

Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia caused by Sacroiliac Joint Dysfunction



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ABSTRACT

Introduction: Persistent genital arousal disorder/genito-pelvic dysesthesia (PGAD/GPD) consists of persistent or recurrent unwanted sensations of genital arousal that may include other types of genito-pelvic dysesthesia, which occur without concomitant sexual interest or thoughts. There are multiple triggering factors for PGAD/GPD.

Aim: To report the case of a 38-year-old woman with low back pain and PGAD/GPD triggered by sacroiliac joint dysfunction.

Methods: The medical data of the female patient with low back pain and PGAD/GPD were reviewed and analyzed.

Results: Resetting of the subluxated sacroiliac joint resulted in complete remission of the patient's symptoms.

Conclusion: Sacroiliac joint dysfunction may be a trigger for PGAD/GPD in some cases. Lack of relevant knowledge among patients and healthcare providers is the biggest challenge of the proper diagnosis and treatment of PGAD/GPD at present. **Zhang Y, Su L, Ge H, et al. Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia caused by Sacroiliac Joint Dysfunction. Sex Med 2022;10:100544.**

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Key Words: Persistent Genital Arousal Disorder/Genito-Pelvic Dysesthesia; Low Back Pain; Sacroiliac Joint Dysfunction; Nerve Entrapment

INTRODUCTION

Persistent genital arousal disorder/genito-pelvic dysesthesia (PGAD/GPD) is an under-recognized clinical entity that causes spontaneous genital arousal without sexual interest or thoughts.¹ The symptoms of PGAD/GPD can negatively affect patients' daily lives (eg, driving or working), and some patients even experience suicidal ideation.² Since it was first described by Leiblum and Nathan in 2001,³ reports about this disorder are being presented by clinicians worldwide. In 2019, the International Society for the Study of Women's Sexual Health (ISSWSH) issued

the first expert consensus for the diagnosis and treatment of PGAD/GPD.¹

Although the pathophysiology of PGAD/GPD remains unclear, the ISSWSH experts panel agrees that a complex combination of diverse biopsychosocial factors contribute to its development, maintenance, and exacerbation.¹ PGAD/GPD can be triggered by injuries (eg, vehicle accidents, falls, childbirth, and overstrain) that disrupt the neural pathway (commonly irritative) from the terminal receptors of the pelvic organs to the peripheral somatic and visceral sensory afferent branches, pudendal nerve, sacral spinal nerve roots, cauda equina, spinal cord, spinothalamic tract, and eventually the paracentral lobule in the somatosensory cortex of the brain.^{1,4}

The therapy approaches to PGAD/GPD are diverse, and there is limited information about the effectiveness of different treatments. Physical/manual therapy can help reduce muscle hypertonicity and edema at the pelvic floor and thus eliminate the corresponding nerve compression in some cases.⁵

We present here a case report of a 38-year-old woman who developed PGAD/GPD related to sacroiliac joint dysfunction. Her symptoms ultimately resolved by manual therapy. Institutional Review Board approval was obtained for

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this case report and the patient provided a written informed consent.

CASE REPORT

A 38-year-old woman presented to our clinic on September 7, 2021 with the complaint of low back pain and “agonizing physiological responses” (quoting the patient). She had previously sought help from orthopedic surgeons, neurosurgeons, neurologists, gynecologists, and psychologists of different hospitals. Lumbar disc protrusion, piriformis syndrome, restless leg syndrome (RLS), anxiety, and dissociative disorder were proposed by previous clinicians, and corresponding therapy approaches had been performed, but the results were not effective.

The patient reported that she first experienced low back pain approximately six months ago, as she was getting up from a squatting position. She visited the orthopedic clinic of a local hospital on February 24, 2021, and magnetic resonance imaging of her lumbar vertebra was performed. The results demonstrated protrusion of the L4/L5 and L5/S1 discs and high-intensity zone at the posterior margin of the L5/S1 disc (Figure 1A). Lumbar discs protrusion and discogenic low back pain were diagnosed, and the patient was advised to rest and take melilotus extract tablets orally. Her symptoms continued to progress, and she began to experience soreness in the right buttock and muscle twitching in the right leg. At her second orthopedic clinic visit on March 3, 2021, she was diagnosed with piriformis syndrome, and physical therapy and nonsteroidal anti-inflammatory drugs were prescribed.

