Turk J Phys Med Rehab 2019;65(1):80-83
DOI: 10.5606/tftrd.2019.2863

Case Report

A rare and late presentation of pudendal neuralgia in a patient with fibromyalgia after pilates exercises

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Received: March 07, 2018 **Accepted:** September 03, 2018 **Published online:** January 30, 2019

ABSTRACT

A 32-year-old female patient was admitted to our clinic about two years ago with complaints of widespread pain, tiredness, fatigue, and insomnia for six months. She was diagnosed with fibromyalgia and initiated medical treatment with the recommendation of aerobic exercises. Her medical history revealed widespread pain disappeared one month after exercising with localized pelvic pain. She had also pain and burning sensation while sitting and lying down. She suffered from severe pain during defecation. Based on her medical history and physical examination findings, the patient was diagnosed with pudendal neuralgia. The patient was put on medical treatment and, then, referred to the anesthesia department due to persistent pain. Computed tomography-guided transgluteal pudendal nerve block was applied by the anesthesiologist. Subsequently, pain disappeared. In conclusion, pudendal neuralgia should be considered in the differential diagnosis in patients with pelvic pain and burning sensation or following compelling exercises.

Keywords: Fibromyalgia, pilates, pudendal neuralgia.

Pudendal neuralgia is a painful, neuropathic condition involving the dermatoma of the pudendal nerve.[1] Firstly defined by Amarenco et al.,[2] this condition was described with a neuropathic pain emerging in the form of burning on the pudendal nerve distribution. The pain is localized on vulva, vagina, clitoris, perineum, and rectum in females and on testicles, perineum, scrotum glans penis and rectum in males.[2]

The pain is in the form of burning, tingling, and numbness as in neuropathic pain. Also, it can emerge in the form of hyperalgesia, allodynia, or paresthesia. Symptoms may appear suddenly or gradually increasing over time.[3-5] Pudendal nerve originates from the sacral plexus and involve the segments S2, S3, and S4. It is a mixed nerve which ensures the sensation of the sexual organs such as penis, scrotum, clitoris and labia, and innervates the muscles of the perineus and the pelvic floor. The nerve comes out of the greater sciatic foramen, and passes through the ischial spine, sacrospinous ligament, and sacrotuberous ligament.[6,7] Pudendal neuralgia develops after mechanical damage to the pudendal nerve, viral infections, and immunological processes.[8] The mechanical damage to the nerve can be also called as pudendal nerve entrapment. This entrapment may occur following the pelvic floor muscle spasm (levator ani or obturator internus), the pressure from the surrounding ligaments (sacrospinous, sacrotuberous), and the trauma of the scar tissue or surgery. For operated patients, the entrapment may result from the mesh or suture with direct nerve injury.[9,10] The main manifestation in almost all patients is that the hip flexion activities (i.e., sitting down, climbing, crouching, cycling, and exercising) lead to urogenital pain, chronic pelvic pain, or prostatitis-like pain.[11] The major components for diagnosis include the history and the physical examination method. The conditions which may mimic pudendal neuralgia should be excluded. In fibromyalgia, the presence of a central pain and a pain perception failure have been proven.

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Cite this article as:
Yıldırım MA, Gökşenoğlu G. A rare and late presentation of pudendal neuralgia in a patient with fibromyalgia after pilates exercises. Turk J Phys Med Rehab 2019;65(1):80-83.

This is also the reason for allodynia and hyperalgesia seen in these patients. Considering emerging signs and symptoms, fibromyalgia is considered to be relevant with neuropathic pain.[12]

Herein, we report a rare and late presentation of pudendal neuralgia in a patient with fibromyalgia after pilates exercises in the light of literature data.

CASE REPORT

A 32-year-old female patient was admitted to our clinic about two years ago with complaints widespread pain, tiredness, fatigue, and insomnia for six months. Based on her detailed medical history and physical examination findings, she was diagnosed with fibromyalgia. At the time of admission, her body weight was 43 kg with a height of 165 cm and she was a single teacher. She was initiated medical treatment with the recommendation of aerobic exercises. Treatment started with pregabalin 75 mg/day, and the dose was gradually increased to 300 mg at first and then to 450 mg; however, the dose was maintained as 75 mg (twice per day) due to the adverse effects. The patient could tolerate this dose and achieved pain relief. However, her pain started again about one year ago, and she suddenly discontinued the administration of pregabalin with an antidepressant. Later, she was prescribed pregabalin again intermittently. Although the patient was unable to spend time for exercise before, she started pilates exercises. During follow-up visits in the clinic, she reported reduced pain and continued a single dose of pregabalin per day. Her widespread pain disappeared one month after exercising. However, localized pelvic pain became apparent and she described a pain and burning sensation while sitting and lying down. She had also severe pain during defecation. Based on the examination of the gynecology and general surgery departments, her symptoms were non-specific and she was recommended a non-steroidal anti-inflammatory drug for pain relief. In our clinic, we increased the pregabalin dosage again (75 mg in the morning and 150 mg in the evening), considering that the pain experienced by the patient was related to somatic symptoms of fibromyalgia and she had benefited from pregabalin before. At the time of admission about one month after, her symptoms slightly improved with medical treatment. She was doing exercises for two to three hours, although she had difficulty in doing thigh stretching exercises. The range of motion of both hips was normal on physical examination; however, flexion and external rotation were painful. The patient was recommended pelvic magnetic resonance imaging (MRI); however, it was unable to be performed due to claustrophobia of the patient. Pelvic radiography showed normal results.

