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To the Editor

Spinal Cord Injury after Prolonged Neck Flexion, is it an Underestimated Risk?

Prolonged extreme neck flexion is a reported risk factor for cervical spinal cord injury.[1-4] This is usually precipitated by altered level of consciousness as in intoxicated patients or patients under general anesthesia. We present a case of cervical spinal cord injury secondary to prolonged neck flexion after opioid abuse and discuss the postulated mechanisms of this rare injury and provide a review of the available literature on this topic in order to increase awareness of this complication.

History and Examination

This is a 20-year-old male with a past history significant only for intravenous abuse of opioids. He was brought to the hospital after he was found in the morning with inability to move arms and legs following a six-hour period of loss of awareness. The patient admitted having used a total of 160 mg of long acting oxycodone intravenously, and subsequently fallen asleep for approximately six hours with his neck-flexed forwards against the wall under a confined space. The patient's partner verified the history. On examination, the patient was awake and alert. His vital signs were normal. There were no obvious signs of external trauma. There was no neck stiffness. Cranial nerves were intact. Motor exam of the upper extremity revealed biceps and deltoids power to be 3/5 bilaterally, triceps and wrist extension were 2/5 bilaterally and hand intrinsic muscles were 1/5 bilaterally. Motor exam in the lower extremities was 0/5 with absent reflexes and decreased tone. Anal tone was absent.

Investigation

Relevant laboratory tests revealed absolute white blood count (WBC) count of 10.8 bil/L and erythrocyte sedimentation rate (ESR) of 4 mm/h. Blood and urine cultures were negative. Creatinine was 121 μmol/L. Myoglobin was not tested. Creatine kinase (CK) was 7189 U/L. Magnetic resonance imaging (MRI) of the spine showed marked cord expansion with high signal within the cord from C2 to C7 on T2WI (Figure 1). There was no significant cord enhancement. At the cranial end of the spinal cord signal changes, the signal abnormality assumes an H-shaped configuration indicating selective involvement of the grey matter suggestive of vascular injury (Figure 2). There were extensive signal changes in the paraspinal muscles with some

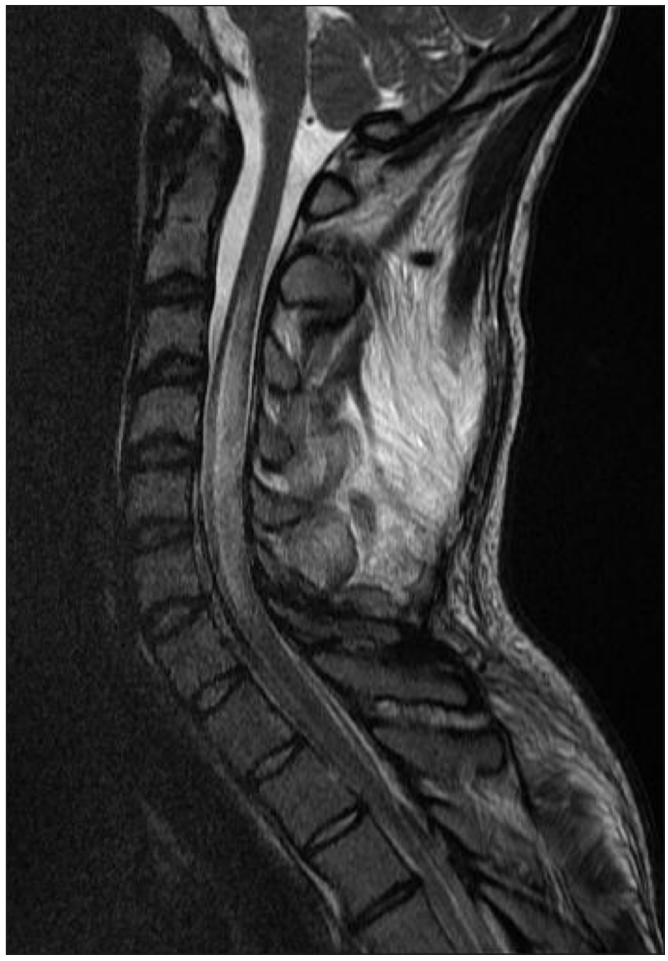

Figure 1: *Sagital T2 MRI image of the cervical spine showing spinal cord signal changes and paraspinal muscle edema .*

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enhancement. The radiological differential diagnosis included infection, inflammatory changes or necrosis/ rhabdomyolysis of the paraspinal muscle and associated spinal cord infarction and ischemia. Echocardiography was normal.

