12-2012

Coccygeal fracture pain cured by sacral neuromodulation: A case report

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Recommended Citation
Hope, E.R., Gruber, D.D. (2012). Coccygeal fracture pain caused by sacral neuromodulation: A case report. Neuromodulation: Technology at the Neural Interface, 16(6), 614-617.

Received: March 8, 2012 Revised: July 8, 2012 Accepted: September 3, 2012

(onlinelibrary.wiley.com) DOI: 10.1111/ner.12002

LETTER TO THE EDITOR

Coccygeal Fracture Pain Cured by Sacral Neuromodulation: A Case Report

To the Editor:

There are many painful syndromes that are often difficult to treat. One option is electrotherapy, also known as neurostimulation. The Gate Control Theory first described by Melzack and Wall (1) in 1965 postulated that nerves carrying painful peripheral stimuli and nerves carrying touch and vibratory sensation both terminate in the dorsal horn, which acts as the gate of the spinal cord. They hypothesized that input to these nerves could "close the gate" to the painful stimuli, thereby eliminating pain.

Based on the Gate Control Theory, Shealy et al. (2) in 1967 showed success in treatment of chronic pain using the first spinal cord stimulator. Continuing this work, Shimoji et al. (3) in 1993 created epidural spinal cord stimulation (SCS) that displayed analgesic properties.

SCS use has been widespread, and is Food and Drug Administration (FDA) approved for numerous conditions. In addition to cervical, thoracic, and lumber stimulators, there is also sacral neuromodulation.

Sacral neuromodulation (InterStim®, Medtronic, Minneapolis, MN) has been indicated by the FDA for the treatment of urinary urgency, urinary frequency, and fecal incontinence. It is the only approved sacral neuromodulator. However, very little data exist on its use for chronic pain or lower back pain. We describe a case where InterStim was used to treat urinary urgency/frequency symptoms and incidentally relieved chronic lower back pain from a previous pelvic fracture.

CASE REPORT

A 74-year-old G2P2 postmenopausal female presented with long-standing urge-dominant mixed urinary incontinence who previously failed two anticholinergic medications, tolterodine and darifenacin. She also underwent one year of pelvic floor physical therapy with little improvement. She received an InterStim Peripheral Nerve Stimulation Phase I with incision and implantation of bilateral tine quadripolar lead electrodes into the S3 foraminae with fluoroscopic guidance under conscious sedation with local anesthesia in December 2008. The placement was successful by patient identification of sensation, great toe flexion, and bellowing of the anus noted. She experienced a suboptimal urgency response (defined as <50% reduction in symptoms), but proceeded to InterStim Phase II with incision and subcutaneous implantation of sacral nerve neurostimulator with electronic analysis and complex programming with the left lead as that side had a great reduction in symptoms. She elected to continue with the InterStim implantation because it significantly improved her chronic coccygeal pain that resulted from a motor vehicle-induced pelvic fracture, 25 years prior. She was so used to this debilitating pain, which sometimes required her to have to "crawl to the bathroom at night," that she failed to inform us of this medical condition prior to implantation. Her daily pain rating went from an 8–10/10 to 2–4/10 on the visual analog scale. A sacral x-ray confirmed correct placement of the lead at the S3 level on that patient's left (Figs 1 and 2).

