admitted with transverse myelitis diagnosis. The spinal radiographs showed old L1 and L2 fracture vertebral bodies with minimal plate displacement and sclerosis. Over the next several days, patient had mild improvement but died 6 weeks post because of the bronchial aspiration.

Reference [23] presented a case of a 16 year old girl who developed back pain after doing a handstand for fun. She developed progressive bilateral foot weakness. Touch, pain, and temperature sensations as well as proprioception were reduced below T12. She had urinary incontinence. Knee and ankle reflexes were elicitable. Plain spinal radiographs showed Schmorl's nodes in the thoraco-lumbar region with no evidence of fracture. MRI scan 48 hours later showed enlarged distal cord with a non-enhancing area of increased signal intensity on the T2 weighted sequence. Conventional and computed myelography, spinal angiography was non-contributory. The motor conduction velocity was unobtainable, and EMG showed complete denervation in the lower limbs. On day 10, MRI showed area of increased signal intensity in T2 sequences with poorly defined areas of increased signal intensity appeared in the mid dorsal region. A follow up MRI five months later showed pronounced atrophy of the lower spinal cord and intra-spongious disc material in the T-12 vertebral body.

Reference [24] reported a 14-year-old previously healthy female, who experienced a sharp pain in her upper back while bending forward to pick up an object. Patient was initially admitted; however she left the hospital against medical advice. She had taken chiropractic treatments. During her third chiropractic treatment, patient had a cardiopulmonary arrest and died. The autopsy revealed mild swelling of the mid thoracic area; however, histological examination was significant for area of necrosis in the T5 to T7 segments of the spinal cord, involving anterior 2/3 and small portions of the posterior column of the spinal cord was involved. The anterior spinal artery at T4-7 was occluded with multiple fragments of cartilage.

4. Results

In this literature review most of the patients were young adolescents or young adults, which correspond with [19] study. Reference [19] reported bimodal age distribution with peaks in young adulthood (mean 22 years) and late middle age (mean 60 years). Hypermobility of the cervical cord in children is postulated to increase the susceptibility to hyperflexion, hyperextension, and distraction mechanisms [12]. All events in this literature review followed hyperextension injury and valsalva like mechanisms during the activities, such as dancing, exercise, bending, sit ups, straining while defecating, milking a cow and hand stands or when bending forward. One female developed pain in her back when working; however, no detail is provided if she was involved in an

activity which could have created a valsalva like mechanism. None of these cases had pre-existing conditions except for one case that had an old vertebral fracture.

In this literature review, five patients had FCE during dancing, four during routine exercises, three patient developed symptoms when in bending position, one had FCE from milking a cow, straining during the defecation and two had no cause, other than sitting in her office or coming out of the car. Spinal cord manipulation prior to the onset of the symptoms was not identified in any of the patients. The common site for the FCE infarct was cervical spine followed by thoracic spines. Cervical-thoracic involvement was more compared to throaco-lumbar site (SEE TABLE# 1). Seven patients (39%) had isolated cervical cord involvement. Few patients had more than one spinal segment involvements. One patient had cervical, medulla and brain stem lesion. In this literature review there was a female preponderance, approximately 61%.

All patients in this review had progressive development of symptoms after the acute onset. In most cases, MRI showed swelling or oedematous changes in the cord consistent with ischemic changes. Eleven patients (61%) were treated with steroids, off which five had complete or maximal recovery, five had minimal or no recovery and one died. Only one patient was treated with intravenous plasmapharesis and IVIG in combination with steroids. Three patients were treated with aspirin or clopidogrel, among those two had partial or minimal recovery and one had complete recovery. Over all, five patients died, five had complete or significant recovery, eight had minimal or no recovery. In all five deceased cases the FCE was due to arterial circulation.

5. Discussion

Spinal cord ischemia, as compared to cerebral infarct, is low and accounts for 1- 1.2% of all strokes [20]. The low prevalent rate of spinal cord infarction could be attributed to the rich collateral supply of the spinal cord.10, 20 FCE was once thought to be a rare and fatal event; however FCE is more prevalent than thought and not typically fatal [15].

The exact mechanism and node of entry of embolic material from the intervertebral disc into spinal vessel is lacking [15]. It is speculated that in children and adolescents, vascularity of the nucleus pulposus normally persists [10]. During the activities, the sudden flexion or extension increases the intra discal pressure transiently, which pushes the disc material into circulation. In this review most cases experienced similar mechanisms during their activities, such as when dancing, hand stand, defecation or when milking a cow. In these activities, a valsalva like maneuver was created, which increased the intra-abdominal and intra discal pressure that could have forced the disc material to spinal circulation. Increased abdominal pressure or intradiscal pressure due to valsalva manuver is thought to be the most responsible mechanism [20, 25]. Other important pre-disposing factor in children is the mobility within the spinal column [10, 14].

