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GRAND ROUNDS

Double crush syndrome caused by cervical spondylosis and vertebral artery loop

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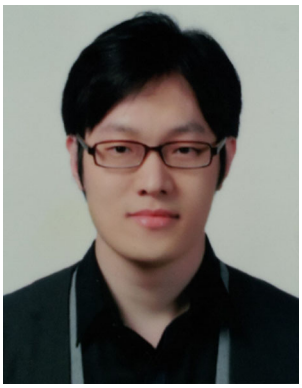
Abstract



Purpose The purpose of this article is to report a successful treatment experience in a rare case of simultaneous cervical nerve root compression by spondylotic cervical foraminal stenosis and a vertebral artery loop.
Methods 51-year-old man presented with a 4-year history of left-sided cervical pain radiating to the left shoulder with progressive exacerbation of weakness on left shoulder girdle muscles for 7 months. The patient had no history of trauma. The patient's CT and MRI revealed impingement of the left C6 cervical nerve root by a tortuous vertebral artery loop and also by narrowed left C5–6 cervical foramen that had undergone spondylotic changes. The patient underwent left C5–6 hemilaminectomy, facetectomy and C5–6 fusion. The procedures were uneventful, and the patient recovered with complete resolution of symptoms.
Results The patient continued to be asymptomatic at a 2-year follow-up examination, and the muscle mass of his left girdle returned normal.
Conclusions This report illustrates the first phenomenon of a double crush syndrome caused by vertebral artery loop and cervical spondylotic changes. When patients with cervical spondylosis present with unexplainably severe pain and weakness, additional underlying pathologies should be considered when making differential diagnoses. The investigation planning should involve electromyography, computed tomography angiography, and magnetic resonance imaging.

Keywords Crush syndrome · Vertebral artery · Loop · Cervical spondylosis

Case presentation

A 51-year-old man was referred to our institution because of a 4-year history of left-sided cervical pain radiating to the left shoulder and progressive exacerbation of weakness on left shoulder girdle muscles for 7 months. The pain was refractory to other forms of treatment, including multiple epidural injections, various anti-inflammatory medications, trigger point injections, and physical therapy. His medical history was negative for malignancy, endocrine disease, or familial disorders. On neurologic examination, Spurling's sign was positive and Lhermitte's sign was absent. Medical Research Council grade of the left shoulder abductor and

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elbow flexor muscles was 3/5. The left shoulder girdle muscles had atrophied slightly but no fasciculation was present. The patient's laboratory findings were within normal limits and showed no evidence of infection or systemic inflammatory disease. Electromyography (EMG) and nerve conduction velocity (NCV) were performed, and were consistent with a left C6 radiculopathy. Cervical X-rays, computed tomography (CT) scan, CT angiography (CTA) and magnetic resonance imaging (MRI) demonstrated a tortuous looped left vertebral artery at C5–6 level with left C5 and C6 nerve root swelling. No bone erosion was observed near the vertebral artery loop (VAL). Degenerative stenosis of the left C5–6 intervertebral foramen with C6 nerve root compression was also identified. To our knowledge, this is the first report concerning a rare case of cervical radiculopathy and shoulder girdle muscle weakness caused by multiple nerve root compressions by cervical spondylotic changes and VAL. We explain this accumulatively increased symptom intensity as double crush syndrome (DCS). The patient underwent left C5–6 hemilaminectomy, facetectomy and instrumented fusion.

Diagnostic imaging section

Cervical spine pre-operative studies showed in Figs. 1 and 2.

Historical review, epidemiology, diagnosis, pathology and differential diagnosis

Our patient initially presented with cervical radiculopathy. Cervical radiculopathy is a disease process caused by the compression of cervical nerve roots, which is commonly a result of degenerative osteophytes or lateral disc herniation. It is also known as cervical neural foraminal stenosis [1]. Infrequent causes of cervical radiculopathy comprise congenital, cystic, metabolic, neoplastic, and vascular conditions. Among these causes, VAL can lead to bony erosion, neurovascular compression, or vertebrobasillar insufficiency [2, 3]. Paksoy et al. reported the incidence of the vertebral artery loop as 7.51% in their 173 patients [4]. A study of 222 cadaveric spines documented the incidence of a tortuous vertebral artery as 2.7% [5]. VAL can occur equally in males and females, and the C4–C5 level is the most common site of unilateral lesions followed by C3–C4 and C5–C6 [6]. Even though the real prevalence of this abnormality is very low, it is not negligible. When patients with cervical spondylosis present with unexplainably severe pain and weakness, additional underlying pathologies should be considered and further study should be examined. Otherwise, it may lead to a possible vertebral artery injury casualty during the surgery.

