Sacral neuromodulation as a treatment for neuropathic clitoral pain after abdominal hysterectomy

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Abstract Sacral neuromodulation (SNM) may be beneficial in the treatment of patients with chronic pelvic pain, although it is not an FDA-approved indication. We present a case of a 51-year-old patient that presented with symptoms of lower urinary tract dysfunction and clitoral pain after an abdominal hysterectomy. Electrophysiological evaluation suggested a pudendal nerve lesion. After failure of conservative treatment, she was offered SNM as a treatment for her voiding symptoms. During test stimulation, she experienced only moderate improvement in voiding symptoms, but a striking improvement in pain symptoms. She underwent a two-stage implantation of a neurostimulator with a successful outcome after 6 months’ follow-up. The results of this report suggest that SNM may be effective in patients with neuropathic pelvic pain.

Keywords Sacral neuromodulation • Pelvic pain • Clitoral pain

Introduction

Sacral neuromodulation (SNM) is an accepted treatment for lower urinary tract dysfunction refractory to conservative treatment. It has also proven to be an effective treatment for fecal incontinence and functional bowel disorders. Although no FDA-approved indication exists, there are several reports showing a beneficial effect of SNM treatment in chronic pelvic pain conditions, such as interstitial cystitis or vulvodynia [1, 2]. The results seem promising, although the study populations are small and long-term success has not been defined yet.

Clitoral pain is an uncommon pain condition, and very little data exist about its prevalence. Gordon et al. reported 21 cases of clitoral pain syndromes, including one patient with clitoral pain after a laparoscopic hysterectomy [3]. Their study showed that a wide variety of conditions are associated with clitoral pain syndromes (trauma, surgery, neurological disease), although treatment was not discussed. We present a case of a patient with clitoral pain following hysterectomy, treated successfully with SNM therapy.

Case

A 51-year-old woman presented to our urological clinic with persistent voiding symptoms, which developed shortly after an abdominal hysterectomy done 6 months earlier. The patient reported impaired bladder emptying requiring abdominal straining, followed by a sensation of incomplete emptying. She also complained of urinary incontinence occurring during day and night, without perceiving urgency. Coughing or sneezing did not elicit urinary incontinence.

Furthermore, the patient reported clitoral pain with paresthesia and a bothersome orgasmic sensation. These complaints also developed after the hysterectomy and were continuously present. A hysterectomy with left ovariectomy was performed for complex atypical endometrial hyperplasia. Post-operatively, a hematoma of the vaginal apex developed, which was treated conservatively. In the acute stage, the patient complained of vaginal pain, with radiation
into the lower abdomen. After 2 months, the pain became more prominent in the clitoral area, and paresthesia also developed. She reported that these symptoms increased when having emotional stress. Due to her symptoms, she was unable to have sexual intercourse and to participate in social activities.

Further relevant medical history noted two vaginal deliveries and a right-sided ovariectomy due to the presence of a mature teratoma of the ovary. There was no history of back injury or surgery, vaginal infections, sexually transmitted diseases, genital trauma, or sexual abuse.

Physical examination showed normal external female genitalia and pelvic examination was normal. Clinical neurological examination was unremarkable, including normal perineal sensation for touch and normal sacral reflexes.

Urodynamic measurement revealed a bladder capacity of 340 ml. During voiding, detrusor hypocontractility and inadequate sphincter relaxation was observed, with urethral pressures up to 130 cmH₂O. Post-micturition residual volume was 321 ml, with a voided volume of 65 ml. Incontinence could not be demonstrated during the investigation.

Electrophysiological evaluation suggested the presence of a peripheral motor neuron lesion of the pudendal nerve. The bulbocavernosal reflex latency was increased to 54.8 ms (normal range 24–41 ms), and bladder–bulbocavernosus reflex latency was 83.6 ms (normal range 47–79 ms). The latency of the pudendal somatosensory-evoked potential was normal with a normal shape of the evoked response.

Psychological evaluation showed signs of mild anxiety and depression. The total score of the Hospital Anxiety and Depression Scale was 16, with scores of 10 on the anxiety subscale and 6 on the depression subscale.

