A novel approach to vulvodynia using targeted neuromodulation

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Vulvodynia is a debilitating disorder which can prove extremely difficult to treat. Neuromodulation is increasingly becoming a frontline therapy in various chronic pain syndromes. We present a relatively simple surgical technique utilizing targeted neuromodulation leading to the successful treatment of vulvodynia.

Key Words: analgesia, pain management, chronic pain, vulvodynia, neuromodulation,

Introduction

Vulvodynia is a debilitating disease with an estimated prevalence of anywhere from 8%-28%.[1,2] The etiology is unclear and often the diagnosis is one of exclusion. There are several proposed theories for the pathophysiology behind the development of vulvodynia including neuro-proliferation of nociceptors,[2] serotonin receptor gene polymorphisms, abnormally heightened inflammatory response,[3] and more. Functional MRI studies have demonstrated increased activation in known pain sensory areas including the anterior cingulate gyrus, somatosensory region, insular cortex, and frontal cortex,[4] suggesting a possible central nervous system link. In addition, both physical and sexual abuse have been associated with the development of vulvodynia.[2]

Finding an exact treatment for vulvodynia has been elusive, and as such, most patients receive numerous multimodal therapies including analgesics, pelvic floor physical therapy, psychotherapy, and more. Several recent reviews on the subject have concluded that cognitive behavioral therapy and pelvic floor physical therapy should be considered first-line in the management of vulvodynia given improvements in pain, sexual function, and the low risk nature of the therapy itself.[1,5] Both authors agreed that botulinum toxin showed promise, though it has been inadequately studied in these patients, and hormonal agents, psychotropic medications, antispasmodics, and topical

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capsaicin have yielded mixed results. Vestibulectomy is recommended for refractory cases of vulvodynia after exhaustion of these and other less invasive therapies.[5]

In recent years, neuromodulation has become an emerging therapy for the management of chronic pain syndromes. In fact, spinal cord stimulation has proven so successful in the management of chronic back pain that it is now an FDA approved treatment for chronic limb pain as well.[6] Similarly, neuromodulation via a more targeted approach for chronic pain at other sites has been recently studied, including targeting of the pudendal nerve for patients with chronic pelvic pain.[7,8]

We present a case of vulvodynia successfully treated via a novel surgical approach using targeted neuromodulation.

Case report

Our patient is a 63-year-old woman who presented to our clinic with a long history of focal vulvodynia involving her left labia after sustaining a laceration from a fall at age 23. Her symptoms were exacerbated by touch or rubbing, including by pants and underwear to the extent that she avoided wearing pants altogether. She believed the pain to be centered around a labial scar resulting from the fall. Her pain would worsen with stress, and she initially presented with urinary frequency and nocturia as well.

In the past, she had attempted numerous interventions with minimal symptomatic relief. Conservative therapies such as topical vitamin E, topical lidocaine, and sesame oil were unsuccessful, as was pelvic floor physical therapy. She underwent an excision of her labial scar without any improvement in her pain. Opioid analgesics offered mild relief. Given her concurrent urinary symptoms and the suspicion that her pain was triggered via a labial branch of her pudendal nerve, she underwent InterStim (Medtronic, Minneapolis, MN, USA) placement along her pudendal nerve which did result in improvement in her urinary symptoms but offered little to no pain relief.

Due to the refractory nature of her symptoms despite exhausting numerous treatment modalities, the patient ultimately consented to trialing placement of a neurostimulation device along the course of her pain. The StimWave 8-contact electrode (StimWave LLC, Pompano Beach, FL, USA) was chosen as it has 8 electrodes that covered the entire length of the labia. It also has an integrated receiver (ASIC) that allows for activation of the electrode using an external energy source which obviates the need for a cumbersome implantable pulse generator and is highly programmable, allowing for high-frequency stimulation to block pain signals. This electrode is activated by placing an antenna over the lead receiver and using a rechargeable external energy source to activate the internal ASIC to stimulate the nerves contributing to her pain. This results in personalized medicine as the patient can choose to activate the lead as often or as little as needed to control her symptoms.

She presented to the office for preoperative planning where her pain was mapped along the left labia via cutaneous stimulation, and the site was further confirmed in the preoperative area and marked. She then was taken to the operating room for placement of a temporary electrode. The patient was placed in the dorsal lithotomy position with the vaginal area exposed. A small skin incision was made superior to the left labia and the StimWave 8-contact electrode was tunneled within the labia along the course of her pain. The electrodes were deployed and an external antenna and energy source were positioned over the lead to activate the lead, Figure 1a. Electromyography

