

Two Cases of Double Crush Syndrome Progressing to Complex Regional Pain Syndrome

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Abstract

The double crush syndrome is described as an increased risk of distal nerve injury after a more proximal injury. This was a case series of two patients who developed Complex Regional Pain Syndrome under circumstances when a double crush phenomenon could have occurred. Both initially had spinal stenosis and subsequent spinal surgery. Both later had crush injuries to a unilateral lower extremity, which progressed to CRPS. There is no documented correlation between double crush syndrome and CRPS; however, these cases raise awareness about a heightened propensity for CRPS in such patients, which will lead to earlier, accurate diagnosis and treatment.

Keywords

Complex Regional Pain Syndrome, CRPS, Double-Crush Syndrome, Spinal Stenosis, Spinal Surgery

1. Introduction

The double crush syndrome (DCS) was proposed in 1973 by physicians Upton and McComas [1]. They described the potential susceptibility of a distal nerve segment to injury in the setting of a more proximal lesion: “neural function is impaired because single axons, having been compressed in one region, become especially susceptible to damage at another site [1].” The syndrome is manifested as a disorder that includes pain, numbness, and weakness due to two or more insults to individual peripheral nerves. Descriptions have included carpal tunnel syndrome (CTS) associated with cervical radiculopathy, as well as CTS with brachial plexus compression [2]. Multiple studies have implicated varied mechanisms contributing to the initial insult. These have included pathological, anatomic, or metabolic origins [2]-[7]. Common pathological risk factors may include stenosis of the nerve root and inflammation of the nerve root. Some suggested metabolic risk factors include diabetic neu-

ropathy, thyroid disease, neural ischemia, and chemotherapeutic agents, along with other various toxicogenic and vasculopathic factors [2]-[8]. Pregnancy and excessive weight gain have also been implicated [2] [3]. Studies of the physiologic mechanism have been completed following the initial insult. They have implicated both morphological and biochemical changes in the biogenesis of pain [8]. The basic premise of DCS was initially suggested to be impairment of axonal flow at one or more sites. Other suggested mechanisms include altered ion channel regulation, inflammation of dorsal root ganglia, and neuroma in continuity [1] [6] [9] [10]. Further implicated causes have included impairment of distal neural microcirculation due to proximal endoneurial edema, decreased neurofilaments, and disruption of lymphatic or venous outflow [11] [12]. No particular cohort of persons has been identified to have an increased propensity to develop double crush syndrome [9].

The two cases presented here provide examples of patients with pre-existing neurologic injury, and a subsequent secondary insult, which develops into Complex Regional Pain Syndrome (CRPS). Two types of CRPS have been defined. CRPS type I is associated with previous trauma, previous surgery, work related injury and female sex. There is no definable nerve lesion. CRPS type II is associated with a previous nerve injury [13]-[15]. Signs and symptoms include: pain disproportionate to the inciting event; hyperalgesia; allodynia; trophic, sudomotor and vasomotor abnormalities; movement disorders; and lack of single nerve distribution [14] [15]. Multiple theories regarding pathogenesis of CRPS include inflammation, central and/or sympathetic nerve dysfunction, autoimmune conditions, limb ischemia, cortical reorganization, small fiber neuropathy, or neurogenic inflammation [16]. Associated conditions have included migraine, osteoporosis, and asthma [16]. Genetic factors have also been implicated [14]. Primary treatments for CRPS include sympathetic blocks, physical therapy, spinal cord stimulation, and oral medications, as well as desensitization and pain psychology [15].

In the two cases to be presented in this article, there was an initial mechanism of insult to the spinal cord, followed by crush injury to a distant part of the body, along the same peripheral nerve pathway as the first insult. The result was development of CRPS type II. Both patients have reviewed this article and written consent stating that this report may be published has been provided.

