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Spinal Cord Stimulation of the Sacral Region as a Treatment for Intractable Coccygodynia: A Case Study

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Background: Coccygodynia is a notoriously difficult condition to manage. Spinal cord stimulation (SCS) may be a promising therapeutic option for those suffering from chronic coccygeal pain. To our knowledge, there are limited reports of using SCS to target the sacral region for the treatment of intractable coccygodynia.

Case Report: A 50-year-old woman with refractory coccygodynia underwent permanent implantation of an SCS device following a successful SCS trial. SCS leads were inserted using a retrograde approach to target S2-S4 bilaterally. During follow-up, the patient reported > 90% improvement of coccygeal pain and her self-reported quality of life dramatically improved.

Conclusion: This case report describes the successful use of the sacral region as a target for SCS in the management of intractable coccygodynia. SCS may be considered an effective treatment option when all other conventional methods have failed.

Key words: Coccygodynia, neuromodulation, sacral nerve stimulation, spinal cord stimulation

BACKGROUND

Coccygodynia is defined as pain in the coccygeal region. Diagnosis of this condition is clinical and made by obtaining a proper history and physical examination. Coccygodynia is notoriously difficult to manage, and often leads to chronic pain (1-8). Chronic, refractory coccygodynia describes coccygeal pain that has persisted despite multiple attempts at therapeutic management.

The pelvic region encompasses a vast array of complex anatomical structures. Contained within this region are visceral organs as well as an intricate network of neural fibers with somatic, sympathetic, and parasympathetic function (9). The anatomical complexity of the pelvic region likely contributes to the challenge in managing this condition.

The number of different etiologies associated with the development of coccygodynia likely also contributes to the difficulties seen in management. Possible causes include direct external trauma, childbirth injury, bone spurs, and even infection or malignancy (10). Other derivatives related to an underlying psychological etiology must be ruled out.

Currently, there is no clear consensus on the proper management of coccygodynia. Conservative treatment measures include cushioning devices, nonsteroidal anti-inflammatory drugs, and coccygeal manipulation (1). Chronic, refractory pain may benefit from more aggressive interventional management including injection therapy (1). In the last decade, surgical removal of the coccyx, or coccygectomy, has grown in popularity for the use of treatment-resistant coccygodynia. Coccygectomies have been shown to be effective in

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relieving pain symptoms; however, this invasive intervention is not without risk (4-8). Complications such as infection, wound dehiscence, delayed healing, wound hematomas, intestinal tract injury, and rectal prolapse have been documented in the literature (8). To prevent unnecessary surgery, identifying efficacious and minimally invasive treatments must be an area of focus. Neuromodulation may prove to be a more reliable and less invasive therapeutic option for cases of treatment-resistant coccygodynia.

Electrical neuromodulation has proven beneficial in the management of a number of different pain syndromes. The use of spinal cord stimulation (SCS) is one of the most widely used modalities of neuromodulation. This technique involves the implantation of electrodes in the dorsal epidural space, most often within the lower thoracic region (9). There are limited reports of the use of SCS within the terminal parts of the spinal cord, such as the conus medullaris, or terminal regions of the dural sac. Studies describing neuromodulation of the sacral region have reported success in the management of treatment-resistant pelvic pain syndromes (9). The use of SCS within the sacral region for the treatment of intractable coccygodynia has yet to be elucidated. We describe the case of a middle-aged woman treated with SCS for the management of refractory coccygodynia.

CASE

The patient presented as a 50-year-old woman with chronic, intractable coccygeal pain. Her pain was described as 8 of 10 on average, persistent, sharp, and throbbing. The pain was worse with bending and sitting, with no radiation to the lower extremities. Sacrococcygeal x-ray and lumbosacral magnetic resonance imaging were unremarkable. The patient had attempted pharmaceutical management with nonsteroidal anti-inflammatory drugs, antineuropathic agents, and antidepressants without benefit. She had undergone a series of ganglion impar blocks, sacrococcygeal ligament blocks, and caudal epidural steroid injections with > 80% temporary relief of symptoms from the ganglion impar block only. Given past success with this block, a ganglion impar radiofrequency lesioning was planned and performed. The patient reported reduction in pain immediately following the procedure; however, within days her symptoms spontaneously returned.

