

# **Successful treatment of intractable complex regional pain syndrome type I of the knee with dorsal root ganglion stimulation: A case report**

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## ABSTRACT

**Objectives:** To report on the efficacy of dorsal root ganglion (DRG) stimulation in a patient with complex regional pain syndrome (CRPS) type I of the knee.

**Materials and Methods:** A 48-year-old woman with CRPS type I of the right knee, diagnosed according to the Budapest criteria set, received DRG stimulation for intractable CRPS type I of the knee.

**Results:** After a successful trial period with three DRG stimulation leads on spinal levels L2, L3 and L4 (covering 90% of the painful area of her knee), a definitive pulse generator was implanted. Three months after implantation, the entire painful was area covered and the patient reported a numeric rating scale score of 1-2.

**Conclusion:** Placement of three DRG stimulation leads at levels L2, L3 and L4 in a patient with intractable CRPS type I of the knee resulted in major pain relief. We recommend further investigation of the effect of DRG stimulation on pain due to CRPS of the knee.

## KEYWORDS

Complex regional pain syndrome type I, dorsal root ganglion stimulation, knee, numeric rating scale, numeric rating scale, treatment

## INTRODUCTION

Complex regional pain syndrome (CRPS) is a collection of locally appearing painful conditions that mainly occur distally and exceed in both intensity and duration the expected clinical course of the original trauma. In a minority of patients, the condition is restricted to the knee. Recently, a systematic review was performed to find evidence for the diagnosis of CRPS of the knee. The authors concluded that this diagnosis has been described before and the best therapy is yet to be found (1).

Dorsal root ganglion (DRG) stimulation is an effective and safe stimulation technique, and previous reports have implicated the DRG in the development and persistence of chronic pain. The ganglion houses cell bodies of primary sensory neurons, including cells that transmit pain information to the central nervous system (2). Deer *et al.* reported the successful use of DRG stimulation in 10 patients with chronic, intractable neuropathic pain of the trunk and/or limbs. These patients received DRG stimulation for 3-7 days. All patients experienced pain relief in the targeted anatomical regions, with reported improvement ranging from 17% to 100%(3). Liem *et al.* reported on 32 patients with pain of the back, leg, and foot treated with DRG stimulation; at 6 months after implantation,  $\geq 50\%$  of the patients had 50% or more pain relief (4).

The above-mentioned favorable results of DRG stimulation of body parts other than the extremities led us to stimulate the DRG in a patient with CRPS type I of the knee that appeared to be intractable.

## CASE REPORT

### History

We retrospectively present a case of CRPS in a 48-year-old woman who suffered complaints of the right knee for five years. One week after a diagnostic arthroscopy to rule out any meniscal problem, she developed CRPS type I of the knee. This patient was referred to our department for a second opinion. She had already been extensively treated with different types of oral medication. In addition, a lumbar sympathetic block resulted in no clinically significant relief of symptoms, and physical therapy also failed. In particular, the patient's pain (reported to be the most disabling factor) did not decrease.

### Examination

When the patient visited our department, the pain on the lateral side and within the knee was described as "irritable and aching" with a score of 6-9 on a numeric rating scale (NRS) ranging from 0 ("no pain") to 10 ("extremely painful"). In addition, she experienced allodynia, hyperalgesia, and tingling around her right knee. The sensory symptoms were not

limited to the innervation area of a single nerve. She also mentioned discoloration of the whole knee, a colder temperature compared to the left knee, trophic changes in leg hair, and motoric dysfunction, but no sweating changes or edema.

Physical examination revealed a swollen, cold, and dark-colored right knee. Palpation was very painful, especially around the patella and the medial tibia head. Allodynia and hyperalgesia around the whole knee and movement restriction also were present. Exploratory neurological examination revealed no other signs as the earlier-mentioned sensory disturbances. The area of the sensory disturbances could not be explained by a mono neuropathy or other neurological etiology. We confirmed the diagnosis CRPS type I of the knee, based on the Budapest criteria set. In addition, thermography revealed the right knee to be colder compared to the left knee.

### Treatment

Our treatment started with different types of oral medication, followed by ketanserin/carnitine intravenously. None of these provided any relief of her symptoms; the allodynia even became worse and edema developed. Therefore, we considered neurostimulation as a treatment. In view of the limited area involved and of earlier experience in our department of disappointing results with dorsal column stimulation in patients with comparable localized CRPS, we decided to apply DRG stimulation and included her in a medical ethical committee-approved observational study in our department for the use of DRG stimulation. The patient was informed and gave written consent.