The initial treatment consisted of pelvic floor physiotherapy combined with clean intermittent self-catherization (CIC). This treatment facilitated voiding, resulting in reduced residual volumes below 100 ml, after which CIC was discontinued. However, the complaints of urinary incontinence persisted, and therefore anticholinergics were added to the treatment. For the clitoral pain, amitriptylin was started but was only temporarily effective. Additional treatment with pregabalin, lamotrigin, and carbamazepin gave partial relief of pain symptoms but was discontinued because of side effects.

Due to failure of conservative treatment of voiding symptoms, sacral neuromodulation was considered a possible treatment option. The patient was screened for SNM therapy with a tined-lead test stimulation. This procedure involved placement of a quadripolar lead into the third sacral foramen in order to stimulate the S3 nerve root. During the screening period of 3 weeks, patient symptoms were self-monitored using voiding diaries. The test was considered successful if more than 50% was noted in any of the studied parameters. Additionally, pain symptoms were recorded before (baseline) and during test stimulation using the visual analogue scale (VAS).

Evaluation of the voiding diaries showed less than 50% improvement in any of the relevant voiding diary parameters (Table 1). The VAS scores however showed a significant reduction in pain symptoms (Table 2). Although the test stimulation was considered unsuccessful regarding voiding symptoms, the marked decrease in pain as well as the high patient satisfaction served as guidance to offer the patient a permanent implantable neurostimulator. After extensive counseling and informed consent that SNM is an off-label treatment for clitoral pain, she received an implant 3 weeks after the tined-lead test stimulation.

After 6 months, VAS scores still showed a significant improvement (Table 2) in pain symptoms. The patient reported to be very satisfied with the treatment and had resumed her daily activities. No improvement in bladder function was noted; bladder emptying was still difficult and incontinence persisted.

### Discussion

The cause of the clitoral pain in this patient is unknown and probably multifactorial. Repeated minor trauma of the pelvic floor and pudendal nerve may have accumulated due to the vaginal deliveries and pelvic surgery, leading to the measurable pudendal nerve lesion. SNM shows to be beneficial in improving clitoral pain and the effect persists

| Parameter                    | Baseline | Test stimulation | Improvement (%) |
|------------------------------|----------|-----------------|-----------------|
| Voided volume/void (ml)      | 217      | 263             | +36             |
| Number of voids/day          | 12.3     | 8.9             | −28             |
| Number of leakages/day       | 5.4      | 3.4             | −37             |
| Number of pads/day           | 1.2      | 1.0             | −11             |

### Table 2 Overview of the VAS pain scores filled out by the patient before and during SNM treatment

| VAS score (0–100) | Baseline | Test Stimulation | 6 months follow-up |
|-------------------|----------|------------------|--------------------|
| Pain at the moment| 75       | 15               | 0                  |
| Pain during last week | 70   | 10               | 5                  |
| Pain in best period | 40   | 0                | 0                  |
| Pain in worst period | 85   | 35               | 10                 |
already for up to 6 months post-implantation. Previous studies that evaluated the effect of SNM on chronic idiopathic pelvic pain have shown positive results. Siegel et al. reported a significant improvement in pain symptoms in six out of ten patients with pelvic pain, which persisted after a median follow-up of 19 months [4]. Yet, no previous studies evaluated the effect of SNM treatment in clitoral pain. Although SNM was used as an off-label treatment for clitoral pain in our case, the results of this report suggest that the application of SNM in patients with clitoral pain might be effective.

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Conflicts of interest T. Marcelissen: none; Ph. Van Kerrebroeck is consultant and speaker for Medtronic; S. De Wachter is speaker for Medtronic.

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References
1. Everaert K et al (2001) The pain cycle: implications for the diagnosis and treatment of pelvic pain syndromes. Int Urogynecol J Pelvic Floor Dysfunct 12(1):9–14
2. Zabihi N et al (2008) Short-term results of bilateral S2–S4 sacral neuromodulation for the treatment of refractory interstitial cystitis, painful bladder syndrome, and chronic pelvic pain. Int Urogynecol J Pelvic Floor Dysfunct 19(4):553–557
3. Gordon AS (2002) Clitoral pain: the great unexplored pain in women. J Sex Marital Ther 28(Suppl 1):123–128
4. Siegel S et al (2001) Sacral nerve stimulation in patients with chronic intractable pelvic pain. J Urol 166(5):1742–1745