

Pudendal nerve entrapment in an Ironman athlete: a case report

Jaclyn A. Durante, BSc, DC*

Ian G. MacIntyre, DC, FCCSS(C)**

Objective: To present the diagnostic and clinical features of pudendal nerve entrapment and create awareness amongst clinicians of this rare and painful condition.

Clinical Features: A 41-year old male ironman athlete complaining of insidious constant penis pain 12–24h after long distance cycling and pain after sexual intercourse. A diagnosis of “cyclist syndrome” also known as pudendal nerve entrapment was made.

Intervention and outcome: Patient was treated twice a week for four weeks using the soft tissue protocol described by Active Release Technique® to the obturator internus muscle. After two weeks of treatment his pain decreased to a 5/10 on the pain intensity scale and he began to cycle again. After four weeks of treatment his pain had decreased to 1/10 in intensity and he continued to cycle. At follow-up, approximately 8 weeks and 12 weeks later the patient communicated that his pain is resolved and he has begun to train for Ironman Lake Placid 2010.

Conclusion: Pudendal nerve entrapment is a rare, painful condition and is often misdiagnosed due to the fact that the clinical manifestations can mimic other pathologies. It is important to be aware of the clinical

Objectif : Soumettre un diagnostic et des particularités cliniques de compression chronique du nerf honteux, et sensibiliser les médecins à cette affection rare et douloureuse.

Particularités cliniques : Un athlète masculine Ironman de 41 ans se plaint de douleur constante et insidieuse au pénis entre 12 et 24 heures après un long périple à vélo, et une douleur après avoir eu des relations sexuelles. Un diagnostic de « syndrome du cycliste », également connu sous le nom de compression chronique du nerf honteux, fut émis.

Intervention et résultat : Le patient fut traité deux fois par semaine pendant quatre semaines à l'aide du protocole du tissu mou décrit par Active Release Technique® sur le muscle obturateur interne. Après deux semaines de traitement, sa douleur avait diminué à 5/10 sur l'échelle d'intensité de la douleur, et il a recommencé à faire du vélo. Après quatre semaines de traitement, sa douleur avait diminué à 1/10 sur l'échelle d'intensité, et il a continué de faire du vélo. Lors du suivi, environ 8 et 12 semaines plus tard, le patient a affirmé que sa douleur avait disparu, et qu'il s'entraînait en vue de la compétition Ironman à Lake Placid en 2010.

Conclusion : La compression chronique du nerf honteux est une affection rare et douloureuse, souvent mal diagnostiquée en raison du fait que les manifestations cliniques ressemblent à autres pathologies. Il est important de connaître les

* Graduate Student, Sport Sciences, Canadian Memorial Chiropractic College, Toronto, Canada

** Associate Professor, Canadian Memorial Chiropractic College, Toronto, Canada

Address correspondence to: Dr. Jaclyn Durante, 6100 Leslie St., Toronto, Ontario M2H 3J1

Phone: (416) 482-2340 ext. 242 Fax: (416) 482-2560

E-mail: jdurante@cmcc.ca

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features to obtain appropriate diagnosis and treatment of this condition promptly.

(JCCA 2010; 54(4):276–281)

KEY WORDS: pudendal nerve, entrapment, neuralgia, obturator internus, pudendal canal, ART®

particularités cliniques pour émettre un diagnostic exact et traiter l'affection rapidement.

(JCCA 2010; 54(4):276–281)

MOTS CLÉS : nerf honteux, compression chronique, obturateur interne, canal honteux, ART®

Introduction

Pudendal neuralgia by pudendal nerve entrapment is described as severe, sharp pain along the course of the pudendal nerve.¹ Genital numbness and erectile dysfunction are two of the major symptoms and the prevalence has been reported to be 50–91% and 13–24% respectively. This can be caused by trauma, infection, tumour, child birth, iatrogenic injury, surgery and/ or microtrauma from cycling.² It has been reported that 7–8% of cyclists on long-distance multiday rides experience pudendal neuralgia.³ Diagnosis of pudendal nerve entrapment is often delayed or misdiagnosed, causing people to suffer with this for 2–10 years.⁴ It is important to understand the clinical presentation and diagnostic criteria to allow early diagnosis and appropriate treatment.

