Case Report

Sacral Nerve Stimulation as a Treatment Modality for Intractable Neuropathic Testicular Pain

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Free full manuscript: www.painphysicianjournal.com **Background:** Chronic testicular pain, or "chronic orchalgia," is defined as testicular pain 3 months or longer in duration that significantly interferes with the daily activities of the patient. For patients failing to respond to conservative treatment, microsurgical denervation of the spermatic cord, epididymectomy, and vasovasostomy have all shown a degree of relief. However, these are all invasive procedures and no treatment has proven efficacy when these options fail.

We present a case of a male who presented with over a decade of chronic right-sided testicular pain secondary to recurrent epididymitis. Before arriving at our clinic the patient had an epididymectomy performed with no appreciable improvement in pain. Initially ilioinguinal, iliohypogastric, and genetofemoral nerve blocks; right-sided S1, S2, and S3 transforaminal epidural steroid injections (TFESIs) with inferior hypogastric blocks; and right-sided T12-L1, L1-L2, and L2-L3 TFESIs all failed to provide pain relief. After conservative therapies had failed, a sacral nerve stimulation trial was done via a caudal epidural approach. The permanent implant has provided the patient with sustained 80% decrease in pain at 4 months status post permanent sacral nerve stimulation implant.

The above case demonstrates the potential benefit of sacral nerve stimulation with neuropathic intractable testicular pain in a patient that failed conservative treatment. In this case, the patient had exhausted medical and surgical management, including advanced interventional pain options. We were unable to find any previous published cases of neurostimulation used as a modality of treatment for testicular pain, and further studies are needed to gain a better understanding of the efficacy in this setting.

Key words: Sacral nerve stimulation, spinal cord stimulation, neuromodulation, testicular pain, orchalgia, intractable, chronic

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he patient is a 31-year-old male with a past medical history significant for hypospadias, chronic urethral strictures, recurrent pilonidal cysts, chronic epididymitis, anxiety, and depression that presented with over a decade of chronic rightsided testicular pain. Around the age of 6 months the patient had construction of a neo-urethra graft performed using penile shaft skin. He then began to develop urinary tract infections in his early teenage

years with increasing frequency to the point where he was being treated every 2 weeks with antibiotics in addition to the prophylactic antibiotics.

As a freshman in college, he woke up with sudden onset right testicular edema, erythema, pustulant urethral discharge, and severe pain. He was diagnosed with epididymitis and treated with NSAIDs, antibiotics, and pain medication, but continued to have recurrent episodes of epididymitis. A second urologist discovered that the neo-urethra graft contained hair follicles necessitating a urethral graft revision. Although the patient did not experience a recurrence of the epididymitis after the revision, his pain progressively worsened to the extent where the patient was making 2 to 3 visits per month to the emergency room. Due to the patient's chronic pain and frequent absences from work, he lost his job and became disabled. After losing his job, the patient became severely depressed and attempted suicide. He was admitted to an inpatient psychiatry ward for 3 days and was discharged in stable condition.

Before arriving to our clinic the patient had been treated with various medications for the epididymitis, and recently had an epididymectomy performed with no appreciable improvement in pain. On presentation the pain was described as sharp, shooting pain on the anterior aspect of his right testicle covering about a 3 cm longitudinal area that was at a constant level of 8/10. The pain reached a level of 10/10 when exacerbated by rising from a seated position, coughing, lifting objects, and with sexual activity. The pain was relieved while lying supine. The patient was on MS IR 15mg QID and Percocet 7.5/325 4-6/day prn, as well as Effexor and Ativan for co-morbid depression and anxiety. He had also tried multiple membrane stabilizing pain medications with little relief. Despite aggressive pharmacotherapy he continued to suffer from chronic intractable pain, which, over the past 5 months had become increasingly more intense and problematic.

Initially ilioinguinal, iliohypogastric, and genetofemoral nerve blocks were performed, but did not provide relief. He then had right-sided S1, S2, and S3 transforaminal epidural steroid injections (TFESIs) with inferior hypogastric blocks, which also failed to provide significant relief. Finally, he had right-sided T12-L1, L1-L2, and L2-L3 TFESIs which also did not provide pain control.

After all other therapies had failed, a sacral nerve stimulation trial was discussed with the patient and approved SNS through his insurance. The trial was done via a caudal epidural approach. Two 14-gauge Touhy needles were placed into the sacral hiatus with AP and lateral fluoroscopic views. An 8-contact St. Jude Medical (SJM, headquarters in St. Paul, MN) electrode was placed through each needle. The first lead was placed just medial to the right-sided S1, S2, and S3 sacral foramen and a second lead was placed just lateral to the first lead for stability and electrical cross-talk (Figs. 1 and 2). The leads were at the upper and middle third of the sacrum at the lateral aspect of the canal. The patient received excellent coverage of



Fig. 1. AP view: Two leads coming through the sacral hiatus towards the right side. First lead is directly medial to the S1, S2, and S3 sacral foramen. Second lead is directly lateral to the first lead.