Owing to limited improvement in six months, the patient was referred to neurosurgeons (March 17, 2021), neurologists (March 18, 2021), gynecologists (April 9, 2021), and psychologists (July 13, 2021) for further therapy. The diagnoses of RLS (by the neurologists), anxiety, and dissociative disorder (by the psychologists) were proposed, and gabapentin and estazolam were prescribed accordingly. Despite those treatments, the

symptoms continued to worsen, and she began to experience paroxysmal stimulation around the perineal area.

After a traditional Chinese chiropractic massage of the low back and buttocks approximately one week before presenting to our clinic, her symptoms worsened severely. The paroxysmal stimulation became persistent, and increased in intensity. The patient described it as “intense low back pain with a continuous agonizing physiological response.”

By careful inquiry, we clarified the term “continuous agonizing physiological response” and recorded the characteristics of the patient’s symptoms as follows: (1) continuous stimulation around the clitoris, labia, vagina, and anus, making the patient feel like she was on the verge of orgasm or reaching orgasm; (2) the symptoms sometimes prohibited the patient from sleeping all night; (3) long periods of sitting, standing, and walking could worsen the symptoms; (4) the symptoms could not be alleviated by masturbation; (5) the symptoms were not associated with any subjective sexual desire; and (6) there were associated problems including low back pain and leg weakness. These complaints just meet the diagnosis criteria of PGAD/GPD proposed by the ISSWSH expert consensus.¹

Physical examination with the patient lying supine and prone on the examination table revealed that (1) her left leg was approximately 1 cm shorter than the right (comparing the height of bilateral medial malleolus); (2) her left anterior superior iliac spine was higher than the right; (3) her left iliac crest was higher than the right; (4) her left posterior superior iliac spine was lower than the right; (5) there was deeper tenderness in the areas around the right sacroiliac joint than that around the left; and (6) tenderness was noted over the symphysis pubis. Blood tests of complete blood count, erythrocyte sedimentation rate, and rheumatoid factor were performed and the results revealed no abnormalities. A pelvic radiograph demonstrated that the bilateral pubes were uneven, and the sacrococcygeal bone tilted to the right (Figure 1B).

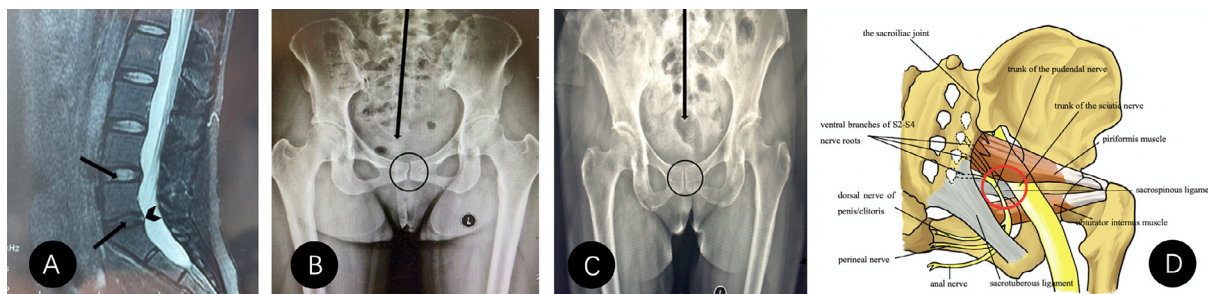


Figure 1. (A) Magnetic resonance imaging of the patient’s lumbar intervertebral discs demonstrating protrusion of the L4/L5 and L5/S1 discs (arrows) and high-intensity zone at the posterior margin of the L5/S1 disc (fork tail arrow). (B) Pelvic radiograph of the patient (pre-therapy) demonstrating that the bilateral pubes are uneven (circle), and the sacrococcygeal bone tilts to the right (long arrow). (C) Pelvic radiograph of the patient (post-therapy) demonstrating that the bilateral pubes are even (circle), and the sacrococcygeal bone is centered (long arrow). (D) Schematic illustration of the course and branches of the pudendal nerve in the pelvic cavity, and the surrounding muscles, ligaments, and nerves. The red circle represents where the entrapment most likely occurred in this case.

