

Treatment of chronic refractory coccydynia with peripheral nerve field stimulation: A novel case

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Abstract

Coccydynia or coccyx pain is a rare form of low back pain located below the sacrum and above the anus. The causes of this condition vary, including previous trauma, underlying conditions (such as infection or neoplasm), and idiopathic causes. Our purpose was to describe the successful treatment of chronic refractory coccydynia with the application of sacral area neuromodulation. A 54-year-old female patient had a 9-year history of chronic drug-resistant coccydynia associated with low back pain and right-sided sciatica. Her clinical examination revealed bilateral pain on palpation of the sacroiliac joint areas. Following a successful trial, the patient underwent implantation of a peripheral nerve field stimulation (PNFS) system under local anesthesia. The electrodes were placed bilaterally at the sacroiliac joint area (S1-S2 level), which was the optimal position at the site of maximal pain, and the implantable pulse generator was placed in a subcutaneous pocket at the right gluteal area. She showed significant pain relief immediately postoperatively, became pain-free a few days later, and remained pain-free at 6-month follow-up. Data regarding neuromodulation, and particularly PNFS, as a treatment for coccydynia and other non-specified kinds of low back pain are still limited. Our case is therefore useful to depict this minimally invasive technique as a modern option in the armamentarium of specialists who treat patients suffering from chronic refractory pain syndromes. PNFS seems to be a promising therapeutic option for chronic refractory coccydynia and larger studies are necessary to confirm the value of this finding.

Keywords

coccydynia, neuromodulation, neuropathic pain, peripheral nerve field stimulation, sacroiliac joint pain

Introduction

Coccydynia or coccyx (tailbone) pain is a rare form of low back pain, which is located below the sacrum and above the anus.^[1-4] It may worsen with prolonged sitting or standing, leaning back, or rising from a seated position.^[4] Risk factors associated with the development of coccydynia in adults include obesity, female gender, and pregnancy.^[1,2,5] The causes of this pathology vary. It may be idiopathic, or

caused by former trauma or an underlying condition, such as infection or neoplasms.^[3,5]

Sacroiliac joint pain is also a form of mechanical low back pain. The patients suffering from this condition can experience unilateral pain bellow the L5 dermatome.^[5] Risk factors for this pathology include several lower extremity deformities, inflammatory arthritis, previous spine surgery, trauma, and pregnancy.^[2,4]

Peripheral nerve stimulation (PNS) and peripheral

nerve field stimulation (PNFS) (or targeted subcutaneous neurostimulation)^[6] are methods of neuromodulation of the peripheral nervous system, involving the delivery of electrical stimuli using implanted electrodes either over a target nerve or over the painful area (field) with the goal of modulating neuropathic pain.^[6] Due to their simplicity and minimal invasiveness, PNS and PNFS may be preferable to more invasive neuromodulation procedures and have been increasingly used in recent years for the management of chronic pain.^[6,7]

With this report we aim to present the application of sacral area neuromodulation in the treatment of a patient with chronic refractory coccydynia, accompanied by lumbar pain, sciatica, and sacroiliac joint pain.

Case description

A 54-year-old woman with chronic drug-refractory coccydynia with concomitant lumbar pain and right-sided sciatica (S1 dermatome) for 9 years was referred to our Department's Stereotactic and Functional Neurosurgery service by her pain medicine physician, who considered her a candidate for neuromodulation therapy. Upon physical examination, the patient showed no motor or sensory deficit. Laseg-

ue sign was negative and she had normal tendon reflexes on both sides. However, the patient experienced pain (visual analogue scale score: 5/10) during palpation of the bilateral sacroiliac joint areas. She agreed to undergo a neuromodulation treatment.

In the operating room, the patient was placed in a simple prone position, and the lower lumbar, sacral and gluteal areas were prepared bilaterally in a sterile manner (as in lower lumbar surgeries). Implantation of an external Peripheral Nerve Field Stimulation (PNFS) system with cylindrical electrodes was performed, using a percutaneous technique (Tuohy needle), under local anesthesia and C-arm guidance. The electrodes were placed bilaterally at the sacroiliac joint area (S1-S2 level), which was the intended optimal position centered at the site of maximal pain (which was marked on the skin), and the implantable pulse generator (IPG) was left externally. Following a trial period, where the patient showed remarkable improvement in pain severity, she was considered candidate for implantation of a permanent PNFS system. Again under local anesthesia, the permanent IPG was thus placed in a subcutaneous pocket at her gluteal area (**Fig. 1**). No perioperative, early or late complications were observed. The patient showed significant improvement in pain relief post-operatively, became pain-free a few days later, and remained pain-free at 6-month follow-up.

