Anatomical Basis of Chronic Pelvic Pain Syndrome: the Ischial Spine and Pudendal Nerve Entrapment

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SUMMARY

Antolak SJ Jr, Hough DM, Pawlina W, Spinner RJ: Anatomical basis of chronic pelvic pain syndrome: the ischial spine and pudendal nerve entrapment.

Chronic pelvic pain syndrome is a conundrum that may be explained partly by pudendal nerve entrapment (PNE), which causes neuropathic pain. In men with PNE, aberrant development and subsequent malpositioning of the ischial spine appear to be associated with athletic activities during their youth. The changes occur during the period of development and ossification of the spinous process of the ischium.

INTRODUCTION

Chronic perineal pain may be caused by pudendal nerve entrapment (PNE). Patients with PNE typically present with pain in the penis, scrotum, labia, perineum, or anorectal region that is aggravated by sitting, relieved by standing, and absent when recumbent or when sitting on a toilet seat. PNE is a clinical diagnosis of conditions in patients with the typical history. Robert et al. (1) and Shafik (2) described how the pudendal nerve is entrapped between the sacrotuberous (ST) and sacrospinous (SSp) ligaments and may engage the falciform process of the ST ligament. Stretching of the pudendal nerve from chronic constipation causes neuropathy (3). Normal vaginal delivery causes measurable neuropathy lasting approximately 3 months (4).

Prostatitis-like urogenital pain and voiding and sexual dysfunction are the hallmark of pudendal neuropathy. Symptoms of prostatitis-like pain occur in 11% of American men, and approximately 95% of the men whose conditions are diagnosed as chronic prostatitis have no evidence of bacterial infection or inflammatory cells in the prostatic fluid (5).

Misdiagnosis begets expensive testing, serious misapplication of surgical procedures, and prolongation of nerve trauma. Treatment response is better in men who receive an early diagnosis and who meticulously avoid traumatic activities. In our practice, many of these men have PNE. At surgical treatment of PNE, Robert noted that 11% of patients required excision of an elongated ischial spine that was interpreted by the surgeon as conflicting with the pudendal nerve. Frequently, in male patients operated on for pudendal neuropathy at Mayo Clinic, the ischial spine is elongated with a posterior orientation,

occasionally shaped like a scimitar. In men, the typical ischial spine is a low-lying spike. Magnetic resonance imaging (MRI) studies of the pudendal nerve, especially at the ischial spine, have been attempted at Mayo Clinic, but MRI is not adequate for diagnosis of PNE. Mayo Clinic radiologists have reviewed MRI scans from other institutions and cannot confirm purported pudendal nerve compression (Maus T: Personal communication, June 2001). Our paper discusses the clinical syndrome of chronic pelvic pain syndrome, how it may be explained by PNE in men, and the etiology and suspected pathophysiology.

CLINICAL EVALUATION AND OBSERVATION

Men with urogenital pain or rectal pain or both, with or without voiding symptoms and with or with out ejaculatory pain, may have chronic pelvic pain syndrome. Prostatitis (reproductive tract infection) is ruled out by the absence of infection or inflammatory cells in the prostatic secretions (voided bladder 3 urinalysis, as described by Meares and Stamey) and the seminal fluid. These patients have National Institutes of Health (NIH) category IIIB chronic prostatitis and chronic pelvic pain syndrome (6) (Table 1).

Within our patient population, we have identified 5 symptom groups:

I. Short-term pain, preceded by voiding complaints; usual onset is after 2 to 5 months of identifiable trauma.

II. Insidious, long-standing symptoms with many past consultations and treatments or surgical procedures.

III. Sudden onset of pain while squatting and lifting. Often this pain precipitates a visit to an emergency department, but no pathologic features are identified (the pain is often treated, however, as epididymitis or testicular torsion).

IV. Pain after pelvic radiation therapy, typically for carcinoma of the prostate. (This group is estimated to be less than 1% of all men with urogenital pain.)

V. Pain from inflammatory processes after perineal surgery or drainage of phlegmon. (This group is estimated to be less than 1% of all men with urogenital pain.)

ANATOMY AND PATHOPHYSIOLOGY OF EXERCISE-INDUCED REPETITIVE MICROTRAUMA

The striking common feature in all patients is that flexion activities of the hip (sitting, climbing, squatting, cycling, and exercising) induce or aggravate urogenital pain, chronic pelvic pain, or prostatitis-like pain. Many of the men played American football, lifted weights, and wrestled as teenagers and young adults.