Case

A 41-year old male ironman athlete complained of insidious onset of constant penis pain and a dull ache in the perineum 12–24h after long distance cycling. His cycling training consisted of 6–11 hours per week, 3 days a week. The pain began in the beginning of September 2008 and by mid-September he was experiencing pain in the perineum after sexual intercourse and burning during urination. He visited with his medical doctor who diagnosed him with prostatitis and he was prescribed antibiotics for 30 days. He returned to his medical doctor who informed him that STDs were ruled out and he was told that the prostatitis was under control, however the pain was unchanged. At that time, the prostate was palpated for enlargement, an ultrasound was performed to examine the prostate and prostate serum antigen was measured. These tests proved to be normal. In early October he was referred to a urologist who prescribed him with another antibiotic for 30 days. By November 2008, the pain was rated 9/10 in intensity. He saw another urologist who explained that the infection and cycling caused inflamma-

tion of the pudendal nerve. At that point he was prescribed naproxen, 750 mg/day. He proceeded to see a sport medical doctor, who told him to experiment with new saddles for his bike. He completed the ironman in late November 2008 and ceased to ride from November 2008 to January 2009. With rest and the use of naproxen the pain intensity decreased to 5/10.

In January 2009, he began to cycle again. However, by February the pain increased to 7/10. For relief he would sit on a frozen water bottle placed in the perineal region and use a foam roller to massage his buttock region. He experimented with a new saddle and it seemed to provide relief. He competed in the ironman in July 2009 and discontinued to ride from July to September 2009 and the pain decreased to 3/10 in intensity. He began to cycle again in September to train for a half-ironman in late fall. The pain intensity increased to 9/10. At this point he was seen by a neurologist who diagnosed him with “cyclist syndrome” and he was prescribed naproxen, rest and to experiment with a new saddle.

In October 2009, he visited a chiropractor to receive treatment. Upon physical examination, hyperalgesia was found during palpation of the lesser sciatic notch and the obturator internus muscle. Palpation in this area reproduced the 9/10 pain in his penis and perineum and caused fasciculations in the surrounding buttock musculature. A working diagnosis of pudendal nerve entrapment was reached. He was treated twice a week for four weeks with only Active Release Technique (ART)® obturator internus muscle protocol. After the first treatment it seemed to aggravate the symptoms, however at two weeks of treatment his pain was decreased to a 5/10 and he began to cycle again. After four weeks of treatment his pain had decreased to 1/10 and he continued to cycle. Eight weeks later his pain after sexual intercourse and the pain in his penis and perineum resolved and he has begun to train for another ironman.

Discussion

Pudendal neuralgia, due to entrapment of the pudendal nerve (PN) has a varied clinical presentation and some suggest that it is due to the anatomical course of the nerve and the fact that the nerve is mixed. The PN arises from the roots of the sacral plexus S2-S4 where 20% consists of motor fibres, 50% are sensory fibres and the last 30% are autonomic.^{5,6,7} The PN descends medially and caudal in relation to the trunk of the sciatic nerve.^{5,6,7} From there it passes laterally and enters the gluteal region and then traverses the greater sciatic foramen.^{5,6,7} Along its course the PN is accompanied by its artery (internal pudendal artery) and vein (internal pudendal vein). The pudendal bundle passes around the termination of the sacrospinous ligament just as it attaches onto the ischial spine.^{5,6,7} The PN is now situated between the sacrospinous ligament ventrally and the sacrotuberous ligament dorsally. The PN travels into the perineal region via the lesser sciatic foramen, where it lies underneath the plane of the levator ani muscle.⁵ From here it enters a duplication of the fascia of the obturator internus muscles and its medial aspect, which forms the pudendal canal.⁵ It is at this location where the PN divides into its three terminal branches: inferior rectal nerve, perineal nerve and the dorsal nerve to the penis/clitoris.⁵

The inferior rectal nerve supplies the integument around the anus, provides sensation to the distal aspect of the anal canal and to the perianal skin.^{5,8} It also provides motor innervation to the external anal sphincter.^{5,8} The perineal nerve supplies motor innervations to the pelvic floor muscles (transverse perineal muscles, bulbospongiosus, ischiocavernosus, sphincter urethra and the levator ani muscles).^{5,8} The sensory branches of the perineal nerve supply the perineum and the ipsilateral posterior surface of the labia majora. Finally, the dorsal nerve of the penis /clitoris is the most superficial branch found at the level of the pubic symphysis.^{5,8} This nerve carries afferent sensory information from the clitoris/penis.^{5,8}

The anatomical course of the PN has great implications regarding the potential sites for its entrapment. The following are areas in which the PN can become entrapped leading to injury and symptoms of pudendal neuralgia: the zone at the ischial spine, the zone between the sacrospinous and sacrotuberous ligaments, the falciform process (formed by the dense fanning out bands of the medial portion of the sacrotuberous ligament as it attaches to the



Figure 1 *Right arrow: piriformis; left arrow: pudendal nerve.* Image courtesy and copyright Primal Pictures Ltd.