Hospital Course

After sending blood and urine cultures, the patient was started on intravenous antibiotics as empiric coverage for any possible infection. The patient had a CT-guided aspiration of the paraspinal muscles, which showed no evidence of infection. During the CT-guided aspiration, a core muscle biopsy was also performed which on pathological analysis showed skeletal muscle with degenerating and regenerating fibers and occasional macrophages suggestive of necrosis. Once all cultures were confirmed to be negative, antibiotics were stopped. Our diagnosis by exclusion was therefore that of cervical spinal cord ischemic injury/ infarction secondary to cord compression from prolonged neck flexion.

The patient made slight improvement neurologically in his deltoid power bilaterally (4/5 from 3/5 at presentation) and wrist extension (3/5 from 2/5 at presentation) but the rest of the neurological examination remained unchanged. The patient eventually was transferred to spinal cord rehabilitation center.

Discussion

We present a case of spinal cord injury secondary to prolonged neck flexion after impairment of consciousness. We hypothesize that the patient also had rhabdomyolysis related to prolonged muscle compression based on the biochemical, radiological, and pathological findings.[5]

We hypothesize that the prolonged neck flexion is the cause of this injury given lack of any other trauma. The selective involvement of the grey matter suggests a vascular-mediated injury given grey matter higher vulnerability to ischemia. One possible mechanism is an embolic event secondary to intravenous substance abuse. However, the selective involvement of the spinal cord segment that sustained the flexion injury and the normal echocardiogram are against this possibility.

The term "Cervical Flexion Myelopathy" has been used in few case reports to describe cervical spinal cord injury related to similar mechanism of prolonged neck flexion. The cases reported to date are due to prolonged neck flexion either as a consequence of surgical positioning[4] or loss of consciousness following substance abuse.[1-3]

1. Cervical Flexion Myelopathy *secondary to surgical positioning:*

Martínez-Lage et al[4] reviewed nine case reports of spinal cord infarction after cranial procedures. The commonest scenario is cervical cord infarction after posterior fossa surgery in the sitting position although some cases were done in the prone position.

2. Cervical Flexion Myelopathy *secondary to drug overdose:*

There are three case reports of spinal cord infarction secondary to prolonged neck flexion following substance abuse.[1-3] Valproic acid was used in one case and a benzodiazepine in the second one. In the third case, the patient used a combination of quetiapine fumarate (anti-psychotic), oxycodone/acetaminophen, and chloral hydrate.

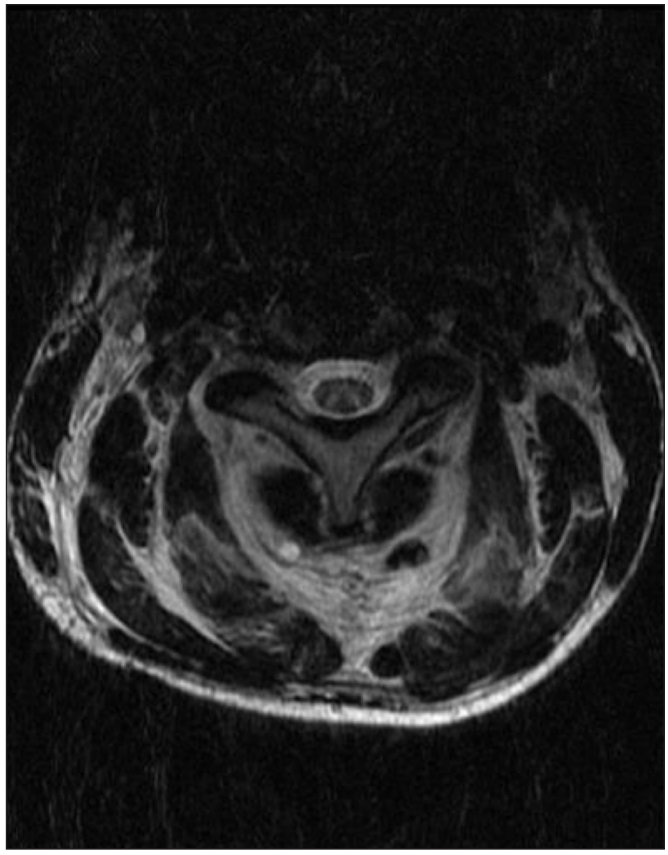

Figure 2: *Axial T2 MRI showing the selective grey matter involvement at C3.*

Pathophysiology

Several mechanisms have been suggested to explain spinal cord injury following prolonged neck flexion. One postulate is mechanical compression created by degenerative disc that leads to vascular compromise of anterior spinal circulation and subsequent spinal cord infacrtion. Kaye et al[1] suggests modified vascular theory such as mechanical compression causing failure of perfusion at the level of microcirculation. In this case report, there was a post-mortem examination that showed necrosis selectively involving the grey matter at the transition of normal to abnormal area of the spinal cord which is in keeping with the MRI findings in our case (Figure 2) and it supports the role of ischemia given the higher vulnerability of grey matter to diminished blood supply. Kaye's hypothesis is supported by the fact that MRI findings are usually not limited to a specific vascular territory and a pure mechanical compression would not explain the selective grey matter involvement.