References [2,26] proposed a hypothesis, that increased intra vertebral-discal pressure forces the nucleus pulpous initially to venous sinusoids and then eventually to basivertebral vein leading to spinal vein occlusion via retrograde flow. Other hypothesis is the extrusion of the nucleus pulposus into the cancellous portion of the vertebral body- a Schmorl's nodes; and then embolizing to vertebral sinusoids and draining into the

basivertebral veins [19]. A valveless communication between the extrathecal and intrathecal vessel facilitates the retrograde transport of embolus to enter the spinal circulation via the Batson plexus [12, 24, 27,28]. On the other hand Davis [13] proposed alternate hypothesis that rupture of disc material embolizes to an adjacent radicular artery and into the arterial circulation. They reported 30 cases and in majority of their cases, nucleus pulposus was found in the radicular artery. Embolisation of the spinal artery eventually causes progressive spinal infarct. Clinically, patients would present with “stroke-in-evolution” with intervening symptom-free intervals [11, 23]. FCE are always arterial type, even when both arterial and venous emboli are present. Venous embolis are mostly asymptomatic and end up in the lungs [23].

As mentioned earlier, presentation of the spinal cord infarction due to FCE is of a "spinal stroke in evolution”. The symptoms presentation depends on the site of the cord and the vessels involved. In this case series, most of the patients had cervical cord involvement followed by thoraco-lumbar involvement and sacral involvement. Only one patient in this literature review had medulla and brain stem involvement secondary to anterior spinal and basilar artery involvement. All patients had sudden but progressive neurological deficits.

Anterior spinal artery (ASA) supplies anterior 2/3 of the spinal cord including the base of the posterior horn. Occlusion of the ASA, presents with an abrupt onset of back pain, bilateral weakness, areflexia and spinothalamic sensory deficit with the sparing of proprioception and vibration sense and autonomic dysfunction involving the sphincter incontinence, Posterior spinal artery supply posterior columns, dorsal root entry and a variable portion of the lateral corticospinal tract [21]. Posterior spinal artery occlusion causes bilateral motor deficit with leminiscal sensory deficit such as vibration proprioception [21]. Transverse infarct with bilateral motor deficit and complete sensory deficit is rare, but certainly a possibility [21]. However, infarcts with no detectable sensory deficit could be due to infarction of the spinal cord mainly involving in the motor area [11].

The diagnosis of the FCE is mainly clinical and should be considered in clinical differentials based on physical examination and confirmed by magnetic resonance imaging (MRI). There is no single non-invasive test to confirm the diagnosis of FCE spinal cord infarction [15]. However, MRI has negated the need for biopsy in suspected FCE cases. Before 1994, only two cases of FCE were imaged by MRI; whereas after 1994, all FCE suspected patients underwent MRI [17]. In this case review, 5 cases of FCE were report before 1994 and none were investigated with MRI out which four patient died. On MRI, the spinal cord infarct secondary to FCE is shown as lack of contrast enhancement in the acute phase, progressive enlargement or spinal cord infarct in evolution. Schmorl nodes on MRI are nonspecific and are not seen in all FCE infarct patients [23, 27, 29, 30]. In this case series, only few patients had schmorl nodes. Children and adolescents with neurological deficits without radiographic abnormalities also required diagnostic screening or appropriate imaging, somatosensory evoked potential and electromyography to assess the spinal activity and rule out psychogenic paraparesis or quadriparesis. CSF analysis FCE is usually normal as also noted in this review except for the one case.

So far, there is no definitive treatment recommendation for spinal infarct secondary to FCE. In majority of the cases, the treatment is supportive such as hospital or ICU admission, stabilizing patient hermodynamically in the presence of spinal shock and active rehabilitative therapy [10]. Role of steroids is controversial and spinal surgery is not indicated in majority of the cases [21]. In this review, steroids were administered to eleven

patients but the response was variable. Secondary management with aspirin [3, 10, 21,] and clopidogrel [20] is also reported; however, the results were variable among the studies. In this review only one patient was treated with the plasmapharesis or immunoglobulin. In some cases anticoagulation with heparin and aspirin has appeared to improve the outcome [31, 32]. Thone [21] recommended low dose aspirin as a secondary preventive treatment; however longer-term treatment includes intensive physiotherapy and physical rehabilitation remains the key [21, 33].