ischial tuberosity), the zone between the levator ani and obturator internus muscle, within the pudendal canal and lastly compression from the piriformis muscle (Figure 1).^{1,5,6,9} A new entrapment site was proposed, which described a distinct narrow osteofibrous canal measuring 3cm long found in all 10 male dissected cadavers.⁷ This canal was bordered by the pubic symphysis dorsal-cranially, the inferior pubic ramus and suspensory ligament laterally and the corpus cavernosum medially.⁷ The dorsal nerve to the penis was found to travel in this canal and thought to be a potential site for entrapment.⁷

The exact pathophysiology of pudendal nerve entrapment is not completely understood, however there are proposed theories. Neuronal insult can be caused by stretching or compression of the nerve, which may lead to ischemic neuropathy due to transient hypoxemia of the PN.² This can result in changes to the intraneural micro-circulation and fibre structure, such as fibrosis and or inflammation, leading to impairment of axonal transport of the PN.² During cycling, repetitive impacts generate extreme perineal pressure leading to increased friction in the pudendal canal and compression from the surrounding

muscular (levator ani, obturator internus, and piriformis) and ligaments structures (sacrospinous and sacrotuberous).^{2,10,11} This constant pressure can lead to loss of gliding of the PN putting it at risk for entrapment.^{2,10,11} Also, the biomechanical position adapted while cycling, forward lean posture with arms on the aerobars of the bike, further increases the compression in the perineal region due to pressure from the nose of the saddle.^{2,10,11} This chronic perineal microtrauma results in the pathophysiological changes mentioned above.²

As mentioned previously, the clinical presentation of pudendal neuralgia is varied due to the fact that the PN is a mixed nerve and has numerous potential entrapment sites creating diverse symptomatology depending on the location of compression. Patients will experience remitting and relapsing neuropathic pain in the distribution of the PN often described as burning, tearing, stabbing, lightning-like, sharp, electrical, and shooting.⁶ Genital numbness, the most common symptom with a prevalence of 50–91% in bicyclists, is often the first sign and may be considered a warning sign to clinicians.^{10,12} Erectile dysfunction is often associated with pudendal nerve entrapment having a prevalence of 13–24% in bicyclists.¹⁰ Pain and paresthesia, which often begin in one location and spread, may extend as far as the groin, inner leg, buttock, and abdomen.⁵ Patients may also experience hyperesthesia and allodynia so intense that they avoid wearing certain clothing that may irritate the area.^{5,6} Pudendal neuralgia presents most commonly as unilateral, but in certain instances may be bilateral.^{5,6,13} The symptoms are usually aggravated by sitting or cycling and absent or relieved by standing, recumbent position and sitting on a toilet seat.⁸

The physical examination findings of patients suffering from PN entrapment are less concrete. Pain may be elicited during application of pressure on the PN at the ischial spine and palpation inferio-medially to the sciatic notch.^{6,14} The most common finding is tenderness with direct palpation of the obturator internus muscle.⁴ The symptoms may be worsened by performing passive internal and external rotation of the hip and resisted abduction and adduction of the hip flexed to 90 degrees.⁴

There is no widely confirmatory test available to diagnose pudendal neuralgia explaining why it is often misdiagnosed or delayed.⁸ It has been reported that proper diagnosis can take anywhere from 2–10 years.⁴ Pudendal

neuralgia is a diagnosis of exclusion, therefore all other causes of the symptoms must be ruled out. However, diagnostic criteria for pudendal neuralgia was established and validated by a multidisciplinary working party in Nantes, France and by members of the Francophone Perineal Electrophysiology club in 2008.¹⁵ The criteria are broken down into four diagnostic domains (Table 1).

Following diagnosis of pudendal neuralgia by PN entrapment, treatment usually involves a course of conservative medical care management. Practitioners may begin by prescribing anti-inflammatory and or nerve pain medications. Patients may opt to have a pudendal nerve block with or without the addition of steroids, which have the potential to relieve the symptoms for a longer period of time.⁵ Surgical decompression of the PN may be an option for those who encounter only temporary relief with a nerve block.^{5,9,14} There are three different approaches and these include: transperineal, transgluteal, and via the ischio-rectal fossa.^{5,14} Working in collaboration with medical management, there are several recommendations regarding cycling that one may choose to implement to reduce the symptoms. These include: stretches, short-term cessation of cycling (3–10 days), weight loss, wider saddle, absent or flexible nose on the saddle, tilt the saddle slightly downward, use of a gel saddle, change body posture to a more upright position, rise out of the saddle for 20–30 seconds every 20 minutes, take breaks during long rides and ensure that the bike has been properly fitted.^{1,2,3,10,13,16}

