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Treatment of Long-Term Complex Regional Pain Syndrome Type I of the Upper Extremity with Stimulation of the Cervical Spinal Cord: A Case Report

Frank F*

Department of Neurosurgery, University Hospital Leipzig, Leipzig, Germany

***Corresponding author:** Frank F, Department of Neurosurgery, University Hospital Leipzig, Leipzig, Germany; E-mail: Franziska.Frank@medizin.uni-leipzig.de

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Abstract

Background: Conservative treatment of patients with complex regional pain syndrome (CRPS) can be challenging and unsatisfactory due to a lack of pain reduction and dysfunction. CRPS can lead to serious mental health impairment with catastrophic pain perception. Spinal cord stimulation is a common treatment for mixed and chronic neuropathic pain syndromes that has been shown to be effective in early stages of CRPS type I.

Case Description: We present a case of a young woman with long-term CRPS of her left wrist. Under conservative therapy, she developed intolerable pain with catastrophizing pain perception and a desire for distal amputation of the upper extremity. Even after a prolonged course of disease, she was successfully treated with cervical spinal cord stimulation.

Conclusion: SCS is an effective treatment for patients with chronic upper extremity CRPS that is unresponsive to conventional therapy and should be considered at an early stage to prevent serious psychological and physical health effects. Even with long-term CRPS, SCS has positive effects on pain perception, mental health, and catastrophizing pain perception.

Keywords: Chronic pain syndrome; Spinal cord; Hyperalgesia; Allodynia

Background

Complex regional pain syndrome (CRPS) is a chronic pain syndrome that affects the upper and lower extremities and occurs primarily after trauma or surgery. It is more common in middle-aged women [1, 2]. The pain intensity is disproportionate to the initial event and is associated with motor, sensory and autonomic deficits. Clinical diagnosis should be made early to avoid delayed treatment and is facilitated by the Budapest criteria [3]. Pathophysiology is poorly understood and conventional therapy with pharmacological, physical and psychological aspects is challenging and applied in an interdisciplinary environment. Psychological factors and catastrophic perception can increase stress, aggravate pain and dysfunction [4]. CRPS can lead to serious mental health impairment and can drastically affect the quality of life. Interventional methods should be considered if conservative therapy does not respond within 12 to 16 weeks. However, these surgical measures should be accumulated gradually in order to prevent further somatization of the complaints. SCS was first introduced in 1967 and has been shown to be highly effective in the treatment of CRPS type I with reduction of pain, allodynia, muscle dysfunction and improvement in quality of life [5]. We present a case of a young woman with long-term distal upper extremity CRPS type I with catastrophic pain perception and a desire for an upper extremity amputation who was successfully treated with cervical spinal cord stimulation. The patient gave written informed consent.

Case Description

The patient presented herself to our neurosurgical outpatient department for the first time in May 2019 with increased pain in her left wrist that did not respond to conservative treatment. In 2011, the patient worked as a paramedic and had an accident

while working in medical service. The accident caused an avulsion fracture of her left wrist that resulted in several follow-up operations, i.e. 13 surgical procedures, including an arthrodesis of her left wrist, with no lasting effect on pain or function. As a result, neuropathic pain developed in 2015 and a complex regional pain syndrome was diagnosed in November 2016 (Figure 1). The pain symptoms started in her left wrist and spread over to the entire hand and forearm.

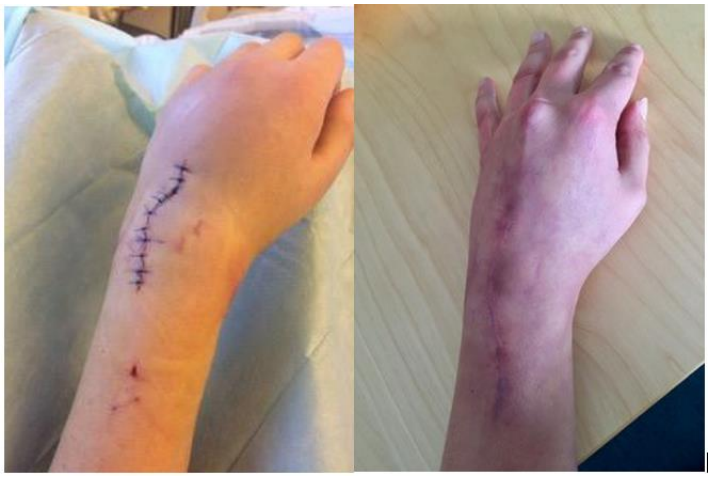

Figure 1: *Left picture: Patient´s left upper extremity after several operations in 2015. The hand was swollen for weeks and burning pain began at the metacarpophalangeal joint including the thumb. Right picture: Decrease in swelling of the left hand after lymphatic drainage. Livid discoloration and marbling of the hand with difference in skin temperature with up to 4,5°C were present. The pain increased and became unbearable. The whole hand was affected and became more and more sensitive.*

