Abstract

Study Design. Case report.

Objective. To report a previously undescribed association between complex regional pain syndrome (CRPS) and surgery for cervical disc protrusion.

Summary of Background Data. Although CRPS has been associated with disc protrusion and lumbar spine surgery, there is no record in the literature of a similar association with cervical disc protrusion.

Methods. Description of a clinical case.

Results. A patient with radicular pain due to cervical disc protrusions underwent posterior foraminotomy, which did not relieve his pain. Within 3–4 weeks, he developed features of CRPS. Review at 16, 22, and 34 months revealed some, but limited, improvement.

Conclusions. This case provides circumstantial evidence of a hitherto undescribed association between CRPS and cervical disc protrusion. The mechanism is elusive, but may involve damage to small nerves in or near the intervertebral foramina.

Key Words. Complex Regional Pain Syndrome; Cervical Disc; Surgery

Introduction

Complex regional pain syndrome (CRPS), previously known as reflex sympathetic dystrophy [1–5], sometimes occurs as a complication of various medical conditions [4–6], but more often it occurs following trauma, spinal cord injuries, fractures, dislocations, burns, or crush injuries [7,8], or as a complication of surgical procedures [8,9]. Although the syndrome has been reported following surgery for lumbar disc protrusion [10–14], our search of the literature has not detected reports of CRPS in patients with cervical disc protrusion or cervical radiculopathy, with or without surgical intervention.

Case Report

A 39-year-old patient suffered a twisting strain and a vertical compression injury to his neck, both within a short period of time. He developed pain in the left shoulder and arm, radiating posteriorly down to the elbow. Additional information on clinical features at the time was not available to the current authors. Magnetic resonance imaging disclosed the following: a left-sided C6–7 disc protrusion slightly compressing the C7 nerve root; and similar findings, but to a lesser degree, at C5–6 (Figures 1 and 2).

Because of the nerve root involvement, the patient consulted a neurosurgeon, who performed a two-level, posterior foraminotomy at C5–6 and C6–7; but the patient did not benefit from the surgery (details about how the operation was performed or what happened during surgery were not available to the present authors).

During the first 3–4 postoperative weeks, the patient developed a burning pain along the left arm, left forearm, and hand. This was associated with paresthesia in all fingers, swelling of the hand and forearm, and increasing inability to use the left hand. The symptoms were intensified during the succeeding year, and eventually, a second
opinion was sought from the surgeon in our group (GMW).

At the initial consultation, some 16 months after the surgery, the main features were a constant burning pain, hyperalgesia, and allodynia in a dysfunctional left upper extremity, with vasomotor and sudomotor changes. The left hand was dependent, supported by the other hand or a narrow sling (cuff and collar type), which irritated the skin even with its minimal slips on the forearm. The shiny, swollen skin was cyanotic, mottled, and very tender upon digital pressure. Attempts to mobilize the joints elicited severe pain. No dystrophic nail or hair changes were recorded, but the skin was cold and wet. The circumference of the left arm was 4 cm less than the right, but the circumferences of the left forearm and left hand were each 2 cm greater than on the dominant right side. Neurological testing revealed absence of the triceps reflex; sensation was diminished throughout the affected limb in a nondermatomal distribution. A diagnosis of CRPS was made (Table 1).

The patient was reviewed on two further occasions. At 22 months postoperatively, slight improvement in the clinical features of the arm and hand was recorded (Table 1). The patient was cared for by a pain clinic, which provided better control of the pain, using opioids, tricyclics, and hypnotics on a daily basis, resulting in improvement to the physical features and associated psychological and social problems.

On the last occasion, some 34 months postoperation, although somewhat less intense, several features of CRPS were still evident (Table 1). Swelling, paleness, and cold and wet skin persisted. Active mobility of the left elbow had been regained. The hand was slightly functional, and no longer required support. Some active movement in the fingers and wrist were present, but passive movement of the joints still elicited a severely painful reaction, with spasm of the neck and the entire limb.

**Discussion**

CRPS has been described in association with lumbar disc protrusion and following lumbar spinal surgery [9–14]. The etiology of CRPS in these cases is elusive. Explicit explanations have not been forthcoming in the literature. Some investigators have proposed that the sinuvertebral nerve can be "traumatized" and develop ephapses [10]. Injury to other nerves affected by the disc protrusion or by the surgery is not less appetite as a conjecture.