In the case presented the chiropractor treated the patient using Active Release Technique (ART)[®], which is described as a hands-on touch and case-management system that allows a practitioner to diagnose and treat soft-tissue injuries.¹⁷ It is performed by contacting the effected muscle, applying a tension and asking the patient to actively contract/shorten the muscle followed by actively stretching/tensioning the muscle.¹⁷ The ART[®] protocol for obturator internus treatment is described as follows. The patient is to begin in the side lying position, involved side up, and the hip in the anatomical position or abducted. The practitioner then places their contact on the distal portion of the muscle and draws tension medially. The practitioner must maintain or increase tension and move the hip into flexion, adduction and external rotation (an assistant may be used to externally rotate the hip).¹⁷ This procedure is conducted several times along

Table 1 Summarized from Labat et al 2008, Diagnostic Criteria for Pudental neuralgia by Pudental Nerve entrapment (Nantes Criteria)

Essential Diagnostic Criteria	Complementary Diagnostic Criteria	Exclusion Criteria	Associated Signs Not Excluded the Diagnosis
Pain in the territory of the PN	Neuropathic pain	Exclusively coccygeal, gluteal, pubic or hypogastric pain	Buttock pain on sitting
Pain predominately with sitting	Allodynia or hyperpathia	Pruritus	Referred sciatic pain
Pain with no objective sensory impairment	Rectal or vaginal foreign body sensation	Paroxysmal pain	Pain referred to the medial aspect of the thigh
Pain relieved by diagnostic nerve block	Pain worsening during the day	Imaging abnormalities able to account for the pain	Urinary frequency or full bladder
Pain does not wake the patient at night	Predominately unilateral		Pain after ejaculation
	Pain triggered by defecation		Dyspareunia and or pain post-coitus
	Presence of exquisite tenderness on palpation of the ischial spine		Erectile dysfunction
	Clinical neurophysiology findings of motor latency		Normal clinical neurophysiology test

the muscle until the practitioner subjectively determines that the tissue is moving properly and the adhesions are no longer palpated.¹⁷

To date there is no research describing the therapeutic effect ART[®] has on muscle injury. Contemporary soft tissue literature has been focusing on the role of fascia. Fascia is dense irregular connective tissue that allows for continuity throughout the body.^{18,19,20,21} Not only does it envelop muscles, fascicles, fibers, tendons, but it penetrates and surrounds organs, bones and nerves.^{18,19,20,21,22} Fascia is primarily composed of fibroblasts, mast cells and macrophages while the extracellular matrix is made up of ground substances (dense gel), collagen and elastin fibers.^{18,19,20,21,22} Contractile cells have also been identified in fascia and have been termed myofibroblasts.^{20,21,22,23,24,25,26} Ruffini organs, a type of mechanoreceptor found in fascia, have been shown to respond to deep pressure.^{23,24,27,28} Once ruffini organs are stimulated, altered input is transmitted to the central nervous system,

which changes the signal received by the motor unit creating tonus change in the muscle.^{23,24, 27,28} Perhaps this is an explanation for the therapeutic effect of ART[®], since the protocol involves the practitioner applying pressure to the muscle. Another proposed mechanism rationalizing the role of ART[®] can be extrapolated from the work of Cottingham in 1985 and Schleip in 2003.^{20,21,22,29} Golgi receptors, which are a sensory ending, are traditionally found in tendons, but are also found in the muscular portion of the myotendinous junction, ligaments, capsules, fascia and aponeuroses.²⁹ Golgi receptors are arranged in series in fascia and have been found to respond to muscular contraction/tension.^{20,21,22,30} Afferent signals sent from golgi receptors in response to contraction/tension, reach interneurons in the spinal cord, which, in turn, have an inhibitory effect on alpha-motor neurons thereby relaxing muscle.^{20,21,27,30} Perhaps ART[®] stimulates the golgi receptors since the protocol involves the patient actively shortening/contracting followed by actively stretching/

tioning the muscle. It should be noted that the suggested explanations regarding the mechanism of ART[®] are theories and more research is needed in the future to fully understand the mechanism.

Conclusion

Pudendal neuralgia by pudendal nerve entrapment is a rare diagnosis, but it is one that causes extreme pain and limitation in suffering individuals. Patients may be enduring this injury for 2–10 years before the diagnosis is made and finally receive appropriate treatment. Pudendal neuralgia has a varied clinical presentation due to its intricate anatomical structure, which may confuse practitioners. Therefore, it is important to be aware of this condition to avoid delayed and/or misdiagnosis and provide prompt treatment.

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