

Diagnosis and Conservative Chiropractic Care of Chronic Idiopathic Pudendal Nerve Entrapment Causing Saddle-Like Paresthesia and Restless Genital Syndrome: A Case Study

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Abstract

Introduction: To describe and discuss the diagnosis and chiropractic treatment of a patient with chronic idiopathic pudendal nerve entrapment that presented with saddle-like paresthesia and caused Restless Genital Syndrome.

Aim: A 43-year-old female patient had symptoms consistent with a pudendal nerve entrapment, saddle-like paresthesia, and restless genital syndrome for 1.5 years prior to starting care with a chiropractor. She had been to numerous medical providers, none of which offered the correct diagnosis or treatment options.

Methods and Results: Chiropractic manipulative therapy, myofascial release, and instrument-assisted soft-tissue mobilization was directed at the pelvic musculature with the primary focus on the right obturator internus. Through one month of chiropractic care the patient's symptoms were resolved including restoration of regular sexual function.

Conclusion: Chronic perineal pain can be found in cases with numerous diagnoses, with pudendal nerve entrapment being one that is rarely documented. There are a wide variety of causes of PNE that include but not limited to direct injury, tumor, ganglion cysts, anatomical anomaly, and extended time cycling. Documented treatment for pudendal nerve entrapment is limited, primarily to pain injections. This case demonstrates how chiropractic care and soft tissue mobilization can correct the dysfunction.

Keywords: Chiropractic; Neuralgia

Introduction

Pudendal neuralgia (PN) is a distressing condition and its management is often not evidence-based [1]. PN is a rare neurological condition and is defined by burning vaginal or vulval pain (anywhere between the anus and the clitoris) associated with tenderness over the course of the pudendal nerve [1,2]. PN due to pudendal nerve entrapment (PNE) is related to loss of mobility of the pudendal nerve over its gluteal or pelvic course. As the pudendal nerve courses its way through the pelvis, its loss of mobility is due to anatomical impingement [3] in various locations to include: in the reflection of the obturator fascia, in the space between the sacrospinous ligament and the sacrotuberous ligament or in the infrapiriformis canal. The loss of mobility induces compression of the pudendal nerve against the falciform process of the sacrotuberous ligament while sitting. As in all canal syndromes, these anatomical structures are also present in asymptomatic patients. Onset of pain and the range of symptoms can involve other elements related to central sensitization phenomena or muscular or visceral reactions/reflexes.

The pudendal nerve provides both sensory and somatic function to the genital region, including the urethra, anus, and perineum. PN is also known as pudendal neuropathy, pudendal nerve entrapment, pudenda canal syndrome, or Alcock's syndrome. PN was first described in a case in 1987 where it was called "Perineal Paralysis of Cyclist." This pathology was due to a history of repetitive cycling [2]. The prevalence of this condition in the general population is thought to be ~1%, with women being affected more than men [4]. Patients can experience a wide variety of symptoms that are neuropathic in nature due to its various functions as a sensory and somatic nerve. Sensory symptoms can include severe pain, paresthesia, and/or altered sensitivity in the nerve's dermatome [5]. In rare cases, saddle-like paresthesia is noted. A change in sensation with urination or defecations may also be manifested as pain or an altered sense of urgency [6]. Changes in sexual function may also be noted, including pain, reduced sex drive, or an individual may have constant unwarranted genital stimulation, also known as Restless Genital Syndrome (ReGS) [6,7]. Due to the

somatic function of the pudendal nerve within the act of urination and defecation, dysfunction within those processes is also possible. Due to a high variance of PN symptoms, the diagnosis of PN is often difficult for clinicians [3].

The treatment for PNE is primarily symptomatic. Described treatment has included neuropathic pain medications, transcutaneous neurostimulation, physiotherapy, body psychotherapy, and hypnosis. Anesthetic blocks of the pudendal nerve have been deemed necessary to confirm the diagnosis. A greater than 50% reduction in pain while sitting immediately after infiltration confirms the role of the pudendal nerve. Corticosteroids are usually injected at the time of the anesthetic nerve block with the therapeutic goal of minimizing the inflammatory component [8,9]. The largest series of 170 patients with PN reported resolution in 45% and improvement in 22% with surgical neurolysis. In the same study, imaging-guided pudendal nerve blocks offered short-term improvements in 65% of the patients [3].