The workup of VAL involves cervical X-rays that may show erosion of adjacent bone structures with sclerotic margins induced by high pulsatile arterial pressures [7]. CT is the most efficient way to evaluate the origin of root compression, and the loops of the vertebral artery are seen as hyperdense tissue masses enlarging the intervertebral foramen and transverse foramen [7]. CT scan findings typically include neural foramen widening and homogenous enhancement of the vertebral artery with preservation of fat planes in the foramina on postcontrast CT scan. MRI shows signal void on T1- and T2-weighted images, with extension into the neural foramen [8]. Conventional angiography or magnetic resonance angiography presently remains the standard in all cases where a vascular origin of compression is suspected, but the disadvantages include the risks of intervention needed and the high cost. In our case, multislice CT angiography was sufficient to assess VAL. Ganiyusufoglu et al. have also reported the potential benefit of multislice CT angiography as an alternative method to evaluate vertebral artery tortuosity [7].

DCS is a distinct compression at two or more locations along the course of a peripheral nerve that can coexist and synergistically increase symptom intensity. It was first described in 1973 by Upton and McComas, who theorized that asymptomatic compression at one site predisposed a peripheral nerve to increased susceptibility for impairment at another anatomic location [9]. Double crush eventually leads to disruption of axonal transport along the nerve, thus increasing the vulnerability of distal axons to compression syndromes and symptomatology [10].

The etiology and pathophysiology of DCS is dubious. In 1987, Nemoto et al. studied a canine compression model and concluded that a double lesion was greater than the sum of the deficits after each separate lesion [11]. This idea of a summation injury was supported in findings from another animal model by Dellon and Mackinnon; the authors examined a rat sciatic nerve compression model and concluded that "the existence of two sites of simultaneous compression will result in significantly poorer neural function than will a single site of compression" [12]. Multiple authors have proposed various explanations; the most accepted principle for DCS involves a primary nerve disorder that predisposes the nerve to further injury [10]. The proposed pathophysiology of DCS is the disruption of nutrient flow in both antegrade and retrograde directions along the axon. In a Delphi study, international experts concluded that four mechanisms were highly plausible: axonal transport, immune-response inflammation of the dorsal root ganglia, ion channel regulation, and neuroma-incontinuity [13].