A coccygeal nerve block was then scheduled. She reported > 80% reduction of the coccygeal pain, therefore coccygeal nerve radiofrequency ablation was performed. One month's follow-up revealed worsening of symptoms. Given that the patient's symptoms were unrelieved with both medication management and interventional pain procedures, we tried SCS of the sacral nerves for her worsening pain and new-onset radiculopathy. The patient was counseled regarding the risks and benefits of the procedure and elected to proceed.

SCS Trial

The trial was performed using an anterograde approach with cephalic advancement of the electrode via the sacral hiatus to the caudal canal. The patient was conscious during the procedure to enable monitoring of neurologic symptoms. The patient was positioned prone and the lumbosacral region was prepared in a typical sterile fashion. A 14-gauge Tuohy needle was inserted into the sacral hiatus. Two 8-contact lead wires were advanced in a cephalad direction into the vertebral canal crossing the sacral nerve roots and the coccygeal nerves bilaterally (Fig. 1). Placement was confirmed through continuous fluoroscopy with intermittent lateral imaging. The Tuohy needle was removed, and the leads were secured with Steristrips (3M, Saint Paul, MN) and Tegaderm (3M, Saint Paul, MN). Electrode programming ensured that adequate paresthesia of the affected coccygeal region was obtained.

The patient achieved > 95% pain relief during her 6-day SCS trial period. Therefore, we proceeded with permanent SCS implantation using the Medtronic IntellisTM with Adaptivestim™ implantable neurostimulator, model 97715 (Minneapolis, MN).

SCS Permanent Implant

Permanent implantation of the stimulator was achieved using a retrograde approach with caudal advancement through the epidural space into the sacral canal. A 17-gauge Tuohy needle was inserted to access the epidural space of L4/L5. An 8-contact lead was advanced caudally into the sacral canal to the level of S2-24. This process was repeated on the right side (Fig. 2). The permanent SCS generator was then subcutaneously implanted into the area of the right buttock and connected to the leads. Following the procedure, the stimulator electrode settings were programmed, and the patient was instructed on the use of her hand-held programmer. The patient was satisfied with the following SCS parameters: rate 50 Hz, pulse width range: 300-350 μs.

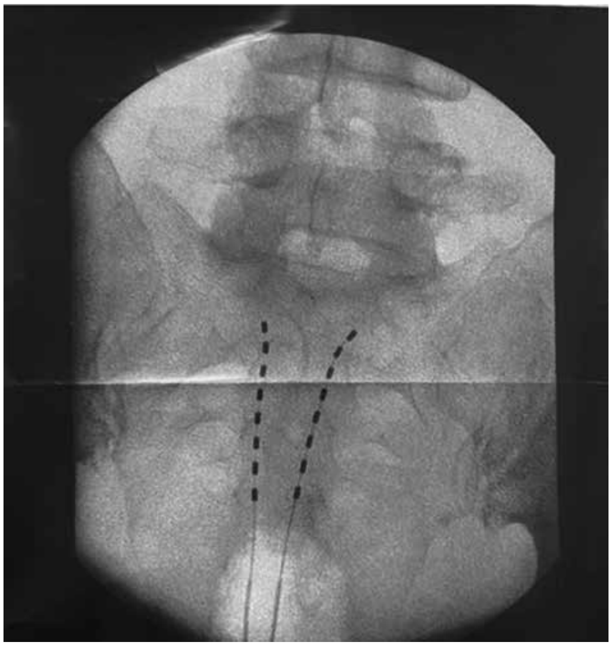

Fig. 1. Trial placement of spinal cord stimulator leads.