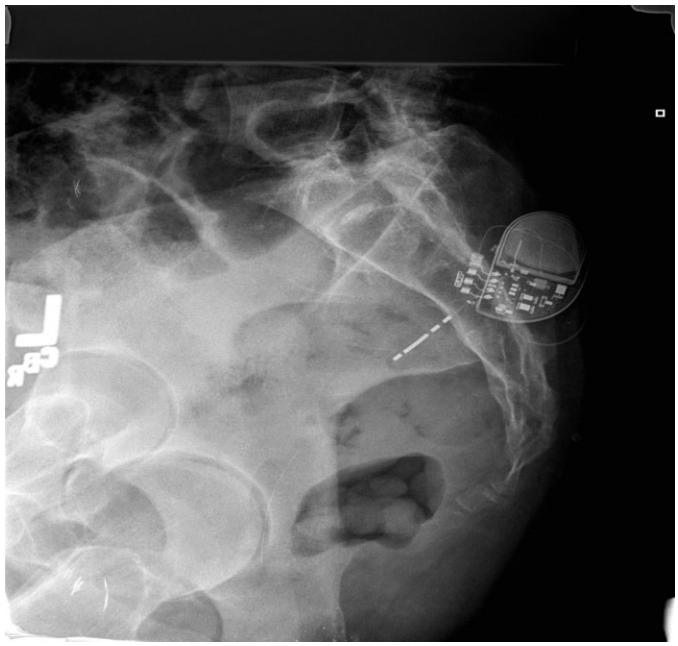

Figure 1. Lateral sacral x-ray showing correct InterStim lead placement at S3 level.

She continued to have daily episodes of leakage of urine and began tolterodine again to help with urge symptoms, but greatly desired the InterStim kept on because of considerable coccygeal pain relief. The medication was subsequently discontinued as it did not reduce her urge symptoms.

The patient subsequently lost the remote device for the InterStim two months after placement and it was turned off for several months. She noted prompt return of her chronic coccygeal pain

The authors have no disclosures or conflicts of interest.
The authors listed each had a substantial contribution to this case report.
The views expressed in this article are those of the authors and do not reflect the official policy of the Department of the Army, Department of Defense, or the U.S. Government.

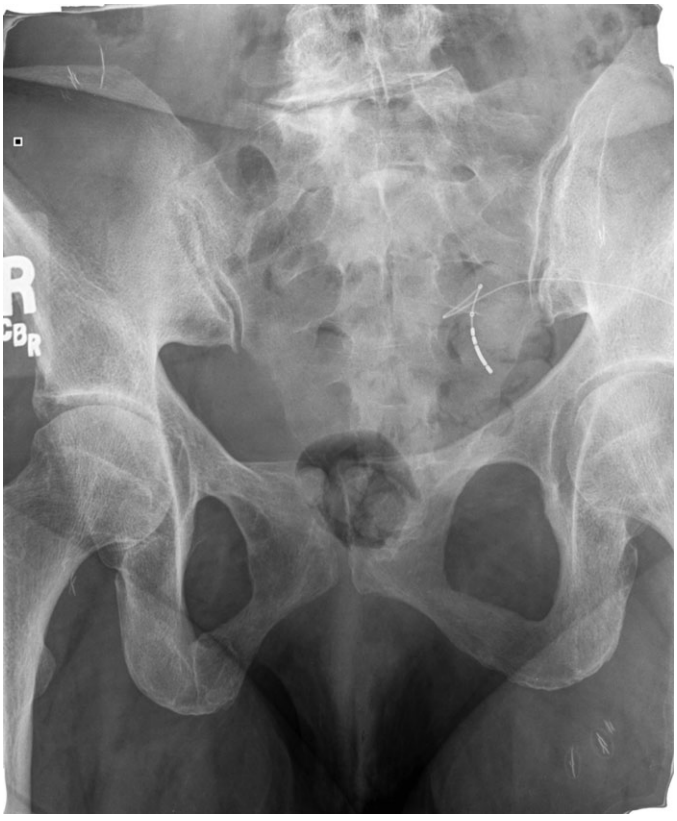

Figure 2. Anterior-posterior sacral x-ray confirming lead placement at patient's S3 level, left side.

after losing the remote. After presenting to the office and obtaining a new remote with the device turned on, the patient stated that she again felt immediate relief from her pain.

She underwent collagen injections in November 2009 and January 2010 (one year postimplant) for stress urinary incontinence symptoms, with significant improvement in stress and urge urinary incontinence symptoms for one week and then worsening urge symptoms daily thereafter. The patient restarted pelvic floor physical therapy and at the time stated that InterStim helps alleviate urge symptoms at night, but does not reduce daytime symptoms. However, she still wants the InterStim kept on due to the substantial relief of her chronic coccygeal pain from the previous pelvic fracture.