2. Case Description

The first patient is a 50 y/o female that developed left foot pain after a heavy palette fell onto her left foot. She had had a lumbosacral L5-S1 fusion and laminectomy 21 years prior to the aforementioned injury. Her history included coronary artery disease, with a history of myocardial infarction and stent placement, cesarean section twice, gastric bypass, and hysterectomy. Medications included citalopram 20 mg daily, celecoxib, and acetaminophen 500 mg. She reported a sharp and burning pain at her left foot. She had mild tenderness at the lateral aspect of her left foot. A lower extremity temperature difference was noted, with the left foot at 26 C, and the right foot at 27.5 C. Her vital signs were within normal limits. She was started on pregabalin 50 mg. Over six months, she underwent multiple sciatic nerve blocks, and yet had unsatisfactory pain control after blocks. Subsequent exams revealed faster toenail growth on the left, color change in the foot, hyperalgesia of the lateral portion of the left foot, decreased range of motion of the toes, and strength limiting pain with movement. A TENS unit was prescribed, which improved pain to a limited degree, and subsequently a lidocaine patch was prescribed. She underwent a psychiatric review for spinal cord stimulator placement which did not reveal any notable findings, and a trial SCS was placed which offered "adequate coverage of all painful areas". This was followed by permanent SCS placement. In the interim, the patient developed a cervical spondylosis with upper extremity radiculopathy. She was treated with cervical injections and ultimately an ACDF. She returned 9 months later, s/p ACDF for cervical spondylosis, reporting worsened axial lumbar pain and left foot/leg pain after a trauma. Ultimately, it was revealed that she had a SCS lead fracture, requiring replacement.

Patient two is a 41 y/o female with a history significant for an MVC in 2001. She had a subsequent lumbar fusion of L5-S1 in 2007. She had been using duloxetine, diazepam, hydrocodone, oxycodone, temazepam, and tizanidine at the time of evaluation. Imaging was completed in the following year, which noted facet joints L4-5 were impinged, with a screw in the joint. She proceeded to have posterior hardware removal, followed by an anterior L4-5 fusion. A subjective improvement in pain was noted, but she continued to experience unacceptable levels of pain. She was offered epidural steroid injections and trigger point injections. She eventually was evaluated for SCS placement. However, she postponed SCS placement for personal reasons. Four months following evaluation, the patient reported that a horse had stepped on her left foot. Physical exam revealed allodynia of the whole foot and ankle with warmth and dry skin compared to the right. There was decreased muscle mass of the

left calf. She was diagnosed with CRPS of the left foot. The patient was started on amitriptyline. She later underwent multiple left lumbar sympathetic blocks which ultimately offered minimal relief. A series of popliteal nerve blocks and aggressive physical therapy were employed, and she reported that the overall foot pain was improved since injury. Ten months after crush injury, the SCS placement was again discussed, and the patient again followed up with the pain psychiatrist. Most recently she had not yet undergone SCS placement. She also reported an aggravation on left foot injury, when a person had fallen upon her foot.

3. Discussion

The current cases give evidence of a pre-existing neurologic injury with subsequent lumbar surgery, and followed by a secondary injury; this is a set up for the double crush syndrome. However, these injuries resulted in CRPS. Both of these patients had lumbar fusion surgery years prior to a second event and yet had continued symptoms of neuropathy. Neither had symptoms of CRPS until an injury to the foot resulted in subsequent presentation of symptoms. Other similarities included female gender.

These cases suggest an as yet undocumented, possible correlation between the double crush syndrome and development of CRPS. There certainly are factors which are consistent in the literature between the two syndromes. Injury and inflammation of the nerve (root) is discussed as a pathogenic factor for both syndromes. This was documented in these patients with prior back surgery for stenosis, and yet with continued lower back pain. Nerve axonal transport dysfunction was the original suggested mechanism by Upton and McComas as a cause of DCS, and was also discussed as a pathogenic factor in CRPS. Finally, there was the suggestion of cortical reorganization in the setting of CRPS; this had also been proposed in other chronic neuropathic pain scenarios, including the radicular pain patterns described in the above patients [17] [18]. The evidence given here is certainly far from conclusive, as the mechanisms of both CRPS and DCS are still heavily disputed. However, it serves to bring attention to the possibility of a heightened propensity for patients who have experienced a “Double crush scenario” to develop CRPS. Obviously there will need to be further research of the discussed syndromes. A heightened awareness to the development of CRPS in this scenario may lead physicians to earlier diagnosis and treatment. This will likely result in better clinical outcomes [19].

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