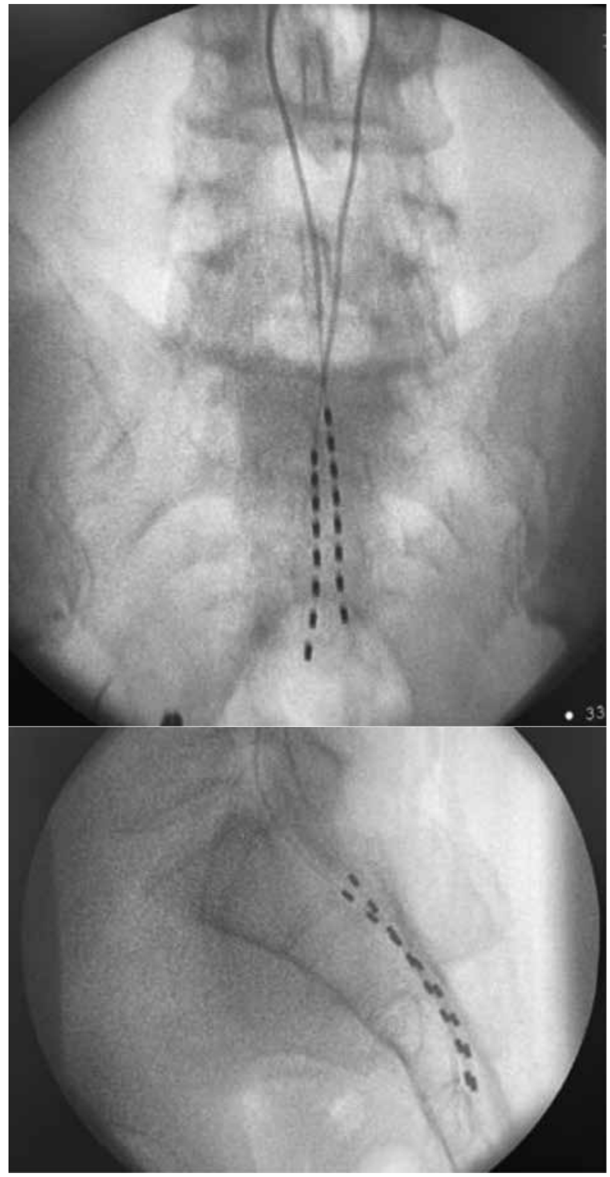

Fig 2. Different views of spinal cord stimulator lead placement, which were permanently connected to a spinal cord stimulator generator into the area of the right buttock..

RESULTS

At the one-week follow-up, the patient reported > 95% pain relief. At the 3-month follow-up, she reported continued significant improvement in her pain symptoms. She noted that the use of SCS had resulted in > 90% improvement of her coccygeal pain and her self-reported quality of life had dramatically improved.

DISCUSSION

The purpose of this case report is to describe the successful use of the sacral region as a target for retrograde placement of SCS leads in the management of intractable coccygodynia. Coccygodynia has proven to be a difficult condition to manage and reports of chronic, treatment-resistant types are abundant in the literature (1-8). There is currently no consensus for optimal management and related studies have failed to report consistent findings (1). Because of its unknown pathophysiology and multifactorial etiology, it is not likely that one therapy will be successful in all patients.

Our patient did not find long-term pain relief from other well-documented interventional therapies such as caudal epidural blocks or ganglion impar blocks (1,2,11). In the present case, we explored SCS as a minimally invasive option for treatment. SCS is an accepted neurosurgical therapy for intractable neuropathic pain. Its indications range widely, including failed back syndrome, refractory angina, and complex regional pain syndrome (12). There is limited published data on the use of SCS in the treatment of coccyx pain. One report in 2018 by Simopoulos et al (13) described the successful use of high-frequency 10-kHz SCS at the level of T9/T10 to treat refractory coccygodynia post coccygectomy. Our approach differs from the above study in that we utilized a retrograde approach to implant bilateral SCS 8-contact leads to stimulate the sacral nerves (S2-S4).

The exact mechanism of SCS is unknown; however, it is thought to be based on the "gate theory." This theory hypothesizes that the gate is opened by excessive small nerve fiber activity in the peripheral nervous system and closed by the stimulation of large afferent fibers (14). This therapy therefore involves electrical stimulation of primarily superficial large fibers in the dorsal columns to cause paresthesia in the painful area. Long-term pain relief may be mediated by the reduction of excitatory amino acids causing antidromic modulation of the gamma-aminobutyric acid and adenosine-dependent systems (14,15).