The pain intensity increased with exercises, cold weather, light touch, cold or warm water. An explanation based on other illnesses was excluded. Another surgical, i.e. causal, treatment could also be ruled out. The CRPS criteria were met for persisting pain and for the clinical categories' hyperalgesia, allodynia, and asymmetry in skin temperature, change in skin colour, asymmetry in sweating, edema, and change in hair growth, decreased mobility, and weakness. In a side comparison, differences in skin temperature of a maximum of up to 4.5 °C were measured. Due to loss of function and persistent pain, she had to train her handedness by being originally left-handed. In professional terms, the patient lost her job in the ambulance service after the arthrodesis in 2015. She began studying in 2017, which she had to finish again due to exacerbated pain and psychological stress. In addition, she developed suicidal ideation in 2015, which took a certain form in 2017 due to a borderline personality disorder. During the presentation in our outpatient department, the pain intensity was rated with a maximum of 10/10 on the visual analog scale (VAS), a minimum of 5/10 on VAS and a pain average of 6/10 on VAS. On the intensity scale of the brief pain inventory 26 out of 40 points were achieved [6]. The pain syndrome was refractory to comprehensive conservative treatment and only 30% of pain reduction could be achieved with previous therapy. A significant influence on mood (7/10), sleep (9/10) and enjoyment of life (7/10) with the brief pain inventory was described. On the pain interference scale, 39 out of a maximum of 70 points were achieved. A total value of 46 (max. 52) was achieved on the pain catastrophizing scale [7]. Clinically, she showed marked allodynia of the distal forearm and the entire hand, accompanied by swelling and changes in skin colour (Figure 2).

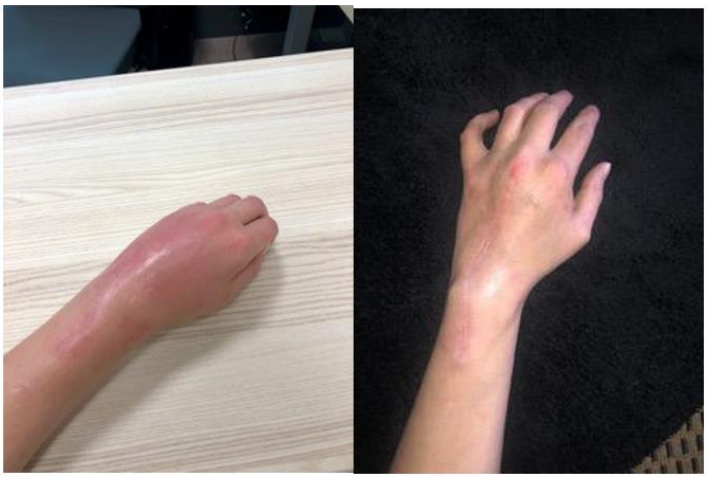

Figure 2: *Left picture: Patient´s left upper extremity one week before implantation of the neuromodulation system in May 2019. Acute swelling, severe burning pain und shiny skin dominated the clinical appearance. Movement of the fingers was hardly possible. The desire for an amputation was great at this point. Right picture: Two weeks after the operation the swelling was completely gone, the skin color was normal, and movement of the fingers was possible. The pain and sensitivity of the hand improved significantly.*

Motor function tests of the arm and hand could not be performed in severe pain and arthrodesis with functional disorder of the wrist and thumb. There was an intense desire for a forearm amputation and a significantly reduced confidence in medical activities. As a last attempt, we provided the indication for stimulation of the spinal cord, which could be carried out in May 2019 eight years after disease progression. The patient underwent cervical SCS trail using an Abbott Medical Octrode lead (Prodigy MRI™) inserted at level T8 and ended at level C5. After a successful trial of two weeks, a permanent implantation of the impulse generator was carried out. During follow up consultation 12 month later, pain at minimum decreased to 2/10 on VAS with an average intensity of pain of 3/10 on VAS and a maximum of 6/10 on VAS. Physical and occupational therapy and psychotherapy were continued as before. Reduction of pain was rated with 70% with the ongoing stimulation plus conservative treatment. The intensity scale of the brief pain inventory was reduced to 14 points (max. 40). Furthermore mood (5/10), sleep (5/10) and enjoyment of life