Our primary hypothesis is that hypertrophy of the muscles of the pelvic floor during the years of youthful athleticism causes elongation and posterior remodeling of the ischial spine. The SSp ligament then rotates, causing the ST and SSp ligaments to overlie one another. The ligaments act like a lobster claw, with the pudendal nerve traversing the interligamentous space where it can be crushed (Robert RR: Personal communication, March 2000). In addition, in this position the pudendal nerve travels a longer course because it is posterior or dorsal to the SSp ligament. In this course it may stretch over the SSp ligament or the ischial spine during squatting or during sitting or standing from a seated position. We surmise that the gluteus muscle, intimately attached to the ST

ligament, exerts a shearing effect as it extends the hip while the pelvic floor is forced inferiorly during the Valsalva maneuver.

The greater sciatic notch provides egress for the piriformis muscle. The pudendal nerve exits the pelvis at the inferior aspect of this muscle. In the athlete, flexion and abduction of the thigh are common motions, and they may lead to hypertrophy of the piriformis muscle. If the sciatic notch is narrowed because of the posterior orientation of the ischial spine, the cross-sectional area of the greater sciatic notch is reduced. Concomitant hypertrophy of the piriformis muscle may cause compression of the pudendal nerve against the posterior edge of the SSp ligament. Pain that suggests this process includes the pain that is induced during sports activity such as that of a baseball catcher (squatting and rising to throw the ball—motions that require extension of the gluteus muscle and abduction and extension of the hip). Another graphic example is a Canadian skater no longer able to turn (a motion requiring bending, squatting with 1 leg flexed while extending, rotating, abducting, and then adducting the crossing leg). Remodeling of the border of the sacrum, a broadening of that structure, also narrows the aperture of the greater sciatic notch (Fig. 1).

ISCHIAL SPINE: ONTOGENY, DEVELOPMENT, AND PATHOLOGY

The ischial spine is absent in quadrupeds, and it is largest in hominids (7). The progression to upright posture in bipeds requires increasing development of the musculature of the pelvic floor to assist the sphincters and to prevent evisceration in the erect position. All pelvic floor muscles have a direct or indirect attachment to the ischial spine: 1) The SSp ligament has muscular attachments. 2) The tendinous arch rises from

the ischial spine, traverses the fascia of the obturator internus, and attaches to the pubis. 3) The coccygeus muscle may affect the posterior remodeling of the ischial spine during development. The enormous forces required to appose the 2 sides of the pelvic floor during the Valsalva maneuver could well lead to malformation and malpositioning of the ischial spine with dire outcomes leading to PNE syndrome.

The ischial spine develops from a separate ossification center that arises between the ages of 13 and 15 years (8). Ossification is complete between the ages of 23 and 25 years. This interval of development is when youth are involved with athletic pursuits that induce hypertrophy of the pelvic floor muscles and the extenders and rotators of the hip. Palpation of levator ani muscles during digital rectal examination in young athletes reveals thick musculature. (Note that hypertrophy of the pelvic floor muscles can be measured with use of ultrasonography in women performing Kegel exercises.) Thus, in young athletes anatomical changes are established for future chronic pelvic pain syndrome.

These anatomical changes can be evaluated by use of various methods. Judet views of the pelvis highlight the ischial spine and the greater sciatic notch. In symptomatic patients with PNE (chronic pelvic pain syndrome and NIH Category IIIB chronic prostatitis), Judet views of the pelvis show interesting variations. Normative data are being obtained for comparison with these variations. For example, the diameter of the sciatic notch has been measured. The Thoms method is a classic means of pelvimetry in women, and comparative studies in men are available (9). Review of the bony structure of the male pelvis in teaching specimens from medical school anatomy laboratories shows a wide variation (age and weight of decedents are not available) (Pawlina W:

Personal communication, May 2001). This variation is consistent with the wide variation of bony pelvic measurements in published literature (9).

Imaging techniques that permit 3-dimensional reconstruction of the pelvis have been used to evaluate pudendal artery abnormalities (10). With 3-dimensional reconstruction, one may be able to measure the positional variation of the ischial spine over the ischial tuberosity and the distance between the ischial spine and the sacrococcygeal notch.

The radiographs in Figures 1 through 3 are representative of those of many of the patients in our study group. Figure 1 is a radiograph of the pelvis of a man with unilateral symptoms of PNE. Note the "normal" ischial on the right and the elongated, broad ischial spine on the symptomatic left side. The radiograph in Figure 2 is from a patient with exstrophy of the bladder and diastasis of the pubic rami who presented with severe symptoms of pelvic pain due to malignancy.