In this review, eight patients had minimal or no recovery, five had full or substantial recovery, five died due to respiratory complications. Cheshire [34] also reported variable outcome in their spinal cord infarct patients such as with variable recovery to no recovery. The important prognostic predictors in spinal cord infarction secondary to FCE are the severity of neurological presentation, MRI appearance of the spinal cord and segment of the cord involved [35]. Patients with higher cervical cord involvement often have medulla oblongata lesions and yield poorer outcome [10, 14, 15, 30, 36]. Age is also an important prognostic factor such as younger patients with cervical cord involvement have poor outcome as compared to older patients with lower segment involvement [10]. Other important prognostic factor is the development of the respiratory compromise or infections. In this review, all the four deceased patients had respiratory compromise or complications. One of the limitations in this review is that most cases were obtained from the case report or case series. But this is a challenge when reviewing rare cases. Secondly, literature search was restricted to the English language only. The main goal of this review is to provide better understanding about FCE induced spinal cord infarct particularly when patients do not have predisposing factors. A better understanding about FCE- spinal cord infarct pathophysiology and clinical presentations in similar clinical situations may help to identify and contribute to our understanding of this disorder and its natural history.

6. Conclusion

We presented a review of the cases of spinal cord infarction secondary to FCE. In these cases, spinal cord infarction often occurred apparently following the harmless events or physical effort, but an obvious relation is noted between the forward bending or neck rotation,5 and the onset of neurological symptoms. One should be aware of the potential adverse neurologic outcomes of inappropriate posture, when performing seemingly harmless activities. Reference [21], proposed the idea of valsalva manuver that is created during the activities, increasing the intra-discal pressure. In all these cases there was no major trauma. Secondly, in our opinion, in the event of unexplained acute onset paralysis, FCE should be considered in differential, and workup with a focus on surrounding vertebral vasculature. Only few cases have been reported about the spinal cord infarct secondary to FCE, therefore it is important to increase the awareness in our neurological and neurosurgical team about this entity. I agree with [19], that the addition of more cases in future will add into the better understanding of this entity and differentiating it from other acute myelopathies. Detailed history, clinical presentation and MRI can help to corroborate the diagnosis.

Conflict of Interest

There is no conflict of interest to declare. The manuscript has not been published elsewhere, and is not under

simultaneous consideration by another journal.

References

[1] F J Mateen, P A Monrad, A N Hunderfund, et al.; "Clinically suspected fibrocartilaginous embolism: clinical characteristics, treatments, and outcomes"; European Journal of Neurology, 18: 218–225; 2011

[2] I Feigin, N Popoff, M Adachi: "Fibrocartilaginous venous emboli to the spinal cord with necrotic myelopathy". J Neuropathol Exp Neurol; 24:63–74; 1965

[3] J R Nance, M R Golomb; "Ischemic spinal cord infarction in children without vertebral fracture"; Pediatr Neurol.; 36; 209–216; 2007.

[4] HS Bhatoe; "Cervical spinal cord injury without radiological abnormalities in adults". Neurol India; 48: 243-248; 2000.

[5] I Koyangi, Y Iwasaki, K Hida, et al; "Acute cervical cord injury without fracture or dislocation of the spinal column"; J Neurosurg; 93: 15-20; 2000.

[6] T Velnar, V Smrkolj, O Cerovic, et al; "An unusual case of high cervical spinal cord injury"; Folia Neuropathologica; 48: 134-138; 2010

[7] I Jurkovic, Eiben; "Fatal myelomalacia caused by massive fibrocartilaginous venous emboli from nucleus pulposus"; Acta neuropath, 15, 284--287, 1970

[8] K L Ho, J M Gorell, M T Hayden; "Fatal spinal cord Infarction caused fibro cartilaginous embolization of anterior spinal cord by the artery"; Human pathology; 11; 471-475; 1980

[9] D F Moorhouse, M Burke, C Keohane, M A Farrell; "Spinal cord infarction caused spinal artery by cartilage embolus to the anterior spinal artery"; Surg Neurol; 37: 448-52; 1992

[10] A Beck. F Gebhard, L Kinzl, et al; "Spinal cord injury without radiographic abnormalities in children and adolescents: case report of a severe cervical spine lesion and review of literature"; Knee Surg, Sports Traumatol, Arthrosc; 8;186–189 ; 2000

[11] N C Chen, C H Yeh, C Hui, R Lin; "Exercise related spinal cord infarction: a case report"; Acta Neurol Taiwan;19: 194-198: 2010

[12] J J Cuello, S Ortega-Gutierrez, G Linares, S Agarwal, et al. "Acute Cervical Myelopathy Due to Presumed Fibrocartilaginous Embolism A Case Report and Systematic Review of the Literature"; J Spinal Disord Tech;27:E276–E281; 2014

[13] GA Davis, GL Klug: "Acute-onset nontraumatic paraplegia in childhood: fibrocartilaginous embolism or acute myelitis?" Childs Nerv Syst 16:551–554, 2000

[14] R D Dickerman, M A Mittle, C Warshaw, J A Epstein; "Spinal cord injury in a 14-year-old male secondary to cervical hyperflexion with exercise"; Spinal Cord; 44; 192–195; 2006

[15] J J Han, T L Massagli, K M Jaffe, "Fibrocartilaginous embolism—an uncommon cause of spinal cord infarction: a case report and review of the literature"; Arch Phys Med Rehabil; 85: 153-7; 2004.