The purpose of this case report is to demonstrate a treatment approach in which chiropractic manipulative therapy and soft-tissue mobilization was utilized to treat a patient who had a pudendal nerve entrapment at the obturator internus muscle. This condition further manifested itself causing saddle-like paresthesia and ReGS. There is little evidence in the literature discussing manual therapy and chiropractic to various nerve entrapment sites that may affect the pudendal nerve, causing neuralgia-like symptoms. This case provides

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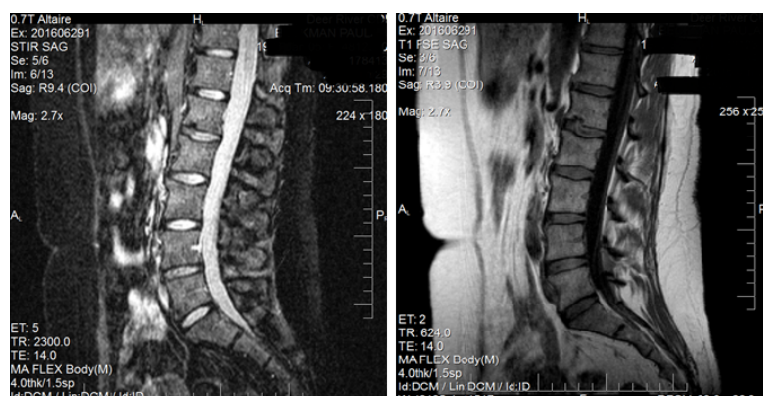


Figure 1: Findings on MRI.

more evidence towards the possible presenting symptoms of PN and complicating factors, as well as a treatment plan that was successful in resolving the condition.

Case Report

Methods

A 43-year-old female secretary presented to a chiropractic clinic with chronic saddle-like paresthesia. She had experienced these symptoms for 1.5 years. The patient described the symptoms as a numb and tingling sensation in the anterior and posterior groin. She reported that it feels like “biofreeze” was constantly being applied to the area in which would contact a saddle if sitting. Her prior course of care had consisted of 3 gynecological exams which included several hormonal and blood tests done to help diagnose her condition. Hormonal testing and blood work came back negative. She had also consulted with her primary medical physician. Multiple times she was told to seek psychological counseling, which only frustrated the patient as she did not feel it was a psychological condition. Further symptoms consisted of constant, unwarranted genital stimulation that has been co-occurring with the paresthesia. She had urgency with bowel and bladder movements, indicating that when she had the urge, she had to go immediately, however she still could maintain control of her bowel and bladder function. She was unsure of the cause of her symptoms and denied a traumatic injury. She did have a previous history of horseback riding and had injured her coccyx in the past but previous sacrococcygeal x-rays were negative for fracture. She indicated that she was treated conservatively for that condition with full resolution of coccygeal pain. She did indicate that she had been under chiropractic care as needed for the past 5 years for various conditions but never mentioned her saddle-like paresthesia since it had been ongoing the past 1.5 years.

Upon examination, there were no neurological deficits with sensory, motor, and reflex testing. Sensory testing was performed with the patient’s eyes closed having her distinguish between sharp and dull. She could identify all stimuli successfully in L1-S1 dermatomes of the lower extremity. Low back and pelvic orthopedic exams were all negative. Palpation findings showed tenderness, swelling, and taut and tender fibers primarily in the right obturator internus with associated muscle spasms throughout the right gluteal region. Also noted were taut and tender fibers on the paraspinals from T12-L5 bilaterally.