Fig. 2. Lateral view: Two leads in the sacral epidural space. Covering S1, S2, and S3. Leads are inferior to L5-S1 disc space. Top 2 leads were used for stimulation during the trial to cover the patients right-sided testicular pain.

his right-sided testicular pain at the S1 neuroforaminal level (top lead of each electrode on the trial). Just inferior to this at the S2 level, the patient had rectal paresthesias. At follow-up 5 days later, the patient reported an increase is his daily activities and an 80% reduction in his testicular pain.

Permanent implant was scheduled with a skilled neurosurgeon in the area. The surgeon made a midline incision from the top of the sacrum to the middle third. Paraspinous muscles were dissected to expose the sacrum. The dorsal elements of the sacrum were unroofed with a drill. A small footprint 8-contact SJM Exclaim® Tripole Paddle lead was surgically inserted to match the trial electrode position. The electrode was anchored to the bone with a silk suture through a pre-drilled hole in the bone. A strain relief loop was left in the sacrum and the leads were tunneled to the battery pocket above the left buttock area. The leads were connected to a SJM Eon-Mini battery and tested. The wounds were then irrigated and closed in anatomical layers.

The permanent implant has provided the patient with sustained 80% decrease in pain at 4 months status post permanent spinal cord stimulation implant. He is using 2 higher frequency programs (90 Hz/387 PW/4.5-9 mA/466 Ohms and 110 Hz/387 PW/4.5-9 mA/466 Ohms) and one lower frequency program (10 Hz/412 PW/3.5-8.5 mA/466 Ohms). Coverage of his painful area continues to be just medial to the S1 foramen. The patient says, "I am amazed at how well the stimulator is working and have regained my life." The patient has had dramatic improvements; he is working full-time, has had no emergency room visits, is less depressed, and is currently being weaned off of opioid medications with a current regiment of Percocet 7.5/325 QID prn. He currently ranks his average pain as 2-4/10 (compared to 8/10 previously), his least pain is 0-2/10 (compared to 4/10 previously), and his worst pain at 7/10 (compared to 10/10 previously).

DISCUSSION

The above case demonstrates the potential therapeutic benefit of SNS for neuropathic intractable testicular pain in a patient that failed conservative treatment. In this case, the patient had exhausted medical and surgical management, including advanced interventional pain options. Experiencing severe, chronic testicular pain for over a decade, the patient had become hopeless. After the epididymectomy and multiple nerve blocks failed to give significant relief, a SNS trial provided a reasonable treatment choice.

Although our patient's initial problem was chronic epididymitis, our patient ultimately suffered from associated chronic neuropathic testicular pain. Chronic testicular pain, or "chronic orchalgia," is defined as "intermittent or constant testicular pain 3 months or longer in duration that significantly interferes with the daily activities of the patient so as to prompt him to seek medical attention" (1). For patients failing to respond to conservative treatment, microsurgical denervation of the spermatic cord, epididymectomy, and vasovasostomy have all shown a degree of relief (2). Unfortunately, for the group of patients that fail to respond to both conservative and more invasive treatment methods, the only traditionally available therapeutic option is inguinal orchiectomy (2). A recent study suggested that pulsed radiofrequency of the spermatic cord may be effective in the treatment of chronic testicular pain (3). Pulsed radiofrequency was first performed for chronic back pain and sciatica by Dr. Shealy in 1975 (4). It also appears that SCS has some therapeutic potential for chronic intractable visceral pelvic pain (5), and when SCS is not effective SNS may play a role in treatment of chronic pelvic pain (6). Tanagho and Schmidt were the first to perform SNS with initial indications for urinary urge incontinence, urgency-frequency, and urinary retention in 1981 (7). However, we were unable to find any previous published cases of SNS used as a modality of treatment for testicular pain in humans in a thorough PubMed search. We are optimistic that we may provide an alternative treatment to orchiectomy, and possibly an option to use before attempting any invasive microsurgery.

HISTORY

Let us briefly review the relevant neuroanatomy. Textbooks have traditionally taught that the pudendal nerve is derived from the S2, S3, and S4 nerve roots. However, an evidence-based review article reports that dermatomes are much larger than are currently described in textbooks (8). These authors explain that the reason for such great overlap and individual variability is due largely to that fact that 2 or more spinal roots innervate most areas of skin, and also secondary to the presence of intrathecal intersegmental anastomoses between dorsal spinal rootlets (8). Furthermore, a surgical anatomy study found that 25% of the cadavers had pudendal nerves with contributions from S1 and 5% had contributions from S5 in addition to being comprised of roots from the 2nd, 3rd, and 4th anterior sacral rami (9). This lends a rational explanation to the therapeutic effects we witnessed for testicular pain with the lead positioned lateral to the S1 neuroforamen.