[16] C Kase, J Varakis, J Stafford; J P Mohr; "Medial medullary infarction from fibrocartilaginous embolism to the anterior spinal artery"; Stroke, 14, 413-418, 1989

[17] JJ Millichap, T Bernard, Leacock; "Spinal Cord Infarction with Multiple Etiologic Factors"; Society of General Internal Medicine; 22: 151–154; 2007

[18] J Redmond, A Thompson, M Hutchinson; "Acute central cervical cord injury due to disco dancing";

British Medical Journal; 286; 1704; 1983

[19] A Reisner, M F Gary, J J Chern, J D Grattan-Smith; "Spinal cord infarction following minor trauma in children: fibrocartilaginous embolism as a putative cause Report of 3 cases"; J Neurosurg Pediatrics; 11:445–450; 2013

[20] K Spengos, P Toulas, S Sameli, et al; "Spinal cord stroke in a ballet dancer"; Journal of the Neurological Sciences; 244, 159 – 161; 2006

[21] J Thöne, A Hohaus, A Bickel, F Erbguth; "Severe spinal cord ischemia subsequent to fibrocartilaginous embolism"; Journal of the Neurological Sciences; 263; 211–213; 2007

[22] G Toro, G C Roman, N Roman, et al; "Natural history of spinal cord infarction caused by nucleus pulposus embolism"; Spine, 19, 360-366; 1994

[23] L Tosi, G Rigoli, A Beltramello; "Fibrocartilaginous embolism of the spinal cord: a clinical and pathogenetic reconsideration"; Journal of Neurology, Neurosurgery, and Psychiatry; 60:55-60; 1996

[24] O M Yousef, P Appenzeller, M Kornfeld: "Fibrocartilagenous embolism: an unusual cause of spinal cord infarction" Am J Forensic Med Pathol; 19:395–399; 1998

[25] J M McLean, G L Palagallo, J P Henderson, J Kimm; "Myelopathy associated with Fibrocartilaginous emboli: review and two suspected cases"; Surg Neurol 229; 44: 228-35; 1995

[26] C Masson, Y Boukriche, JL Berthelot, JM Colombani; "Vertebra, rib, and spinal cord infarction caused by probable fibrocartilaginous embolism". Cerebrovasc Dis;12: 142–3; 2001.

[27] J W Kestle, C Tator; "Intervertebral disc embolization resulting in spinal cord Infarction"; J Neurosurg 71:938-941, 1989

[28] DJ Mikulis, SC Ogilvy, A McNee, et al. Commentary: "spinal cord infarction and fibrocartilaginous emboli". AJNR Am J Neuroradiol;13:155–160; 1992

[29] A Raghavan, E Onikul, MM Ryan, et al: "Anterior spinal cord infarction owing to possible cartilaginous embolism". Pediatr Radiol; 30:503– 506; 2004

[30] T P Duprez, L Danvoye, D Hernalsteen, et al; "Fibrocartilaginous embolization to the spinal cord: serial MR imaging monitoring and pathologic study"; AJNR Am J Neuroradiol; 26:496–501; 2005

[31] K Hakimi, T Massagli; "Anterior spinal artery syndrome in two children with genetic thrombotic disorders". J Spinal Cord Med; 28:69–73 2005

[32] D Cooper, D Magilner, J Call; "Spinal cord infarction after weight lifting"; Am J Emerg Med; 24: 352–5; 2006.

[33] S Kirshblum, M Priebe, C Ho, et al; "Spinal cord injury medicine. 3. rehabilitation phase after acute spinal cord injury"; Arch Phys Med Rehabil; 88, Suppl; 12007

[34] W Cheshire, C Santos, E Massey, J Howard Jr: "Spinal cord infarction: etiology and outcome"; Neurology 47:321– 330, 1996

[35] P Bosch, M Vogt, W Ward; "Pediatric spinal cord injury without radiographic abnormality: the absence of occult instability and lack of indication for bracing"; Spine; 27: 2788–2800; 2002.

[36] S. Weidauer, M. Nichtweiss, H Lanfermann, F Zanella "Spinal cord infarction: MR imaging and clinical features in 16 cases". Neuroradiology; 44: 851– 7; 2002