She subsequently had Botulinum-A Toxin (Botulinum-A Toxin (Botox®, Allergen Pharmaceuticals, Irvine, CA, USA) bladder injections for worsening urge symptoms in March 2011 (2.5 years postimplant). As of May 2012 (3.5 years postimplant), she reports that her urge symptoms are significantly improved and her coccygeal pain is still controlled with the continued use of the InsterStim.

DISCUSSION

SCS has incomplete coverage for many areas of the body, including, but not limited to, low back, buttocks, feet, groin, pelvis, and neck. The S2-5 dermatomes are supplied by pathways that are somatotopically deep in the spinal cord and are often times difficult to reach with SCS (4).

Peripheral nerve stimulation works by activating inhibitory interneurons as well as primary afferent delta fibers, and has the potential to recruit more nerve fibers than SCS. SCS has to penetrate through multiple dural layers as well as cerebrospinal fluid, and the primary afferent delta fibers are not accessible (5).

Chronic pain disorders are more common among women than men. Women also experience increased frequency of pain as well as higher disability rates and greater allocation of healthcare resources (5). Chronic pain in the sacral area has traditionally been difficult to control, and the unique anatomy in this region may contribute. Many chronic pelvic pain conditions are suspected to be neuropathic in nature, including coccygodynia; however, this has not been proven (5). This is further supported by the fact that there are extremely limited medical and/or surgical interventions that alleviate pain in a majority of patients (6). Patients with pelvic pain typically respond best to bilateral S2 or S3 electrodes as opposed to unilateral (4).

Alo et al. in 1999 (7) (Table 1) used selective nerve root catheters in the epidural space for five chronic pain patients who failed several types of conservative treatment and had at least one abnormal objective finding. The chronic pain syndromes involved ilioinguinal neuralgia, discogenic pain at L4-5, failed back syndrome, vulvodynia, and interstitial cystitis. Multielectrode systems were placed in the thoracic, lumbar, and/or sacral areas. All patients had >50% reduction in pain level, with their visual analogs declining significantly after the seven-day trial and all requested permanent implantation of a nerve root stimulation system. They also postulated that sacral electrodes appeared less vulnerable anatomically to patient movement than electrodes at other levels, possibly because they are contained within the root sleeve in the spinal canal.

Again in 1999, Alo et al. (8) reported a cohort of 80 patients with 36-month follow-up after lead placement, primarily in thoracic and cervical regions with a majority diagnosed with low back and extremity pain. They were initially given a single program after multiple electrodes were placed, then given access for multiple program stimulation with patient control. This allowed patients to control their level of success and alleviate the all-or-nothing phenomenon. Eighty percent of patients would have the procedure again and 72% were still using the SCS system daily after 36 months.

In 2001, Alo and Mckay (9) published a case report of a 32-year-old woman with interstitial cystitis with intractable 10/10 pain that severely disrupted her quality of life. Two sacral nerve root stimulator electrodes were placed at the S2-3 level bilaterally. During her seven-day trial phase, she had increased bladder volumes, decreased pain, and stopped all bladder pain and spasm medications. Her pain level was significantly reduced at 1/10 by the visual analog scale. She subsequently underwent permanent implantation at the same level. At publication, patient was one year after implantation, only requiring occasional use of the stimulator and has rare symptoms, with her preinterstitial cystitis quality of life restored. A literature search for coccygeal fracture or neuromodulation revealed one case report of a female that had previously undergone InterStim implantation for urinary incontinence and also had bilateral lower extremity pain and back pain from postlaminectomy syndrome (10). In this case, however, the InterStim did not improve her pain, but instead they used SCS at levels T8-T9-T10 which helped both her urinary incontinence and relieved her pain completely. In addition, they tried the InterStim vs. the SCS (turning one off and then the other for two-month period) and they were equal in terms of urinary continence symptom relief, but only the SCS improved both her lower extremity and back pain.

Table 1. Literature Search Description and Outcomes.