Conclusion

There are 3 basic approaches to a spinal cord stimulation trial for SNS. The first to be developed was a retrograde technique, which allowed selective stimulation of the S2, S3, and S4 nerve roots (10). The retrograde nerve root stimulation is performed by entering the lumbar epidural space and advancing the leads inferiorly to the desired location medial to the foramen. The second is a transforaminal sacral placement, which is commonly done for overactive bladder and urinary retention with the Medtronic InterStim® device through the S3 foramen (11-13). The final is an anterograde technique, which approaches from the caudal epidural space, similar to that used for a caudal Racz procedure, known as epidural lysis of adhesions (14). Our approach was to enter the caudal epidural space with the SJM electrodes and advance the leads to the right of the midline just medial to the S1, S2, and S3 sacral foramen openings. The location was verified by both AP and lateral fluoroscopic views.

Many theories on the mechanism of action of SCS have been suggested including the original theory by Melzack and Wall (15) of activation of gate control mechanisms at the dorsal horn nucleus. More recently, additional studies have indicated neurotransmitters, such as GABA and adenosine, may be involved in the pathway through which SCS works (16,17). Regardless of which theory is correct, SCS has been proven to have efficacy in the treatment of complex regional pain syndromes (18,19), intractable pain due to peripheral vascular disease (20-22), intractable pain due to angina (23,24), and failed back surgery syndrome (25-27). Long-term studies have indicated that SCS has a diminishing effectiveness over time for patients with complex regional pain syndromes (28), and may not be cost effective for management of critical limb ischemia (29). In addition to bleeding, infection, and nerve damage, a significant risk includes lead migration, which may require surgical revision. However, with no other alternative therapeutic options presently available for chronic testicular pain, SNS offers many desirable benefits, which include being less invasive, reversible, adjustable, and testable. SCS may be a promising modality for the treatment of neuropathic testicular pain.

Beyond the scope of this case report, neurostimulation may be considered in other painful conditions such as chronic testicular pain status post orchiectomy, penile pain, vulvar pain, vaginal pain, pelvic pain, interstitial cystitis, and other chronic pelvic pain conditions. Ultimately, a prospective study is needed to gain a better understanding of the efficacy of SNS as a treatment modality for intractable testicular pain for those who have failed conservative therapies.

References

- Davis BE, Noble KJ, Weigel JW. Analysis and management of chronic testicular pain. *J Uro* 1990; 143:936-939.
- 2. Granitsiotis P, Kirk D. Chronic testicular pain: An overview. *Eur Urol* 2004; 45:430-436.
- Mirsa S, Ward S, Coker C. Pulsed radiofrequency for chronic testicular pain-A preliminary report. *Pain Med* 2009; 10:673-678.
- Shealy CN. Percutaneous radiofrequency denervation of spinal facets. Treatment for chronic back pain and sciatica. *J Neurosurg* 1975; 43:448-451.
- Kapural L, Narouze SN, Janicki TI, Mekhail N. Spinal cord stimulation is an effective treatment for the chronic in-

tractable visceral pelvic pain. *Pain Med* 2006; 7:440-443.

- Haque R, Winfree CJ. Spinal nerve root stimulation. *Neurosurg Focus* 2006; 21: E4.
- Pettit PD, Thompson JR, Chen AH. Sacral neuromodulation: New applications in the treatment of female pelvic floor 11. dysfunction. *Curr Opin Obstet Gynecol* 2002; 14:521-525.
- Lee MW, McPhee RW, Stringer MD. An evidence-based approach to human dermatomes. *Clin Anat* 2008; 21:363- 12. 373.
- Shafik A, el-Sherif M, Youssef A, Olfat ES. Surgical anatomy of the pudendal nerve and its clinical implications. *Clin*

Anat 1995; 8:110-115.

- Alo KM, Yland MJ, Redko V, Feler C, Naumann C. Lumbar and sacral nerve root stimulation (NRS) in the treatment of chronic pain: A novel anatomic approach and neuro stimulation technique. *Neuromodulation* 1999;2:23-31.
 - Schmidt RA, Jonas U, Oleson KA, Janknegt R, Hassouna M, Siegel S, Van Kerrebroeck P. Sacral nerve stimulation for treatment of refractory urinary urge incontinence. *J Urol* 1999; 162:352-357.
 - Siegel SW, Catanzaro F, Dijkema HE, Elhilali MM, Fowler CJ, Gajewski JB, Hassouna MM, Janknegt RA, Jonas U, van Kerrebroeck PE, Lycklama a Nijeholt AA, Oleson KA, Schmidt RA. Long-term re-

sults of a multicenter study on sacral nerve stimulation for treatment of urinary urge incontinence, urgency-frequency, and retention. *Urology* 2000; 56:87-91.