### DRG stimulation

The DRG was approached in the same way as described by Liem *et al.* (4) We implanted three DRG stimulation leads at spinal levels L2, L3, and L4, at the right side of the spine (Fig. 1).

The treatment started by implanting one DRG stimulation lead at spinal level L3. The patient reported stimulation vibes within her knee, but these did not cover the entire area. A second DRG stimulation lead was implanted at level L4. The stimulation now covered more of the painful area, but still not everything. So we tried a third DRG stimulation lead on level L2. This level was chosen because of earlier experience with DRG stimulation in our department: sometimes the best level for DRG stimulation is above or below the level one would expect based on the dermatome map. After implantation of this third lead, the stimulation covered almost the entire area; only one spot on the lateral knee side was not covered.

When one lead was in place, it was connected to an external neurostimulator. The neurostimulator was programmed to ensure that the paresthesias were directed to the correct location; the same procedure applies to the other two leads. The parameters of stimulation are presented in table 1.

After eight days of stimulation coverage, the patient reported a substantial decrease in pain intensity. One spot on the lateral knee side was still not covered, but she was very pleased with the results and requested to proceed with permanent implantation. Before, she scored the pain as NRS 6-9, whereas now she scored it as 1. One week later, the implantable pulse generator was placed in the patient's left buttock (Fig. 2).



**Figure 1.** Dorsal root ganglion stimulation leads with four electrical contacts at levels L2, L3, and L4



**Figure 2.** Implantable pulse generator placed in the left buttock

**Table 1.** Parameters of dorsal root ganglion stimulation in a 48-year-old female patient

		Lead 1 (Spinal level L2)	Lead 2 (Spinal level L3)	Lead 3 (Spinal level L4)
Electrode configuration	1	N	-	N
(+, - or N)	2	N	N	N
	3	+	+	+
	4	-	N	-
Pulse width ( $\mu\text{sec}$ )		170	170	160
Frequency (Hz)		20	20	20
Amplitude ( $\mu\text{A}$ )		700	1030	500

+, positive; -, negative; N, neutral

One month post-implant, she stated that the entire painful area was covered and graded her pain as 1-2 during stimulation. In addition, movement of the right knee had improved. The device was used 24 hours a day. At 3-months follow-up the NRS score was still 1-2 during stimulation. She did not feel the stimulation vibes anymore, but she certainly could tell when the device was switched off, as her pain returned within minutes. On the other hand, when it was switched on, the effect kicked in within one minute. The patient will further be periodically monitored over one year.

## DISCUSSION

This patient with CRPS type I of the knee failed to respond to multidisciplinary pain management. The symptoms fit the Budapest criteria set, but infrapatellar nerve injury should be considered as differential diagnosis (5). An electromyography was not performed, so we cannot be 100% sure that there was no demonstrable nerve damage. In case of nerve damage, the diagnosis should be CRPS type II. The DRG stimulation therapy resulted in a clinically significant result: the pain level dropped from an initial NRS score of 9 to 1-2 and the movement of the knee became better. We chose DRG stimulation due to earlier experience in our department with CRPS of the knee: most patients barely respond to conservative treatment and dorsal column stimulation. Our first results with DRG stimulation therapy for CRPS of the knee are promising and in line with others (3, 4). However, none included a patient with CRPS of the knee. In addition, our follow-up was three months whereas that of Liem *et al.* (4) was six months, making it difficult to extrapolate their results to ours. We realize that this is a case report and therefore had limitations concerning the generalizability across patients; nevertheless, we consider the present results to be encouraging and the DRG to be a potential new neural target for reducing chronic neuropathic pain due to CRPS of the knee.

## CONCLUSION

This report indicates DRG stimulation to be an effective therapy for reducing chronic neuropathic pain due to CRPS of the knee. Stimulating the DRG covered the entire painful area, and up to three months post-implant the initial NRS score of 9 had decreased to 1-2. Long-term results are not yet available, and more patients with similar complaints need to be investigated. If such studies replicate the present findings, a controlled study should be performed in order to draw more definite conclusions.

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