It is important to note the anatomy of the sacral canal and the correlation between the sacral and coccygeal nerves. This canal is a continuation of the lumbar spinal canal that extends inferiorly until terminating at the sacral hiatus (16,17). The sacral canal contains the caudal termination of the dural sac, the filum terminale, and the roots of the sacral and coccygeal spinal nerves (16,17). The S1-S4 anterior and posterior rami exit the canal through their associated foramina while the S5 and coccygeal nerve roots exit via the sacral hiatus (16,17). The sacral nerves carry afferent and efferent fibers. The anterior and posterior rami of S1-S4 contribute to the sacral plexus, which supplies fibers to the gluteal nerves, sciatic nerve, posterior cutaneous nerves, and the pudendal nerve (16,17). The posterior rami of the S5 and coccygeal nerves supply sensory innervation to the skin overlying the coccyx, and their anterior rami are joined by a branch off the anterior ramus of S4 to make the coccygeal plexus (16,17). The anococcygeal nerves originate from this plexus and function to supply the skin adjacent to the sacrotuberous ligament on the dorsal aspect of the coccyx (16,17). The coccygeal plexus has been theorized to play a role in generating the pain seen in coccygodynia, therefore making it an ideal site for neuromodulation (18).

Electrical stimulation of sacral nerves is a promising therapeutic option for those suffering from refractory coccygodynia (19). Sacral neural stimulation can be achieved through the following percutaneous methods: retrograde lumbar approach, anterograde sacral hiatus approach, and transforaminal approach (20). The anterograde approach was used for the trial in order to minimize the risk of accidental dura puncture associated with the retrograde approach. Using the antegrade approach for the trial seems to be a reasonable alternative with minimal side effects. However, for permanent implantation, a retrograde approach is recommended due to the reduced risk of wound infection (20,21). When compared, the retrograde and anterograde approach have shown no significant difference in the treatment of chronic pelvic pain (CPP) (19).

Based on evidence, the sacral portion of the spinal cord is the ideal site for neuromodulation in the treatment of CPP caused by different etiologies (19,22,23). CPP encompasses a number of treatment-resistant conditions including irritable bowel syndrome, interstitial cystitis, pudendal neuralgia, and coccygodynia (24). Different methods exist to stimulate the sacral spinal nerves. Sacral neuromodulation (SNM) is one approach that has conventionally been used to manage overactive bladder, nonobstructive urinary retention, and fecal incontinence (25). SNM and SCS have shown success in the treatment of CPP (19,22,23,26-30). Zabihi et al (29) reported positive results from SNM via a retrograde approach over S2-S4 for the treatment of refractory CPP, interstitial cystitis, and painful bladder pain syndrome. Additionally, Abd-Elsayed et al (30) reported the successful treatment of CPP via retrograde placement of SCS leads over S3 bilaterally. Furthermore, a 2019 meta-analysis found that patients with CPP treated with SNM had significant improvement in pain symptoms (19). These studies demonstrate that sacral nerve stimulation may be an effective treatment option for those with pelvic pain. It is important to note, however, that these studies focused on the treatment of CPP and not chronic coccygodynia, as in our study.

To our knowledge, there are no reports of SCS achieved via retrograde lead placement over sacral spinal nerves for the treatment of intractable coccygodynia. There is some evidence of SCS of the sacral region being effective via different methods. Lee et al (31) described the use of anterograde placement of SCS leads to achieve sacral burst neuromodulation for the successful treatment of intractable coccygodynia. Although this approach differs from ours, it further highlights the potential use of SCS for coccygeal pain.

CONCLUSION

Past case reports support the use of different neuromodulation techniques to manage refractory coccygodynia. Our report adds to this by demonstrating that sacral nerve stimulation via retrograde placement of

SCS leads can be used to successfully treat intractable coccygodynia. This may be considered an effective treatment option when all other conventional methods have failed.

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Author Contribution: HWH: writing, research; MAE: editing, research; PBH: writing; ADK: editing.

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