

# **DRG spinal cord stimulation as remedial solution for patients with severe pain due to Anterior Cutaneous Nerve Entrapment Syndrome: A case series.**

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## ABSTRACT

**Objectives:** anterior cutaneous nerve entrapment syndrome (ACNES) is a debilitating neuropathic pain condition. A small portion of patients does not respond to any of currently available treatment modalities. These patients, often young women, might benefit from targeted spinal cord stimulation of the dorsal root ganglion (DRG).

**Methods:** This retrospective case series describes 5 ACNES patients who were referred from a Dutch dedicated tertiary referral center to collaborating sites with extensive experience in DRG stimulation to be implanted with a DRG Axium System (St. Jude / Abbott, Illinois, USA) in the period of 2013-2016. Numeric pain rating scores at routine 6- and 12 month follow-up visits were analyzed.

**Results:** Three patients experienced >50% pain reduction at 12 months follow-up. Four patients experienced device-related complications, such as lead dislocation, lead breakage, pain at the battery site and overstimulation.

**Conclusions:** This case series suggests DRG spinal cord stimulation can be safe and effective for some patients with persistent pain due to ACNES.

**Keywords:** Dorsal root ganglion, Anterior Cutaneous Nerve Entrapment Syndrome (ACNES)

## BACKGROUND

Chronic abdominal pain due to anterior cutaneous nerve entrapment syndrome (ACNES) is a potential debilitating condition caused by entrapped sensory nerves within the abdominal wall. Patients present themselves with continuous abdominal pain, attenuated by certain positions or exercise, often severely interfering with daily life. Physical examination will reveal a finger-tip-point of maximum pain over the abdominis rectus muscle, accompanied with local sensory disturbances and a positive Carnet's test. (1) Captured DN4 scores will further indicate neuropathic pain, with burning or painful cold sensations and hypersensitivity to brushing of the skin.

A therapeutic algorithm has been developed consisting of infiltrations with local anesthetics, pulsed radio frequency treatment or TENS and finally surgical resection of the intercostal nerve endings at the level of the anterior rectus sheath.(2,3) However, a small portion of patients does not respond to any of these currently available treatment modalities. These patients, often young women, might benefit from targeted spinal cord stimulation of the dorsal root ganglion (DRG). DRG stimulation has proven to be effective in other neuropathic pain syndromes, such as chronic postherniorrhaphy inguinal pain (CPIP). (4)

Sensory input of the abdominal wall is supplied by intercostal nerves originating from DRG levels T7 to T12. Patients can indicate their point of maximum pain in a specific small area within corresponding dermatomes, which provides the first clue to where lead placement should be. Retrograde paresthesia mapping can confirm if one or multiple DRG's contribute to the painful area.(5)

Up to date, there is no literature available on the efficacy of DRG stimulation in patients with ACNES. This case series is the first to address long-term results of this last resort treatment option for therapy resistant patients.

## METHODS

This retrospective series describes 5 ACNES patients who were referred from a Dutch dedicated tertiary referral center to collaborating nearby implanting sites with extensive experience in DRG stimulation in the period of 2013-2016. Two patients received treatment in the St. Antonius Hospital, Nieuwegein; two in St. Elisabeth Hospital, Tilburg and one in Diakonessen Hospital, Zeist. Screening was performed following the Dutch Neuromodulation Society Guidelines and included a psychological assessment.

Data was gathered from the electronic patient files in the referring center and the three implanting sites. Patients had provided written consent forms for the use of their anonymized patient related outcome measures. Numeric pain rating scores (NRS, rang-

ing from 0-10) were used to assess average levels of pain prior to baseline and follow-up visits per standard protocol. The primary endpoints were pain reduction at 6 and 12 months follow-up, if available.

## Patients

### *Patient 1*

A 35-year-old female (BMI 35,4) was referred suffering from ACNES just below the right lower ribs since 2007. She had undergone her first neurectomy in 2007 after multiple infiltrations only resulted in a few days of pain remission. All other treatment modalities had failed. After this neurectomy she had been pain free for 6 months, but unfortunately complaints recurred. Two more explorations with resection of a neuroma followed, without effect. Using opiates, she still experienced pain NRS 8 and was therefore referred and implanted with a DRG Axiom System (St. Jude / Abbott, Illinois, USA) by the end of 2013 after a successful one week trial. The surgical procedure lasted 46 minutes for one lead at T9 right. There were no postoperative complications. She had complete pain remission at 6 months follow-up (NRS 0). Unfortunately, after 7 months she experienced a lead dislocation with recurrence of her pain. After revision she again obtained adequate pain reduction (NRS 0 at 12 months).