Author	Year	Description	Outcome
Alo et al. (7)	1999	Selective nerve root catheters for chronic pain in thoracic, lumbar, and sacral areas	>50% reduction in VAS pain level; all had permanent implantation
Alo et al. (8)	1999	80 patients with thoracic and cervical leads for low back and extremity pain; single vs. multiple program controls	36-month follow-up, with 72% of patients still using system daily
Alo and Mckay (9)	2001	Intractable interstitial cystitis pain with sacral neuromodulator placed	Significantly improved quality of life; rare symptoms with occasional use of stimulator
Marinkovic et al. (12)	2011	Retrospective, case control to evaluate sacral neuromodulation in interstitial cystitis patients	Statistically significant ($p < 0.01$) improvement in urinary frequency and visual analog pain scale scores at 6+ year follow-up
Pauls et al. (11)	2004	Refractory interstitial cystitis with chronic pelvic pain patients treated with sacral neuromodulation	Decreased narcotic requirements and subjective pelvic pain
Peters et al. (14)	2003	Refractory interstitial cystitis treated with sacral neuromodulation via percutaneous approach or staged procedure	Symptom and quality of life improvement; improved implantation rate with staged procedure (94% vs. 52%)
Peters et al. (13)	2004	Refractory interstitial cystitis treated with sacral neuromodulation	69% patients with moderate or marked improvement in pelvic pain
Yakovlev and Resch (10)	2010	InterStim implantation and SCS for urinary incontinence and postlaminectomy syndrome	InterStim and SCS equal in incontinence relief; SCS improved lower extremity and back pain also
Yakovlev and Resch (15)	2012	Spinal cord stimulation after coccygeal fracture with coccygodynia	T8-T10 leads with excellent pain relief; no longer requiring daily pain medication

SCS, spinal cord stimulation; VAS, visual analog scale.

Several papers referred to postoperative pain in terms of pain after placement and did not refer to preoperative pain that was relieved with InterStim. Pauls et al. reported a prospective study that evaluated sexual function after InterStim implantation for urinary incontinence; however, patients did not have a history of chronic pelvic pain (11). There was a significant improvement in pain during intercourse ($p = 0.015$).

Marinkovic et al. reported patients with interstitial cystitis and painful bladder syndrome that had a significant improvement in pain after sacral neuromodulation ($p < 0.01$) (12). Another paper on interstitial cystitis by Peters et al. describes a moderate or marked improvement in pain after InterStim implantation in treating chronic pain associated with interstitial cystitis refractory to standard therapy (13). Finally, another paper by Peters et al. found that 69% of their patients reported moderate or marked improvement in pelvic pain also with interstitial cystitis (14).

Only one case report was found that discussed SCS for treatment of coccygodynia after a coccygeal fracture, by Yakovlev and Resch (15). A 37-year-old male with a two-year history of coccygodynia after a closed sacral and coccygeal fracture as well as a rectal tear. He received surgery for the perineal wound, but continued to experience pain after the fractures had healed. Two epidural leads were placed with a satisfactory trial response. The final leads were placed at T8, T9, and T10, with excellent pain relief reported. The pain no longer required pain medications and his quality of life improved.

Our patient underwent a lateral sacral x-ray which confirmed correct lead placement at the S3 foramina; however, the pudendal and coccygeal nerve roots are closely related. The S3 nerve root contributes to the pudendal nerve. S4 and S5 nerve roots supply the coccyx, anus, and rectal areas. It is possible that our patient has variant anatomy and that the S3 nerve root supplies a portion of the coccygeal area, which would account for her pain relief associated with the InterStim device.

The majority of pain that the sacral neuromodulation has previously treated has been chronic pelvic pain that is refractory to other therapies, which often coexists with urinary incontinence or refractory interstitial cystitis. For these two indications, it appears that the sacral neuromodulation has a significant improvement in pain. No citations were found that described the use of sacral neuromodulation in terms of coccygeal pain; only SCS has previously been used. In conclusion, sacral neuromodulation has the potential for treatment of coccygeal pain.

Authorship Statement

Dr. Gruber had the idea for the case report and took care of the patient in the clinic setting. Dr. Hope prepared the case report draft with important intellectual input from Dr. Gruber. All authors approved the final case report/letter.

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Keywords: coccygeal fracture, coccygeal pain, InterStim, neuromodulation