Based on her medical history and physical examination findings, the patient was diagnosed with pudendal neuralgia according to the Nantes criteria.[3] Pregabalin dose was increased to 150 mg in the morning and 150 mg in the evening, and she was recommended to do exercises for a certain period of time. She was, then, referred to the anesthesia department due to persistent pain. A written informed consent was obtained from the patient. Computed tomography-guided transgluteal pudendal nerve block was applied by the anesthesiologist. Subsequently, pain disappeared. The patient was advised that neuronal block could be applied again, if pain appeared. She was scheduled for follow-up with the recommendation of walking exercises.

DISCUSSION

The major components of the diagnosis of pudendal neuralgia include the medical history and physical examination findings. The conditions which may mimic pudendal neuralgia should be excluded. In the differential diagnosis, bladder syndrome, vulvodynia, levator myalgia, piriformis syndrome, coxodynia, cauda equina syndrome, neuralgias and inflammation of other nerves such as obturator, genitofemoral, or ilioinguinal nerves should be considered. Examination should start by examining the lesions on perineus, vulva, and vagina. Vulvodynia is frequently seen. Palpation and bimanual examination of the pelvic floor should be performed to eliminate vulvodynia. Palpation of the ischial spine or pudendal nerve precipitates a tingling sensation with paresthesia or pain commonly known as Tinel's sign. In addition to the levator muscles, piriformis and obturator internus and externus should be also evaluated carefully. Although pudendal neuralgia finding is often diagnosed late or incorrectly, its incidence in the population was found to be 1% in a study and the prevalence of pudendal neuralgia is higher in females than males.[13-15]

In the diagnosis of pudendal neuralgia, the Nantes criteria are used. *Accordingly, the five essential diagnostic criteria are as follows: (i)* pain in the anatomical territory of the pudendal nerve extending from the anus to the clitoris (or penis); *(ii)* worsened

by sitting; *(iii)* the patient is not woken at night by the pain; *(iv)* no objective sensory loss on clinical examination; and *(v)* positive anesthetic pudendal nerve block. *Exclusion criteria have been also proposed as follows:* purely coccygeal, gluteal, or hypogastric pain, exclusively paroxysmal pain, exclusive pruritus, presence of imaging abnormalities able to explain the symptoms.[3]

Our case met the all five of the diagnostic criteria with certain exclusion criteria. In a case presentation by Insola et al.,[16] fibrosis of obturator internus was detected on MRI, resulting in pudendal neuralgia. Similarly, we are in the opinion that the compelling exercises in our case was likely to result in edema or fibrosis of the obturator internus. In addition, in two case presentations by Lee et al.,[17] pain caused by the pudendal nerve compression reduced upon the detection of a ganglion cyst on MRI and aspirating the cyst.

In the first-line setting, conservative treatment is recommended for pudendal neuralgia. Various exercises can be applied to help reduce the pain severity during physiotherapy. Patients are provided with a home-based exercise regimen including relaxation training.[2,18] There are several drugs which are frequently used for medical treatment including gabapentin, pregabalin (bid, oral route), cyclobenzaprine, and tricyclic antidepressants. The drugs such as intravaginal diazepam (tablets of 5 mg tid per day) or compound fuses can be also administered.[19] Transperineal, transgluteal, and transrectal approaches are used for pudendal nerve block, and mainly a transvaginal approach is preferred in female cases. In case of the lack of responsiveness to the treatment, nerve decompression surgeries are recommended.[17]

In conclusion, pudendal neuralgia should be considered in the differential diagnosis for patients suffering from pain and burning sensation at the pelvic area following compelling exercises. For the treatment of fibromyalgia, stretching and hip flexion exercises having a high level of evidence should be recommended by gradual increases, particularly for patients who have never done any exercise before.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

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