Due to the saddle-like paresthesia, a lumbar MRI was ordered immediately to rule out cauda equina syndrome. The radiological examination reported a disc bulge at L4-L5 with associated

osteophyte formation. The right L4-L5 foramen was compromised by the osteophyte as well as stenosis. Mild spondylosis was reported throughout the remainder of the thoracolumbar spine. There was no radiographic evidence of the causative agent of the saddle-type paresthesia, nor was there evidence of cauda equina compression. Consultation with a radiologist determined no contraindications to conservative chiropractic care. Furthermore, the chiropractor and radiologist felt that the findings on the MRI did not explain the patient’s current symptoms (Figure 1).

Following the MRI, the patient was diagnosed with Pudendal Neuralgia based on the patient history, clinical presentation, and radiographic findings. It was determined that the PN was causing the saddle-like paresthesia and the ReGS was a complication of the PN.

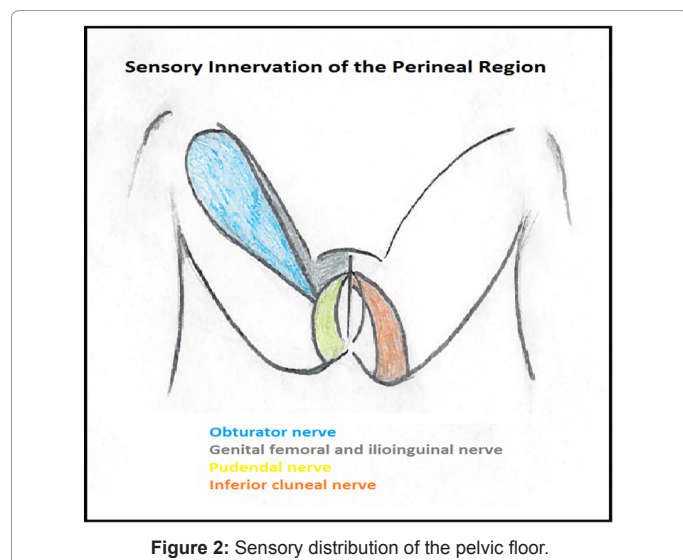
The patient was treated conservatively for a 5-week period, first being seen 2 times a week for 3 weeks, and then 1 time a week for 2 weeks. Each treatment consisted of instrument-assisted soft-tissue mobilization, specifically the Graston Technique, followed by manual myofascial release to the pelvic and gluteal region on the demonstrated taut and tender muscles, with most of the focus on the right obturator internus. Patient’s low back and pelvis were also treated with chiropractic manipulative therapy (CMT) where segmental dysfunction was noted at each visit.

Results

Over the course of the 5-week treatment time, the patient’s intensity and frequency of the paresthesia had completely resolved. She no longer experienced unwanted genital stimulus and could achieve a normal, desired orgasm which wasn’t possible prior to the start of the treatment plan. Better urinary and bowel function was also described with better overall sensation and decreased sudden urgency. A VAS was used to monitor the intensity of the symptoms. Initial VAS score was 5/10, and her score at re-evaluation was 1/10. A Back-Bournemouth Questionnaire (BBQ) was used to monitor the low back pain. Initial BBQ score was 71% and her re-evaluation score was 4%. The standard diagnostic criteria for Restless Genital Syndrome were also used. Her initial diagnosis of ReGS was made following a score of 5/5 from the criteria. At re-evaluation, her score was 0/5. The patient’s final evaluation was done approximately 6 weeks after the final treatment with her score’s staying the same as the re-evaluation scores. The patient was discharged from care at that time. A 3 month follow up via conversation was had and the patient remained symptom free.