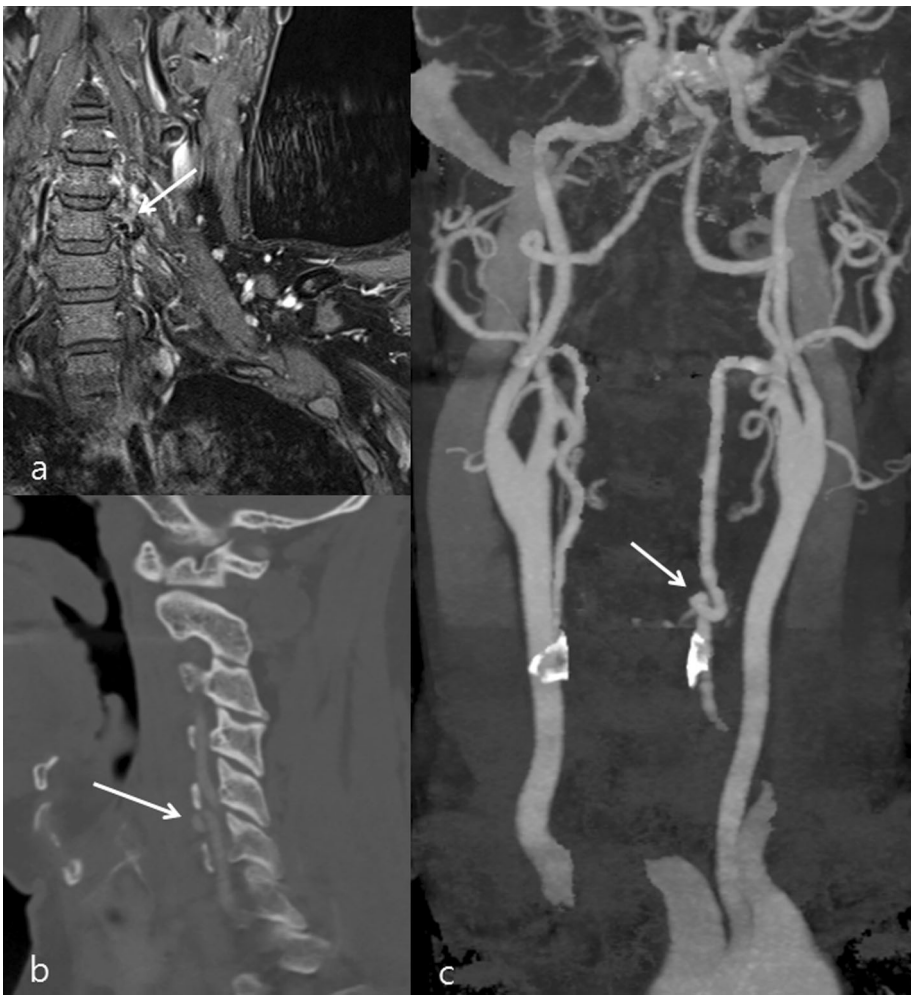

Fig. 1 Preoperative images: **a** T2-weighted coronal magnetic resonance images without contrast show a signal-void vascular structure in the left C5–6 intervertebral foramen (*arrow*). **b**, **c** Computed tomography (CT) angiogram demonstrating vertebral artery loop at the level of the left C5–6 cervical foramina

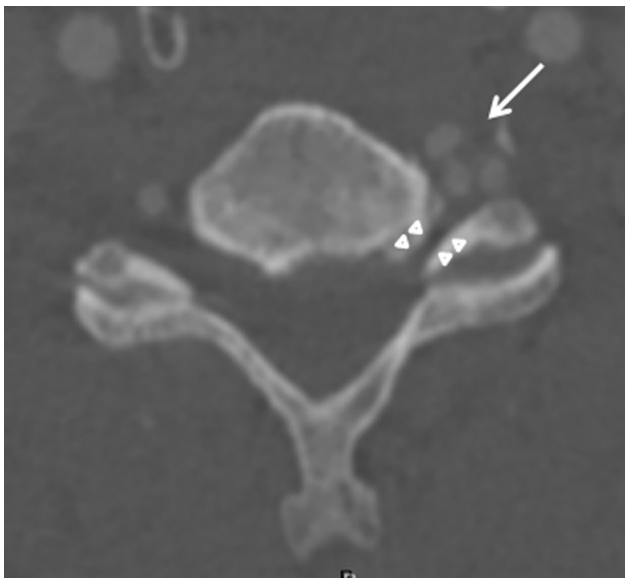

Fig. 2 Axial CT angiogram obtained at the level C5–6 shows a stenosis of the left intervertebral foramina (*arrow heads*) and a vertebral artery loop (*arrow*) compressing left C6 nerve root

Rationale for treatment and evidence-based literature

Given his physical presentations and EMG findings, both the tortuous left VAL and the left C5–6 cervical foraminal stenosis were supposed to be causative of the patient's symptoms.

Surgical procedures reported in the literature are microvascular decompression through the anterolateral [2, 14] or posterior approach [6, 15], foraminotomy with sectioning of the compressed rootlet [16], and vascular reconstruction through the anterolateral approach [17]. Horgan et al. have also reported nonsurgical care for this entity, with reduction of cervical radicular symptoms after improved blood pressure control and intermittent nonsteroidal anti-inflammatory agents [18].

In our case, the C5–6 cervical neural foraminal stenosis compressed the C6 nerve root at the proximal site. The symptom was aggravated because the VAL compressed the ventral ramus on distal C6 nerve root and increased the vulnerability of distal axons. Therefore, it was decided that

surgical decompression of the C6 cervical nerve root at the level of the left C5–6 cervical foramen would be necessary to alleviate pressure from the VAL and the C5–6 foraminal stenosis.

Our case did not have characteristic findings such as enlargement of the intervertebral foramen and transverse foramen. On the other hand, narrowing of the intervertebral foramen due to cervical spondylotic changes with facet joint hypertrophy was present at the pathologic level.

Hence, the case of our patient, the C5–6 disc space was not narrowed and there was no instability of the C5–6 segment, the mechanism of the facet joint arthrosis could only be explained by repeated stimulation by the pulsating VAL.

The episode of high pulsatile arterial pressure induced the instability of facet joint and capsular laxity. Afterwards, facet joint hypertrophy and osteophyte formation occurred during the restabilization phase as a result of foraminal stenosis. Oga et al. also concluded in their study involving 22 cases that the extent of the vertebral artery tortuosity correlated with the grade of cervical spondylosis [19].

Concerning treatment, we performed hemilaminectomy, facetectomy, pedicle screw fixation and fusion in our patient. The goals of treatment were to release the C6 nerve root from multiple compression sites.