### *Patient 2*

A 26-year-old female (BMI 26,7) was referred after developing ACNES during her pregnancy in 2010 in the lower left abdominal quadrant. After conservative treatment measures, she had undergone two neurectomy procedures that also were to no avail. She was referred to an implanting center with an NRS 9 whilst using high doses of pain medication. She was accepted for DRG all-in-one procedure at the start of 2016. Paresthesia mapping according to protocol was carried out preoperatively. The patient described 100% paresthesia coverage at level L2. Just days before the procedure was scheduled, she described intermittent complaints in the right lower abdomen similar to her complaints at the left side. This was left expectant and one lead was implanted in an all-in-one procedure on level L2 left. She reported NRS 5 at 1 month follow-up and was able to lower her medication use. However, the complaints on the right side persisted and a lead breakage after 4 months prompted lead replacement and addition of an extra lead for the right side. She experienced an overall 50% pain reduction (NRS 4) 6 months after the initial implantation.

### *Patient 3*

A 50-year-old male (BMI 25,7) was referred and implanted in November 2015 after a neurectomy for ACNES had improved his pain in the anterior abdominal wall, although radiation pain to flank and back persisted resulting in a high pain level (NRS 9). An all in

one procedure with placement of one lead at DRG level T9 right was performed. Multiple settings were extensively explored, but although the paresthesia coverage was 100%, there was 0% pain reduction at 6 months. Patients medication use diminished after implantation, but pain levels remained high. After the device was turned off for a few weeks, patient had the feeling the device actually might have some small effect and therefore did not opt for explantation.

#### *Patient 4*

An 18-year-old female (BMI 20,5) was referred suffering from ACNES in the right lower quadrant. An infiltration with lidocaine in the point of maximal pain resulted in 6 hours of complete pain remission. Finally, a neurectomy resulted in only two days of pain remission, after which she had posterior lateralization of pain in the according segment of the involved intercostal nerve (clinically T10-11 region). She was referred and implanted in April 2013 with an all-in-one procedure with one lead placed at T10 right. Her NRS scores dropped from 8 pre-operatively to 6 within 6 months, but she experienced severe pain at the battery site. The battery was moved to the abdomen and she reported NRS 3 at 12 months follow-up.

#### *Patient 5*

A 60-year-old male presented himself with excruciating pain in the left lower quadrant of the abdomen in 2014. These complaints arose suddenly without any evident cause, but six years prior to these symptoms he had undergone bladder surgery because of a urinary tract cancer. After extensive lab and imaging tests, there were no clues for cancer recurrence and no other explanation for the pain was found, but ACNES. TENS and medication had no effect on the pain. A neurectomy of multiple anterior cutaneous nerves in the left lower abdominal wall rendered complete remission of pain, albeit temporary, since after three months pain recurred. An all-in-one procedure was performed with lead placement on level T10, T11 and T12 left after positive mapping in February 2016, with a pre-operative NRS of 8. At 6 months follow-up his NRS had dropped to 5, but after a nefrectomy due to cancer recurrence he developed right-sided complaints as well. Two additional leads were implanted at level T10 and T12. Due to overstimulation-like symptoms, T11 left was turned off. Patient came into a palliative setting because of metastatic disease, and therefore no 12 month follow-up could be obtained.

## **RESULTS**

Three patients experienced >50% pain reduction at 12 months follow-up. Four patients experienced device-related complications, such as lead dislocation, lead breakage, pain at the battery site and overstimulation.

**Table 1.** Pain reduction at 6 and 12 months of 5 patients.

	Baseline NRS	6 month NRS	12 month NRS	Pain reduction At 6 – 12 month
Patient 1	8	0	0	100% - 100%
Patient 2	9	5	4	44% - 56%
Patient 3	9	9	9	0% - 0%
Patient 4	8	6	3	25% - 62%
Patient 5	8	5	-	37%

**DISCUSSION & CONCLUSIONS**

This case series suggests DRG spinal cord stimulation can be safe and effective for some patients with persistent pain due to ACNES, although certain pitfalls should be addressed. Two patients developed bilateral complaints in the work-up to or during neuromodulation treatment. In the overall ACNES population, bilateral complaints, which are practically always symmetrical and at the same level, exist in some 13% of patients. Whether this remarkable observation is attributable to an entrapment in the abdominal wall of two separate nerves or to a mirroring phenomenon at the level of the spinal cord/brain is unclear. In our experience, often both sides require treatment. This could pose a technical challenge to the implanting physician if multiple leads are needed. We suggest that it is certainly important to assess in every ACNES patient whether pain distribution is only one-sided.

Retrograde paresthesia mapping preceding implantation is essential to assess whether multiple DRGs contribute to the painful area.

Procedural and device-related complications such as lead breakage, lead dislocation and pain at the battery site mask the positive effect at standard follow-up points. Technical innovations could improve the overall success rate of DRG spinal cord stimulation.

Further investigations on this form of therapy for patients with persistent pain due to ACNES seems very relevant, since there is a group of patients who remain in severe pain after various treatment modalities for this intriguing pain syndrome.

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## COMPETING INTERESTS

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