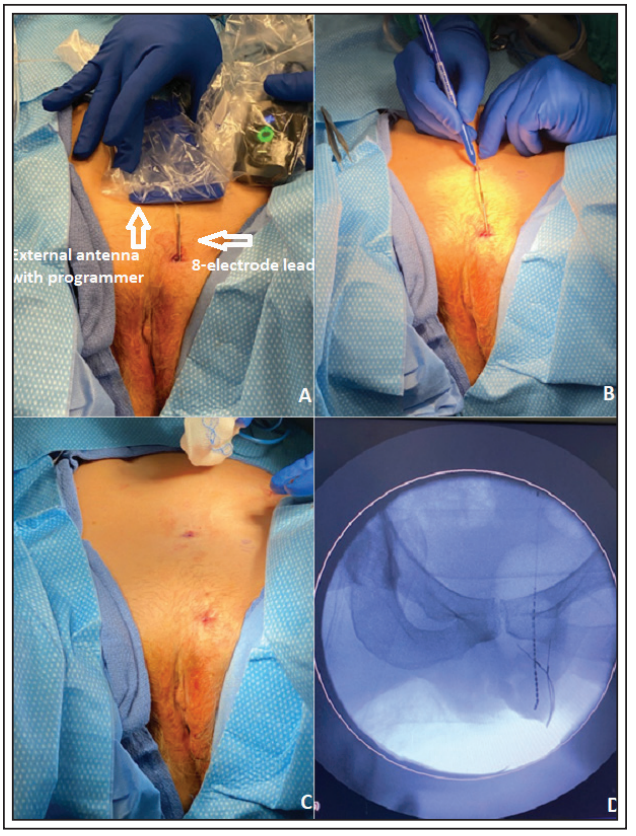

Figure 1. (A) Initial lead placement at superior left labia with application of external antenna (arrows). **(B)** Marking of abdominal wall for tunneling excess lead subcutaneously. **(C)** Final appearance upon closure with lead completely tunneled. **(D)** Fluoroscopic image depicting course of electrodes.

(EMG) electrodes which were placed in the pelvic musculature showed a response when the lead was activated, confirming functioning of the lead. The lead was then secured to the skin before closing the incision.

The patient followed up in the office 1 week later reporting near 100% improvement in her symptoms, and her only complaint was slight swelling and tenderness at the lead's insertion site. The lead was removed in the office without difficulty while awaiting insurance approval for the permanent lead placement, and her pain returned within several days of removal. Approximately 1 month later, the patient returned to the operating room for placement of her permanent lead. This was placed in similar fashion within the left labia with EMG monitoring to ensure lead functioning. However, because this lead was to be permanent, the redundant proximal portion of the lead was tunneled subcutaneously up her abdominal wall, and a small subcutaneous pocket was created for placement of the copper receiver and excess lead, Figure 1b-c. Fluoroscopic imaging was obtained to document lead position, Figure 1d. In the recovery room, the lead was activated using an external antenna and rechargeable energy source and programming parameters were set to provide optimal pain relief. The patient was instructed to stimulate the lead 8 hours per day and to titrate the stimulation duration based on her needs.

Once again, the patient experienced near 100% resolution of her pain within 24 hours of the procedure. At 8 weeks postoperatively, she reported that her pain was essentially non-existent and that she could finally wear pants without pain. She stimulates the lead using the external antenna by positioning the energy source over the implanted receiver on her lower abdomen. She was contacted 1 year postoperatively and her pain remains virtually non-existent. Currently she wears the device once every few days for the majority of the day. She said that the surgery has been "like a miracle."

Discussion

Vulvodynia is a notoriously difficult disease to satisfactorily treat adequately, and this difficulty often leads to frustration for both patients and providers. As discussed earlier, there is no single most effective treatment, and successful management often requires multimodal therapy and trial and error. This leads to a significant burden on the healthcare system, representing up to 72 billion dollars annually.[2]

In our patient, who had exhausted nearly all other treatment options aside from vestibulectomy, a relatively simple, minimally invasive outpatient procedure was able to nearly completely resolve her symptoms with lasting efficacy. She must simply wear her external antenna, which can easily be concealed under clothing, and activate her device for several hours per day with lasting results. This has led to a dramatically improved quality of life to the point that she can once again wear pants without fear of debilitating pain. Perhaps more importantly, she has also been able to avoid the use of narcotics which is becoming increasingly more prescient in the era of the opioid epidemic.

While the treatment of vulvodynia with neuromodulation may be a novel treatment, neuromodulation is being increasingly studied in a large variety of pain syndromes.[7-9] Perhaps the simplest form of neuromodulation is transcutaneous electrical nerve stimulation (TENS) which has been extensively studied in chronic back pain. Though a meta-analysis by Wu et al[9] revealed that TENS was no more effective than placebo in improving functional disability in patients with chronic back pain, other forms of neuromodulation including percutaneous electrical nerve stimulation and percutaneous neuromodulation proved more effective. The authors surmised that the increased efficacy resulted from more direct delivery of electrical stimulation to the nerves in question.[9] Additionally, neuromodulation has been evaluated in chronic pelvic pain, leading Tam et al[7] to conclude that there is evidence to suggest possible benefit of electroacupuncture and TENS in this cohort of patients. Peters et al[8] compared sacral and pudendal neuromodulation in patients with irritative voiding symptoms, demonstrating superiority of pudendal neuromodulation for improving concurrent pelvic pain.

The vulva is innervated by the anterior labial branches of the ilioinguinal nerve, genitofemoral nerve, and branches of the pudendal nerves. Previously, pudendal neuromodulation with the lead positioned near the pudendal nerve via an ischial-rectal approach did not result in any improvement in her pain. However, once we positioned a lead in close proximity to the suspected source of pain, the patient experienced dramatic results, suggesting that the direct application of electrical energy to affected nerves produces the most efficacious results. The exact mechanism of neuromodulation has yet to be fully elucidated, but the most widely accepted theory is the gate control theory of pain signaling in which non-painful afferent signaling from the stimulation device can block transmission of pain signals.

Certainly, it is difficult to make conclusions based off the experience of a single patient. However, given the success of neuromodulation in the management of other pain syndromes, targeted neuromodulation may represent a new avenue in the management of

vulvodynia with minimal invasiveness or side effects while avoiding a larger surgery or chronic narcotics. This novel intervention requires further evaluation, ideally with randomized controlled trials, but it shows significant promise.

Disclosures

Dr. J.R. Stephens has no disclosures. K.M. Peters is a co-founder of Micron Medical and serves as a consultant for Urogen and Urovant. □

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