Kaye's report is the only one with pathological confirmation of the diagnosis on post-mortem examination. In all other cases the diagnosis was made based on clinical and MRI evidence of cervical spinal cord injury and as a diagnosis of exclusion, once all other etiologies had been ruled out. We believe the MRI findings in our case provide radiological confirmation of Kaye's pathological findings and support a compromise of micro-

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circulation as the mechanism of the spinal cord injury.

Given the lack of effective medical intervention for this condition, the emphasis should be on prevention. In the cases associated with operative positioning, use of intra-operative neurophysiological monitoring would be advocated and argued to prevent this complication by detecting early evidence of spinal cord dysfunction during the procedure,[4] and in the cases of substance abuse raising community awareness of the devastating long-term irreversible neurological compromise and quadriplegia is imperative.

Conclusions

We believe that Cervical Flexion Myelopathy is an under-recognized clinical diagnosis and reporting such cases is important to raise awareness of the possibility of this complication, with the goal to improve or increase preventative measures.

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To the Editor

Isolated Unilateral Hypoglossal Nerve Palsy

Hypoglossal nerve palsy is a rare cranial neuropathy with a broad differential diagnosis. Most etiologies typically present with other neurological or systemic sequelae(1). Thus, isolated hypoglossal nerve palsy presents a diagnostic challenge requiring a systematic investigative approach. We report of an interesting case of isolated unilateral hypoglossal nerve palsy following a mononucleosis infection. The investigative work-up, differential diagnosis, and management are discussed.

A 57 year-old male presented with a 6-month history of a swollen left tongue, decreased tongue mobility, and dysphagia. He denied pain or dysarthria and had not experienced recent neck trauma or had any recent operations. Ten years earlier, he developed infectious mononucleosis and remained healthy until a second episode of mononucleosis that began two days prior to his presenting symptoms. On examination, the tongue's left side appeared atrophic, without fasiculations and it deviated leftward upon protrusion. The cranial nerve examination was otherwise unremarkable. Notably, cerebellar testing was within normal limits and there were no other focal neurologic deficits.

Repeated MRIs over a 3-month period failed to demonstrate a structural lesion accounting for the patient's presentation. After 3 months, the patient improved clinically. His tongue mobility and swallowing returned to normal, although he subjectively felt left sided tongue thickening. Based on his clinical course and investigations, a diagnosis of post-infectious hypoglossal nerve palsy was made.

The hypoglossal nerve is a purely motor cranial nerve innervating the genioglossus, styloglossus and hyoglossus muscles in the tongue. Pathology affecting the pathway within and beyond the hypoglossal nucleus may present as ipsilateral tongue paresis, tongue deviation towards the side of the lesion, atrophy, dysphagia, or dysarthria(2). There are many potential causes of hypoglossal nerve palsy. The largest review, a retrospective case series of 100 cases, reported tumors, predominantly malignant, as the most common cause(1). The most common malignancies reported were metastases, chordomas, nasopharyngeal carcinomas and lymphoma. Benign space-occupying lesions causing this pathology include schwannomas, meningiomas, ependymomas and craniopharyngiomas(1). Trauma is the next most common etiology, such as gun shot wounds or blunt injury(1). Fractures of the occipital condyle and odontoid process can disrupt the hypoglossal canal. Vascular causes include internal carotid and vertebral artery dissections or ectasia, vascular insufficiency(1), and dural arteriovenous fistulas(3). Systemic processes such as Guillain Barre Syndrome, multiple sclerosis, and diabetes mellitus are rare causes(1). Certain infections can cause hypoglossal nerve palsy including meningitis, osteomyelitis(1), poliomyelitis, syphilis, herpes simplex virus, cytomegalovirus and Epstein-Barr virus(3). Given that idiopathic isolated hypoglossal nerve palsy is a diagnosis of exclusion, a thorough investigative work-up should aim to rule out all of the above entities(3).

Hypoglossal nerve palsy due to infectious mononucleosis is a rare etiology; only six cases have been reported previously(4,5). The typical clinical presentation is an isolated, unilateral hypoglossal nerve palsy that occurs in children and adolescents. To our knowledge, this is the first case reported in a middle-aged male.

The diagnosis of hypoglossal nerve palsy requires an understanding of the differential diagnosis to guide history-

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