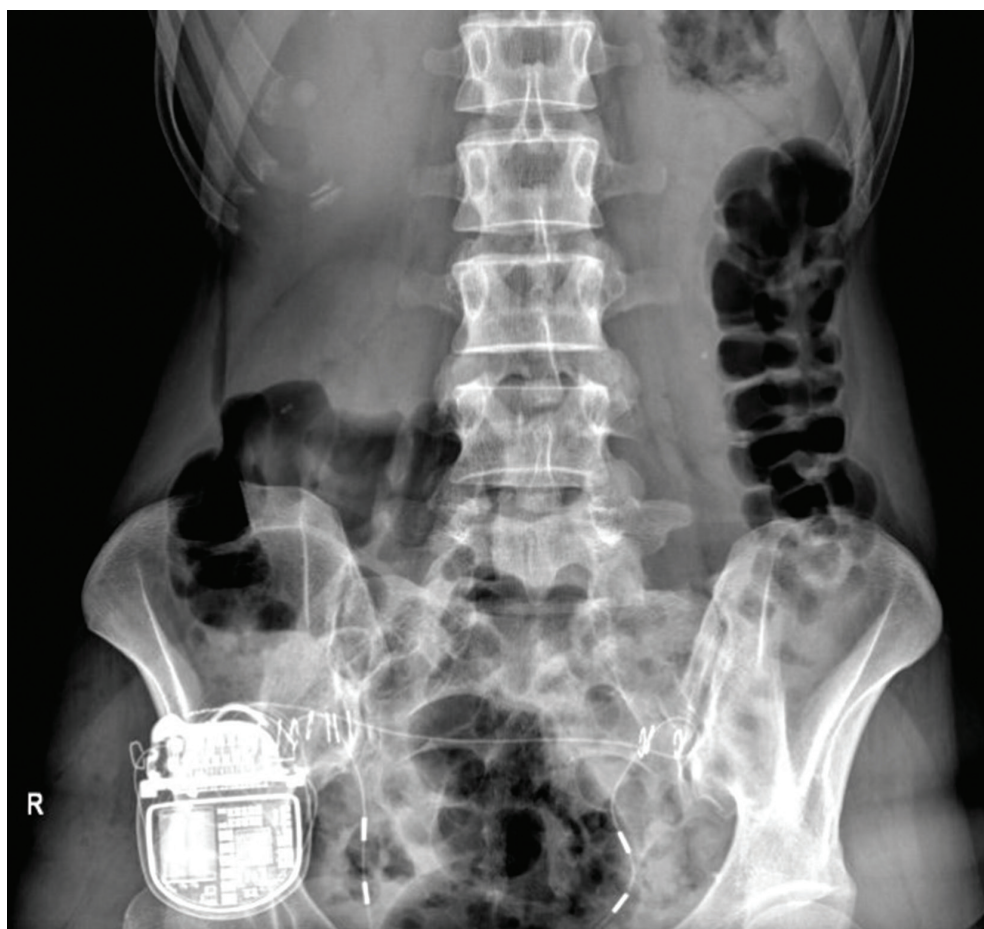


Figure 1. The implanted peripheral nerve field stimulation system of our patient (postoperative anteroposterior X-ray).

Discussion

There are various treatment options available for symptomatic therapy of chronic low back pain, including failed back surgery syndrome (FBSS).^[1] They consist of conservative, pharmacological, and surgical treatments. The treatment for coccydynia and other non-specific forms of low back pain is characterized by progressive invasiveness, usually starting with non-invasive methods such as nonsteroidal anti-inflammatory drugs (NSAIDs), physical therapy, and minimally interventional treatments (e.g. injections), before considering surgery. Non-surgical management provides pain relief in the majority of patients.^[1] Among surgical treatments, coccygectomy is considered to be one of the most thoroughly investigated treatment options and may be beneficial for refractory cases, at the cost of an overall high complication rate.^[1]

Deep brain stimulation (DBS), cortical and subcortical stimulation, spinal cord stimulation (SCS), PNS, PNFS, and also “hybrid” techniques, a term used to describe combinations of the above techniques^[6], are described to be valid neuromodulation treatment options in cases of pain syndromes, such as neuropathic pain disorders (including most types of low back pain, lumbosacral radiculitis that is refractory to pharmacological and non-surgical interventional treatments, FBSS, and diabetic peripheral neuropathy)^[8], complex regional pain syndrome, refractory headache, intractable musculoskeletal pain and fibromyalgia.

Although DBS and cortical stimulation are not established in the management of chronic pain, these techniques may be used in refractory cases.^[9] On the other hand, PNFS has been shown effective and safe to relieve significantly symptoms of chronic low back pain in a recent series.^[10] Overall, there is mixed evidence supporting the efficacy of the above techniques^[9] and the data regarding neuromodulation, and particularly PNFS, as a treatment for coccydynia and other non-specific kinds of low back pain are still limited.^[3,6] Therefore, there are described criteria regarding neither patient selection nor postoperative outcome evaluation. Following current management principles that apply to other chronic pain syndromes, we think that Pain Medicine physicians have the primary role to determine a patient as a candidate for neuromodulation treatment and treatment-resistance should be a prerequisite. Similar to other chronic pain syndromes, the reduction/elimination of pain should be the main criterion for assessing treatment outcome.

Additionally, we would like to note that since there are currently no clear recommendations on when and how PNFS should be applied in the treatment of coccydynia, further research in the field is encouraged. With increasing experience, studies with high quality metadata are needed, aiming to clarify the following aspects: 1) patient selection criteria, 2) proof of the method's feasibility, 3) outcome definition, 4) procedure evaluation, 5) success rate, and 6) complication incidence.

Finally, considering our patient's experience, we would like to underline, as advantages of the used method, the

minimum time needed for postoperative observation (a few hours), the avoidance of hospitalization, the postoperative independency of the patient without restrictions in her socialization and the lack of complications. The long-term outcome of her treatment is the next important aspect to be determined.

Conclusions

PNFS seems to be a promising minimally invasive neuromodulation therapeutic option in carefully selected patients suffering from chronic refractory coccydynia and larger studies are mandatory to confirm the value of this finding.

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Competing interests

The authors have declared that no competing interests exist.

Author contribution

Concept, design, the definition of intellectual content, ‘guarantor’: I.M.; data acquisition, data analysis, literature search: I.M. and G.T.; manuscript preparation, manuscript editing, and manuscript review: all authors.

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