Significant abnormalities of each ischial spine relate to the requirements of pelvic floor musculature to respond to congenital or acquired deformities, resulting in the remodeling of the bones in the absence of athletic activities. For example, a businessman presented with 12 years of severe penile pain originally induced by straining during bowel movements. He had a history of 55 years of chronic constipation. A radiograph of his pelvis (Fig. 3) shows broad, stubby ischial spines similar to those in the man with exstrophy of the bladder (Fig. 2).

A female dancer and dance instructor, age 73 years, had severe pain after a 1,300-km automobile trip. She has abnormal ischial spines (not illustrated) similar to those in

Figure 3, which are thought to be associated with repetitive flexion and extension of the hip joint and associated pelvic floor contraction required in her artistic endeavors.

CONCLUSIONS

Chronic pelvic pain is often caused by a compression neuropathy of the pudendal nerve. The bony remodeling as a result of the activity of pelvic floor muscles leads to juxtaposition of the SSp and ST ligaments which compress the pudendal nerve in the narrowed interligamentous space. Elongation of the ischial spine in response to the same muscular forces presents an additional site for repetitive microtrauma of the pudendal nerve.

Future attention must be paid to 1) the transverse diameter of the ST and SP ligaments which compress the pudendal nerve 2) the dimensions of the greater sciatic notch (diameter and depth), correlated to age, weight, and body habitus; 3) the cross-sectional area of the greater sciatic notch and the piriformis muscle; and 4) sequential pelvis x-rays in youthful and maturing athletes to measure changes in position and appearance of the ischial spine.

Development of this information should aid in the definition and proper treatment of chronic pelvic pain syndrome, which affects a significant percentage of men, is a medical economics drain, and is a personal tragedy with occasional fatal outcomes due to suicide. Chronic pelvic pain syndrome in women should be studied in a similar manner. Application of the same principles in women is of paramount importance lest vulvodynia and other syndromes continue to be misdiagnosed and inappropriately treated.

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Table 1.—National Institutes of Health Classification System for Prostatitis

Type	Classification	Definition
I	Acute bacterial prostatitis	Evidence of acute bacterial infection
II	Chronic bacterial prostatitis	Evidence of recurrent bacterial infection
III	Chronic abacterial prostatitis (chronic pelvic pain syndrome)	No demonstrable evidence of infection
	A Inflammatory	White blood cells in semen*
	B Noninflammatory	No white blood cells in semen*
IV	Asymptomatic inflammatory prostatitis	No symptoms†

*Samples were from expressed prostatic secretions or from voided bladder 3 urine samples (sediment from initial 10 mL of urine after prostate massage during Meares-Stamey 4-glass test).

†Incidental diagnosis was made from the prostate biopsy or from the presence of white blood cells in prostatic secretions during evaluation for other disorders.

LEGENDS

Fig. 1. A 45-year-old man with scrotal and crural pain on the left side for 39 months. Left epididymectomy and subsequent excision of left hydrocele provided no relief. Usually in anteroposterior radiographs of the android pelvis, the ischial spine is not visible. On the right side, the spine is normal (*straight arrow*). On the left side, the spinous process is heightened, broadened, and blunt (*curved arrow*). The sacrum is broad, and the aperture of the greater sciatic notch is quite small. In his youth, the patient wrestled, played American football and lifted weights.

Fig. 2. A 33-year-old man with malignant pudendal neuralgia. Exstrophy of the bladder and diastasis of the symphysis pubis are apparent. The abnormal pelvic floor musculature with this anomaly led to a broadened, heightened ischial spine (*curved arrow*). The aperture of the greater sciatic notch is narrow (*2-headed arrow*). The piriformis muscle may have a direct compressive effect on the pudendal nerve.

Fig. 3. A 55-year-old man had constipation since childhood that required hours of straining and forcing of stool. He had a 12-year history of burning penile pain. Also, he had suprapubic and inguinal discomfort radiating to the scrotum and left testis. Allodynia was present, and hypalgesia was present throughout the distribution of the pudendal nerve. Compression of the pudendal nerve medial to the ischial spine reproduced subjective pain. Over the mid coccygeal area, there was an 8-cm-diameter area of peau d'orange with hypalgesia consistent with chronic regional pain syndrome. The ischial spine is heightened, broadened, and blunt.