

Case Report

Complete Relief of a Severe Feet and Hand Necrotising Raynaud's Phenomenon with Double Epidural Cervical and Dorsal Spinal Cord Stimulation

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Abstract

Several evidences suggest that Spinal Cord Stimulation (SCS) can effectively treat vascular disorders. Raynaud's phenomenon is a vasospastic condition affecting primarily the distal resistance vessels, triggered by cold temperatures, or by emotions such as anxiety and stress. High levels of sympathetic activity seem to be related to Raynaud's phenomenon as well as low levels of calcitonin gene-related peptide expression in the local sensory fibers. Patients with pain due to vascular disease initially receive conservative and pharmacological therapy to treat the underlying cause. If the symptoms persist, amputation could be necessary. We present a case of a severe necrotising Raynaud's phenomenon involving one hand and both feet successfully treated with epidural cervical and thoracic SCS.

Keywords: Raynaud's Syndrome; Spinal Cord Stimulation

Introduction

Raynaud's Phenomenon (RP) occurs frequently with an incidence of 3% to 21%. RP is a vasospastic condition affecting primarily the distal resistance vessels, that manifests itself as an extreme whiteness of the fingers or toes and later as a blue discoloration leading to ulcers [1]. Usually RP is triggered by cold temperatures, or by emotions such as anxiety and stress. High levels of sympathetic activity seem to be related to RP as well as low levels of calcitonin gene-related peptide expression in the local sensory fibers [2]. Patients with pain due to vascular disease initially receive conservative and pharmacological therapy to treat pain and the underlying cause. If the symptoms persist, a trial of Spinal Cord Stimulation (SCS) can be performed in order to cure ulcers, reduce pain and prevent amputation. There are several reports suggesting the beneficial effect of SCS in a variety of vascular-linked pathology, with reduction of pain and analgesics requirements [1, 3]. The decision to implant a SCS system is a multidisciplinary strategy of advanced pain management. In the present report, we present a case of a severe necrotising RP

involving one hand and both feet successfully treated with cervical and dorsal SCS.

Case Report

A 37-year-old woman presented in November 2016 complaining of pain involving both feet with sign of incoming ischemia and necrotic ulcers of toes. Also the fifth finger of the right hand was extremely cold and painful with ischemic signs. The diagnosis was necrotizing RP at both feet and right hand (Figure 1, Panel A). Neither laboratory findings, such as Antinuclear Antibodies (ANA), full blood or serum electrophoresis changes, nor clinical manifestations of connective tissue diseases were present. Nail fold video-capillaroscopy did not detect specific microvascular abnormalities. The character of pain was tearing; touching objects or dresses led to an immediate increase of pain intensity, so that she even could not wear shoes, walk or take objects. The pain ratings were mean 8/10, maximum 10/10, and minimum 6/10 on the nominal analogue scale and didn't respond to any kind of therapeutic manoeuvre or behavioural factors. Gabapentin up to 900 mg /die and oral morphine 120 mg/die did not allow pain relief.

She had been immediately treated with intravenous iloprost (up to 2ng/kg/min for 24 hr for 7 days), with scarce clinical response and increasing pain and digital necrosis. The hypothesis of amputation was therefore taken into account, but a trial of SCS was attempted before. Prior to the surgical intervention written consent was obtained. A standard operative technique for placement of SCS was employed under fluoroscopic guidance in two planes. Two octopolar electrodes (Vectris™ SureScan® MRI 1x8, Medtronic, Minneapolis, MN, USA) were used. One electrode was directed cranially with the tip at C3-C4 interspace. The paresthesia elicited by the single lead covered well the right arm and hand, but the coverage was less efficacious in the legs. Therefore, another catheter was midline positioned at T9-T10 to elicit paresthesia at both legs (Figure 2). The electrodes were connected to two extensions, which were externalized 15cm para spinally.

On the first postoperative day, the SCS led to immediate pain relief and feet and hand perfusion got better very soon. After a testing phase of 14 days, a rechargeable impulse generator (Restore Advanced™ Sure Scan MRI Medtronic, Minneapolis, MN, USA) was implanted under local anaesthesia; amputation was no longer necessary and oral Morphine and Gabapentin were gradually stopped. After 2 months, toes ulcers completely healed (Figure 1, Panel B). The stimulation parameters are showed in table 1 and were maintained at each control. The patient has been followed up through the outpatient department. At present, 2 years after the implantation of the device, she has a total pain relief and continues vasodilator infusion monthly. ANA turned positive (titre 1:320), but the remaining investigations were negative and nail fold videocapillaroscopy findings were still no specific.