Discussion

To our knowledge, this is one of the few case reports demonstrating



the effectiveness of chiropractic intervention combined with manual therapy to treat PN. This case study fully describes an interesting case of PN, presenting with saddle-like paraesthesia and ReGS. This report further provides a complete chiropractic protocol for the complexity of the pudendal nerve entrapment to alleviate the symptoms. Our treatment was the best for the patient because we could identify the cause of the pathology, thus offering conservative treatment to resolve the symptoms and underlying condition. This allowed the patient to forgo invasive procedures surgical procedures or pain injections. The saddle-like paraesthesia and ReGS caused by the pudendal nerve entrapment within the right Alcock's Canal was primarily treated with soft tissue therapy. The myofascial release and IASTM broke up the adhesions within the obturator internus and surrounding structures that had entrapped the nerve, causing it to be chronically irritated. Releasing the nerve entrapment and decreasing the irritation also allowed the resolution of aberrant efferent signals that caused the unwanted genital stimulus. By removing the neurological interference, the patient could have no pain or altered sensation in the perineal area and return to having regular sexual function.

The pudendal nerve is a mixed nerve that originates from the ventral rami of the S2, S3, S4 and sometimes S5 nerve roots. These nerve roots join to form the sacral plexus. From the sacral plexus, these branches form the pudendal nerve. From its origin of the sacral plexus the peripheral nerve travels into the pelvis through the lesser sciatic foramen where the sacrospinous ligament overlies its fibers. Once leaving the foramen it joins with the internal pudendal vessels and travels to the ischiorectal fossa. There it becomes surrounded in the obturator internus fascia known as the pudendal canal, or Alcock's Canal. The nerve then gives off three branches just distal to Alcock's Canal. The first branch is the inferior rectal nerve that travels caudally where it continues as a mixed nerve. The sensory portion relays sensation from the terminal portion on the anal canal and the surrounding area. The motor neurons somatically control the external anal sphincter. The second branch, the perineal nerve, is also a mixed nerve that innervates the pelvic floor musculature and provides sensory innervation to the perineum and part of the labia majora on the respective side. The terminal branch, the dorsal nerve of the clitoris, receives sensory stimuli from the clitoris (penis in males) [6]. The pudendal nerve is predisposed to entrapment at 2 sites: the ischial spine by compression of the sacrospinous and sacrotuberous ligaments and the pudendal canal by compression of the falciform process of the sacrotuberous ligament or thickening of the obturator fascia [10,11].

Sensory innervation of the pelvic floor is shown above in Figure 2. It includes the area between the anus and penis or clitoris [12]. Symptoms from a pudendal nerve injury or entrapment can be purely sensory, purely motor, or a mixture of the two. Most neuralgias resolve quickly, although those that are longer lasting can be very traumatic for the individual involved. The most frequent finding of dysfunction is bilateral decreased sensation to the perineum and labia in women and perineum and scrotum in males. Symptoms of pain usually resolve quickly, while those that are sensory in nature tend to last longer and can accompany sexual disorders as well. Anatomically, the obturator internus is a common entrapment site of the pudendal nerve due to muscular spasms [13,14].

In diagnosing PN, the essential criteria, which has become the gold standard for clinicians to use when diagnosing or ruling out PN includes:

- Pain in the sensory innervation of the pudendal nerve.
- Pain worse with sitting.
- The patient is not disturbed at night by the pain.
- There is no objective sensory loss on clinical examination.
- There is a positive response to an anesthetic pudendal nerve block [12,15].

Complementary diagnostic criteria include numbness in the sensory innervation. Exclusion criteria includes: exclusively coccygeal, gluteal, pubic, or hypogastric pain, pruritus, and exclusively paroxysmal pain [12,15]. This criterion is known as Nantes Criteria and was developed in 2008 by a group of clinicians to aid in diagnosing this elusive condition [15]. The 5 parts of Nantes Criteria highly suggests a case of PN if the parts are fulfilled. However, it should be stated that due to the wide variety of presenting symptoms from case to case that use of the Nantes Criteria in combination with a proper history and quality exam will allow the best chance for a proper diagnosis [12].