- Jonas U, Fowler CJ, Chancellor MB, Elhilal MM, Fall M Gajewski JB, Grunewald V, Hassouna MM, Hombergh UVD, Janknegt R, Van Kerrebroeck PEV, Lylcklama A, Nijeholt AAB, Siegel SW, Schmidt RA. Efficacy of sacral nerve stimulation for urinary retention: Results 18 months after implantation. J Urol 2001; 165:15-19.
- Trescot AM, Chopra P, Abdi S, Datta S, Schultz DM. Systematic review of effectiveness and complications of adhesiolysis in the management of chronic spinal pain: An update. *Pain Physician* 2007; 10:129-146.
- Melzack RA, Wall PD. Pain mechanisms: A new theory. *Science* 1965; 150:971-979.
- Linderoth B, Gazelius B, Franck J, Brodin E. Dorsal column stimulation induces release of serotonin and substance P in the cat dorsal horn. *Neurosurgery* 1992; 31:289-297.
- 17. Meyerson BA, Ren B, Herregodts, P, Linderoth B. Spinal cord stimulation in animal models of mononeuropathy: Effects on the withdrawal response and the flexor reflex. *Pain* 1999; 79:229-243.
- Harke H, Gretenkort P, Ladleif HU, Rahman S. Spinal cord stimulation in sym- 24.

pathetically maintained complex regional pain syndrome type I with severe disability. A prospective clinical study. *Eur J Pain* 2005; 9:363-373.

- Kemler MA, Barendse GA, van Kleef M, de Vet HC, Rijks CP, Furnée CA, van den Wildenberg FA. Spinal cord stimulation in patients with chronic reflex sympathetic dystrophy. N Engl J Med 2000; 343:618-624.
- 20. Jivegard LE, Augustinsson LE, Holm J, Risberg B, Ortenwall P. Effects of spinal cord stimulation (SCS) in patients with inoperable severe lower limb ischaemia: A prospective randomised controlled study. J Vasc Endovasc Surg 1995; 9:421-425.
- 21. Klomp HM, Spincemaille GH, Steyerberg EW, Habbema JD, van Urk H. ESES Study Group. Spinal cord stimulation in critical limb ischaemia: a randomised trial. *Lancet* 1999; 353:1040-1044.
- 22. Broseta J, Barbera J, de Vera JA, Barcia-Salorio JL, Garcia-March G, González-Darder J, Rovaina F, Joanes V. Spinal cord stimulation in peripheral arterial disease. A cooperative study. *J Neurosurg* 1986; 64:71-80.
- 23. De Jongste MJL, Hautvast RWM, Hillege HL, Lie KI; Working Group on Neuroradiology. Efficacy of spinal cord stimulation as adjuvant therapy for intractable angina pectoris: A prospective, randomized clinical study. J Am Coll Cardiol 1994; 23:1592-1597.
 - 4. Hautvast RW, DeJongste MJ, Staal MJ,

van Gilst WH, Lie KI. Spinal cord stimulation in chronic intractable angina pectoris: A randomized, controlled efficacy study. *Am Heart J* 1998; 136:1114-1120.

- 25. Barolat G, Oakley JC, Law JD, North RB, Ketzik B, Sharan A. Epidural spinal cord stimulation with a multiple electrode paddle lead is effective in treating intractable low back pain. *Neuromodulation* 2001; 4:59-66.
- Kumar K, Nath R, Wyant GM. Treatment of chronic pain by epidural spinal cord stimulation: A 10-year experience. J Neurosurg 1991; 75:402-407.
- Burchiel KJ, Anderson VC, Brown FD, Fessler RG, Friedman WA, Pelofsky S, Weiner RL, Oakley J, Shatin D. Prospective, multicenter study of spinal cord stimulation for relief of chronic back and extremity pain. *Spine* 1996; 21:2786-2794.
- 28. Kemler MA, de Vet HC, Barendse GA, van den Wildenberg FA, van Kleef M. Effect of spinal cord stimulation for chronic complex regional pain syndrome Type 1: Five-year final follow-up of patients in a randomized controlled trial. *J Neurosurg* 2008; 108:292-298.
- 29. Klomp HM, Steyerberg EW, van Urk H, Habbema JD. Spinal cord stimulation is not cost-effective for non-surgical management of critical limb ischaemia. *Eur J Vasc Endovasc Surg* 2006; 31:500-508.