The present case constitutes the first report of CRPS in a patient with radiculopathy and cervical disc protrusion treated by foraminotomy. It is not evident from the data acquired, if the CRPS was triggered by the original disc protrusion and radiculopathy (and would have developed even if surgery had not been undertaken), or if it was triggered by the surgery. The nearest related report in the literature describes CRPS developing after bilateral neck dissection [8]. In that case, CRPS developed on the side of radical dissection, but not on the side of partial dissection.
Although foraminotomy is performed from the back, and radical dissection is performed from the front, these procedures share a common feature. Rami communicantes pass from the middle cervical ganglion to the C6 spinal nerve and from the inferior (stellate) ganglion to the C7 spinal nerve. These rami give rise to the cervical sinuvertebral nerves and to the vertebral nerve [15,16]. Lying at the depth of dissection, it is feasible that one or other of these small nerves could be injured in either procedure. This would offer an explanation for the genesis of CRPS similar to that suggested for the lumbar cases.

When first seen, our patient satisfied the clinical diagnostic criteria proposed by Harden et al. [17], and the operational criteria for the diagnosis of CRPS, which require a score of at least 11 on the Galer scale [18] (Table 1). Over time, as the patient improved, his score on the Galer scale dropped, and he no longer satisfied those criteria. Those scores, however, perhaps underestimate the patient’s condition because features such as sensitivity to cold and summation to pinprick [18] were not tested. Nevertheless, he still fully satisfied the clinical criteria of Harden et al. [17].

Sharing case reports, such as this one, can serve several purposes. Firstly it can provide consolation to others who have encountered this same complication, or will do so in the future, in that their experience is not unique. Secondly, putting on record this particular association with cervical surgery renders it a recognized entity, which may have medicolegal significance.

**References**


**Table 1**  The clinical features of a patient with CRPS at various times after posterior foraminotomy at C6–7, C5–6

<table>
<thead>
<tr>
<th>Features</th>
<th>Postoperative Follow-up</th>
<th>I  15 Months</th>
<th>II 22 Months</th>
<th>III 34 Months</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pain</td>
<td>++</td>
<td>++</td>
<td>++</td>
<td></td>
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<tr>
<td>Hyperalgesia</td>
<td>++</td>
<td>+</td>
<td>+</td>
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<tr>
<td>Allodynia</td>
<td>++</td>
<td>+</td>
<td>0</td>
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<tr>
<td>Skin</td>
<td></td>
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<tr>
<td>Temperature</td>
<td>Cold</td>
<td>Cold</td>
<td>Cold</td>
<td></td>
</tr>
<tr>
<td>Color</td>
<td>Cyanotic, mottled</td>
<td>Pale, mottled</td>
<td>Pale</td>
<td></td>
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<tr>
<td>Dystrophy</td>
<td>0</td>
<td>0</td>
<td>0</td>
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<tr>
<td>Edema</td>
<td>+</td>
<td>0</td>
<td>0</td>
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<tr>
<td>Sudomotor</td>
<td>Wet</td>
<td>Wet</td>
<td>Wet</td>
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<tr>
<td>Motor</td>
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<tr>
<td>Elbow</td>
<td>Absent</td>
<td>Present</td>
<td>Present</td>
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<tr>
<td>Wrist</td>
<td>Absent</td>
<td>Weak</td>
<td>Present</td>
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<tr>
<td>Fingers</td>
<td>Absent</td>
<td>Absent</td>
<td>Weak</td>
<td></td>
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<tr>
<td>Muscle atrophy</td>
<td>Left arm</td>
<td>Left arm</td>
<td>Left arm</td>
<td></td>
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<tr>
<td>Sensation</td>
<td>Reduced</td>
<td>Reduced</td>
<td>Reduced</td>
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<tr>
<td>Triceps reflex</td>
<td>Absent</td>
<td>Absent</td>
<td>Absent</td>
<td></td>
</tr>
<tr>
<td>CRPS score (0–20)</td>
<td>12</td>
<td>10</td>
<td>9</td>
<td></td>
</tr>
</tbody>
</table>

CRPS = complex regional pain syndrome.
The CRPS score is according to the operational criteria of Galer et al. [18], under which a score of 11 indicates CRPS. +: present or evident; ++: markedly evident or severe; 0: not evident.