Another contributing factor that makes proper diagnosis difficult is the multiple reported causes of PNE. The most common mechanism is mechanical compression of the nerve [6]. This compression can be from direct contact with muscles in the pelvic girdle, an increase in pelvic pressure, various tumors, surgical mesh from pelvic repair [16] or a ganglion cyst(s). There are numerous locations for this to happen, but the most common place being in the obturator internus, more specifically, Alcock's canal [17]. Other muscles within the pelvis may also cause compression of the nerve during spasm. The irritation caused by this mechanical stress on the nerve causes the neuropathic symptoms. A second mechanism is a direct trauma to the nerve. This trauma could be compressive, tensile, or tearing in nature which causes direct structural damage to the nerve itself [18]. A third mechanism of the condition is biochemical stress to the nerve via other pathologies [19]. Pathologies include, but not limited to, herpes infection, diabetes, and multiple sclerosis. There is also speculation that physiological changes during and after surgery, including bleeding in the pudendal nerve can cause the condition [20]. A fourth mechanism is a central dysfunction of the spinal nerve roots that cause the peripheral dysfunction.

According to the Nantes Criteria, the patient fit 3/5 components; part 1, 2, and 4. In the history the patient states waking up throughout the night due to the various symptoms, which excludes her from part 4. A nerve block wasn't performed because the patient was responding to conservative care, therefore it can't be stated whether the patient would've fit part 5 [12]. Regardless, she had met 3 of the 5 components of Nantes Criteria.

As previously stated, there are many causes of PN. Some can be treated conservatively while others cannot. Due to saddle-like paraesthesia the MRI was ordered to rule out Cauda Equina syndrome or other causes of PN that require a more invasive treatment. However, a more serious pathology, to include Cauda Equina, malignancies, vertebral fractures, discogenic pathology, tumors and cysts were ruled out given the radiographic report. With no radiographic evidence suggesting the cause of the symptoms, it was determined that the pudendal nerve was entrapped at Alcock's canal and the describe symptoms were directly related to the entrapment. Due to the saddle-like paraesthesia, the entrapment would have to be proximal to where the nerve divides. Although in most cases neurologic injuries are transient in nature and resolve within a few days to weeks, this case study illustrates the importance of proper diagnosis and treatment to address longer lasting symptoms. Improving interdisciplinary communication to achieve better patient outcomes is a goal to which every heal care profession should strive. In our case study, it was important following care that the patient did report back to her other healthcare professionals about the care treatment and the results achieved. As mentioned, the patient went through many other primary care providers and numerous exams and treatments before the proper diagnosis was given.

Limitations

There are some limitations with this case as well. The patient had other co-morbidities complicating her condition. She had a known disc herniation at the L4-L5 level, likely contributing to her LBP but not the cause of the PN or the associated symptoms. This was treated with CMT. The chiropractic manipulation to the lumbopelvic region did help correct her LBP but it is also reasonable to suggest that proper lumbopelvic biomechanical function will further help alleviate muscular tension, which was evident in the right obturator internus. Furthermore, as mentioned above, the patient only fit 3 out of the 5 criteria on Nantes Criteria. She was experiencing pain at night, which would exclude her from part 3, but again she never did receive a pudendal nerve block. Further limitations include a lack of blinding. Both the patient and the clinicians were aware of the treatment provided. Another factor was the patient's difficulty in following the treatment plan until the end. She was unable to complete the re-evaluation when scheduled. This was due to scheduling conflicts as well as lack of urgency. The patient stated she was doing so well that she didn't feel the need to return right away. If she could follow the treatment plan more closely her OAT may have been able to improve further and a better observation of necessary treatment could've been made. There are also limitations within the format of a single case study. Without more patients of similar presentations and treatment of PNE, we cannot determine the treatment is effective for the general population.

Conclusion

This is a case description of a patient who was suffering from PN for 1.5 years. The patient responded favorably and quickly to CMT, instrument-assisted soft-tissue mobilization, and manual myofascial release at the entrapment site of the pudendal nerve. To the authors' knowledge, this is a rare case report which demonstrates the resolution of PN using conservative chiropractic care, allowing the patient to avoid more invasive treatment options such as a pudendal nerve block or

surgical intervention. The results suggest that the treatment described in this case report can aid in the recovery of PN. The resolution of this patient's symptoms warrants further studies to investigate the effectiveness of conservative care for PN.

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