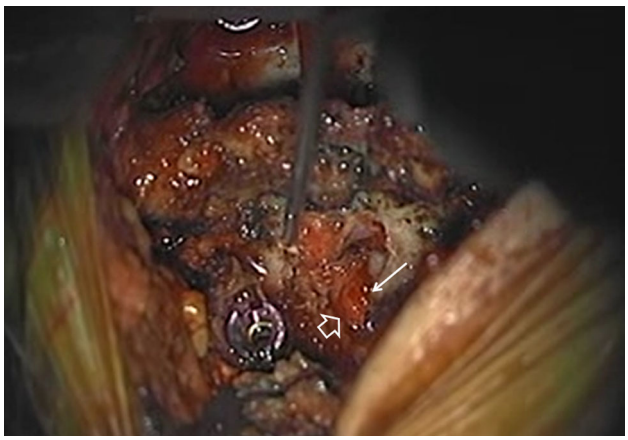

Fig. 3 Intraoperative photograph obtained after hemilaminectomy, C5–6 facetectomy and pedicle screw fixation. Left C6 nerve root (*arrow*) was edematous and decompressed from the vertebral artery loop (*open arrow*) and bone

Operative procedure

The surgery was performed under general anesthesia in a posterior approach to the cervical spine.

The patient underwent left C5–6 hemilaminectomy and facetectomy. The facet was hypertrophied and the C6 nerve root was compressed by a pulsatile mass presumed to be the left VAL. With assistance of a surgical microscope, hypertrophied bone around the tortuous vertebral artery was removed, and the C6 nerve root was freed from direct compression of the tortuous vertebral artery by inserting a Teflon plug in between (Fig. 3). Surgical decompression with or without instrumented fusion were contemplated.

We decided to perform a fusion procedure despite the fact that two of the three structural columns were preserved [20], because after decompression without fusion, repeated stimulation on C5–6 motion segment and disc space narrowing might lead to elongation of the VAL and symptom recurrence. Sakaida et al. suggested that narrowing of the disc space can cause elongation of the vertebral artery, leading to vertebral artery loop [17].

Another point to consider was between placing pedicle screws and lateral mass screws. We used pedicle screws despite higher risk of vertebral artery injury because

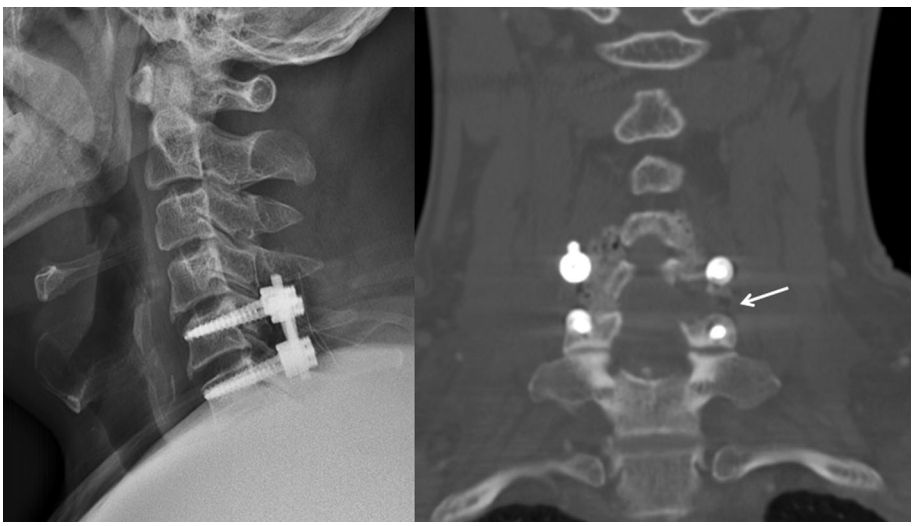

Fig. 4 Postoperative images: (*left*) Plain radiograph showing the final result of the C5–6 pedicle screws. CT scan demonstrating good decompression of the C6 nerve root (*right*)

inserting lateral mass screws were limited due to wide facetectomy state and pedicle screws have higher stability over lateral mass screws fixation as regards to primary and stability after cyclic loading. Intraoperative monitoring was performed during procedures to prevent secondary injury due to vertebral artery thrombosis and dissection.

Clinical outcome

The patient was completely free of pain and weakness by the third postoperative day. A postoperative cervical X-ray and CT scan were achieved and affirmed the C5–6 pedicle screws and good decompression of the C6 nerve root (Fig. 4). The patient continued to be asymptomatic at 2-year follow-up examination, and the muscle mass of his left shoulder girdle was normal.

Compliance with ethical standards

Conflict of interest This paper was written as part of Konkuk University's research support program for its faculty.

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