Figure 1 Panel a: Signs of incoming ischemia and necrotic ulcers of toes and the fifth finger of the right hand at admission. **Panel b:** complete remission of ulcers at 2 months.

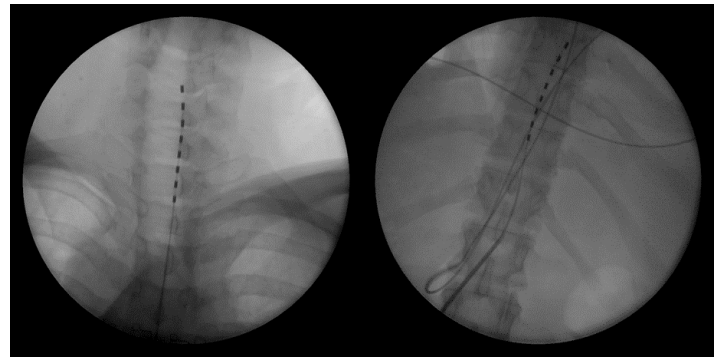


Figure 2: The epidural position of the cervical and dorsal catheters.

Electrode (tip)	cervical (C3-C4)	dorsal (T9-T10)
Amplitude (V)	5	2.2
Impulse duration (msec)	270	270
Frequency (Hz)	80	80
Electrode polarity	0:+,1:-,4:+	1:-,5:+

Table 1: The stimulation parameters for each catheter at implantation. The same parameters were confirmed at follow-up.

Discussion

The use of SCS in Raynaud's syndrome was first described in 1989 by Robaina et al, who observed good-to-excellent results in three patients with idiopathic Raynaud's disease [1]. Other report confirmed the beneficial effect of SCS, both in term of pain reduction and better perfusion [4, 5], even if the mechanism is not yet fully understood [6]. One hypothesis is that SCS activates low-threshold, large diameter Ab-fibers, which synapse onto inhibitory (GABA-ergic or cholinergic) interneurons in the spinal dorsal horn. These interneurons release inhibitory neurotransmitters (eg, GABA), thus reducing the excitability of spinal second order neurons, so that subsequent input from A and C-fibers is attenuated [5, 7]. Others proposed that the increase of cutaneous blood flow by antidromic activation of afferent fibers in the dorsal roots causes peripheral release of calcitonin gene-related peptide. This would enable cutaneous vasodilation by means of nitric oxide release by endothelial cells [8], thus increasing peripheral blood flow.

In addition SCS induces inhibition of efferent vasoconstrictor sympathetic activity, transmitted via nicotinic receptors in the ganglia and mainly via alpha 1 receptors on the vessels [6,9], thereby increasing blood flow. Furthermore, pain relief can be mediated by

suppression of nociceptive transmission via descending inhibitory pathways [10]. This is the first case report, to our knowledge, in which a double cervical and thoracic lead was implanted for RF. Actually, even if a single median cervical catheter can result in stimulation of both legs and upper extremities, a double, cervical and thoracic catheter, can allow to program each catheter separately in order to achieve a better paresthesia coverage and pain control in each single extremity.

As a matter of fact, also Wolter demonstrated that a single median high cervical electrode does not always result in stimulation of both legs and arms [3], since only at higher voltages stimulation might also be felt in the trunk and in the legs (by activation of the smaller amount of fibers in the superficial layer of the fasciculus cuneatus) but then it might already create unpleasant stimulation of the arms and hands. Therefore, we decided to implant a double cervical and thoracic electrode to assure a better stimulation pattern, although risks and costs are more pronounced. We cannot argue if a single cervical catheter stimulation with a paresthesia free system, like "high frequency" or "burst stimulation" [11,12], would have had the same result of our implant with a similar or better comfort. The present case once more highlights that epidural neuro-stimulation is a promising therapeutic option for severe RP. In particular, it shows the feasibility of a double (cervical and thoracic) electrode placement in order to achieve a better control of pain and perfusion of extremities.

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