(5/10) of the brief pain inventory improved with a total of 25 points (max. 70) on the pain interference scale, as well as the pain catastrophizing scale with a sum score of 18 (max.52) points. Allodynia decreased with a residual slight presentation combined with a slight hyperalgesia of the left wrist. Motor function could not be improved due to arthrodesis. Reduced hairiness and skin atrophy remained. Due to a dislocation, the generator was revised into a deeper gluteal layer in May 2020. Today, that means three years later, the patient was satisfied with spinal cord stimulation, even though she described a slight increase in pain intensity during winter while consulting our outpatient clinic in March. Pain intensity was rated with a minimum of 3/10 on VAS, a maximum of 9/10 on VAS and an average of 5/10 on VAS. On the pain intensity scale of the brief pain inventory, the patient achieved 22 points (max. 40). Additional to previous therapy, she was working with a therapy dog. Pain reduction was rated as 50% with the ongoing treatment. Impact on mood (5/10) was unchanged, sleep (7/10) worsened slightly, and enjoyment of life (3/10) improved compared to the years before with an almost stable value on the pain interference scale of 26 points. Even more, sum score of pain catastrophizing scale was stable (18/52). Despite the renewed increase in pain, the patient was overall still satisfied with the neuromodulation and would have it performed again. The patient did not want to interrupt the stimulation at any time, especially given the positive effects on quality of life and pain perception. Oral medication could be reduced over the following years. To avoid habituation effects of stimulation, we recommend switching between BurstDR™ and continuous mode.

Discussion

Several studies having shown before that SCS is a safe and effective procedure for the treatment of pain conditions such as failed back surgery syndrome, refractory angina pectoris, peripheral vascular disease and CRPS type I[8,9]. Neurostimulation of spinal cord in patients with CRPS can reduce pain and improve health-related quality of life in a more effective way than physical therapy alone [10]. Success depends on strict selection criteria of patients with complex regional pain syndrome type I, lasting for at least 6 months with a mean intensity >/= 5 on VAS and being not responsive to conservative therapy. Complications reported after the implantation of a neurostimulation system were mainly related to hardware problems. In our view, the low periprocedural risk to patients justifies offering neurostimulation treatment. Consistent data for long-term efficiency of spinal cord stimulation in CRPS are lacking so far with reported diminishment between 2-3 years after implantation on the one hand and a still significant effect on pain reduction after 88 months [11,12]. Best results were seen within one year after implantation in patients under 40 years and receiving SCS within one year after onset with a slight decline overtime. For our treatment recommendation of pain therapy, the factor of time is a very important predictor for a positive response in patients with chronic pain conditions. Suitable interventions options should be carried out before irreversible changes in pain memory and irreversible restructuring processes of central pain take place. The longer the painful condition persists, the more limited the options for adequate pain treatment. In comparison to current studies on the successful treatment of pain syndromes in lower extremity with dorsal root ganglion (DRG) stimulation with a greater improvement in pain and quality of life, data for upper extremities are missing so far due to lacking approval for the procedure on cervical spine [13-15]. Notable, in a case report of CRPS in upper extremity, DRG stimulation was performed accidentally due to an anatomical obstruction on spinal cord with positive impact on pain reduction [16]. Psychological tests are recommended to evaluate comorbid psychiatric conditions and dysfunctional cognitions before implementing medical treatment strategies. Pain catastrophizing and incorrect beliefs are common in patient with CRPS and may be critically involved in maintenance of primary features of the pain syndrome and in responses to treatment. Even though as described before, that psychiatric disorders should exclude patients from stimulation of the spinal cord, our patient received a positive effect on psychiatric condition and pain catastrophizing through neurostimulation. We cannot conclude that pain catastrophizing should be a contraindication, as similarly reported by Lame. We suggest that SCS is an effective treatment for disabled patients with severe pain that is unresponsive to conventional therapy and should be considered in early stages to prevent severe impact on mental and physical health. Even in long lasting CRPS SCS, as a minimal- invasive procedure, has a positive impact on pain reduction and catastrophizing. Its benefit might be underestimated due to lack of larger studies and should not only be considered as a last chance therapy.

Conclusion

Complex regional pain syndrome in distal upper extremity can be treated successfully with SCS with no side effects. Furthermore, neurostimulation has positive effects on mental health and catastrophic pain perception. We suggest that SCS should be considered early in the course of disease as a possible therapeutic option for complex regional pain syndromes in the upper extremity to prevent extremity amputation and major impact on mental health.

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