Combining the symptoms, medical history, physical signs, and findings from supplementary examinations, we suspected that the patient was experiencing PGAD/GPD triggered by sacroiliac joint dysfunction. The manipulations from Discovery of Posture Secret (DPS) were adopted to reset the subluxated right sacroiliac joint, which was causing the pelvic tilt. The key contents of the DPS technique are the 4R principles: resetting joint malalignment (RJM), resetting abnormal muscles (RAM), resetting joint stabilization (RJS), and resetting sensory motor control (RSMC).⁶ In the first treatment, RJM was obtained by forward to backward manual rotating of the right ilium, and the patient reported that the symptoms were significantly alleviated. RJM and RAM were obtained after three additional treatments (with three days between treatments), in which the patient's spasmed and displaced gluteal and pelvic floor muscles and ligaments were relaxed and strengthened by manual deep massage and resistance training. Finally, RSMC was obtained via rehabilitation exercises, which were performed to restore the patient's impaired proprioceptive input, and her symptoms resolved completely. A second radiograph revealed a normal pelvis (Figure 1C).

The patient was asked to revisit our department every month for follow-up after discharge; 6 appointments have been completed to date, and no recurrence has been reported.

DISCUSSION

The trunk of the pudendal nerve, a branch of the sacral plexus, leaves the pelvis from the lower edge of the piriformis muscle, and then returns to the pelvic cavity around the posterior part of the sciatic spine.⁷ Through the pudendal canal, the nerve reaches the ischial-rectal fossa and divides into the perineal nerve (which distributes to the muscle of the perineum and skin of the scrotum or labia majora), the anal nerve (which distributes to the sphincter of the anus and the skin around the anus), and the dorsal nerve of the penis/clitoris (distributing to the skin of the penis/clitoris).⁷ Any entrapment along the nerve pathway can cause corresponding irritation/inhibition symptoms (Figure 1D).⁷⁻⁹

Sacroiliac joint dysfunction is a common cause of low back pain, which usually centers around the unilateral/bilateral sacroiliac joint, but the accompanying symptoms can be complicated, depending on the adjacent structures involved.¹⁰ The sacroiliac joints are a pair of auricular-shaped, diarthrodial joints located between the sacrum and ilium bones, linking them together and completing the pelvic ring. In addition to the bony joint structures, there are many ligaments, such as the sacroiliac, sacrotuberous, and sacrospinous ligaments, that help stabilize the largest axial joints of the body. In certain settings, such as direct trauma to the pelvic ring, pregnancy and delivery, chronic strain, inappropriate posture/exercise, and systematic diseases, the joints can be dislocated and sacroiliac joint dysfunction (subluxation of sacroiliac joint) occurs.¹⁰ Long-term sacroiliac joint dysfunction can lead to uneven load on the pelvic ring and cause pelvic tilt,

which can lead to injury of the surrounding muscles, ligaments, and nerves.

In the present case, we speculated that the patient's PGAD/GPD developed because of sacroiliac joint dysfunction, pelvic tilt, displacement and spasm of the gluteal and pelvic floor muscles and ligaments, and eventually pudendal nerve entrapment. Given that the patient's symptoms involved the clitoris, labia, vagina, anus, and right leg, we suspected that the trunks of the pudendal and sciatic nerves were entrapped (red circle in Figure 1D).

The DPS is a manual therapy technique featuring 4R principles, which is effective in reducing musculoskeletal pain and correcting abnormal postures.⁶ In the treatment of this patient, we followed the 4R principles with effective results.

We believe that the under-recognition of PGAD/GPD was the reason for the patient's extended process of seeking medical help. Given the fact that there is no relevant literature on PGAD/GPD in Chinese medical databases, such as the China National Knowledge Infrastructure, Wanfang Data Knowledge Service Platform, and VIP databases, we speculate that this disorder is completely unknown to Chinese healthcare providers, let alone the general public. Multiple misdiagnoses were the result of under-recognition in the present case.

There are several limitations of this case report. First, there is no imaging examination to directly show the subluxation of the sacroiliac joint. Second, the position where the pudendal and sciatic nerves were entrapped was speculated, but not confirmed via tests. Third, no assessment scale was used in the process of diagnosis.

CONCLUSIONS

This report demonstrates that sacroiliac joint dysfunction and pelvic tilt can trigger PGAD/GPD, and DPS can be a useful manual therapeutic option for restoring the misplaced joints, muscles, and ligaments in these cases. Greater emphasis should be placed on professional research, as well as public education among healthcare providers and the general public in China to raise awareness regarding this potentially debilitating disorder. Correct understanding is essential for correct treatment.

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STATEMENT OF AUTHORSHIP

Conceptualization, YXZ; Writing-Original Draft, YXZ, LS, HG; Writing-Review and Editing, QW.

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