Case Report

Priapism associated with lumbar stenosis: case report and literature review

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Abstract: Lumbar spinal stenosis (LSS) is characterized by narrowing of the central canal, lateral recesses, or foramina leading to compression of neural tissue. The clinical syndrome associated with LSS is usually neurogenic claudication, which often presents as lower back and extremity pain, numbness, and tingling upon ambulation. Autonomic dysfunction is rarely observed in patients with LSS; however, a subset of male patients has been reported to experience intermittent priapism associated with the onset of neurogenic claudication symptoms. We present the case of a 33-year-old male who was diagnosed with LSS associated with neurogenic claudication and priapism who underwent minimally invasive decompressive surgery. Complete resolution of claudication and priapism was observed at the 6-week follow-up visit. This case report highlights minimally invasive lumbar decompression as an effective treatment for the rarely observed presentation of priapism associated with LSS.

Keywords: Lumbar spinal stenosis (LSS); priapism; neurogenic claudication

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Introduction

Lumbar spinal stenosis (LSS) is characterized by narrowing of the central canal, lateral recesses, or associated neural foramina (1). Unlike its more common degenerative counterpart, primary LSS is congenital in origin with short pedicles predisposing patients to central canal narrowing (2,3). As a result, patients with primary LSS have an onset of symptoms at an early age and present without degenerative changes. Acquired LSS is the most common etiological form and is the direct result of spondylotic changes such as disc degeneration, disc herniation, facet osteophyte formation, and ligamentous hypertrophy (4). Less common etiological forms of LSS include iatrogenic, post-traumatic, or secondary to endocrinopathies or skeletal diseases such as Cushing's syndrome or Paget's disease. The clinical syndrome associated with LSS is usually neurogenic claudication, which often presents as lower back and extremity pain, numbness, and tingling (1). Priapism, defined as a prolonged and undesired erection in the absence of sexual stimulation, is a rare finding in the context of LSS. Herein, we describe a case of LSS with neurogenic claudication and intermittent priapism, which were completely resolved following minimally invasive surgery.

Case presentation

History and physical

A 33-year-old male experienced onset of lower back pain and neurogenic claudication symptoms in 2013. The patient was evaluated at an outside clinic and diagnosed with degenerative joint disease. At that time, he noted back pain rated as a 7/10 with lower extremity tingling, weakness, and numbness. He denied bladder and bowel incontinence. The treatment regimen was conservative, consisting of analgesics, nonsteroidal anti-inflammatory drugs (NSAIDs), and physical therapy. Conservative therapies reduced pain to a 5/10 rating; however, the patient still required multiple leaves of absence from his occupation.

The patient presented to the senior author (LAT) for progressively worsening symptoms in 2018. His lower

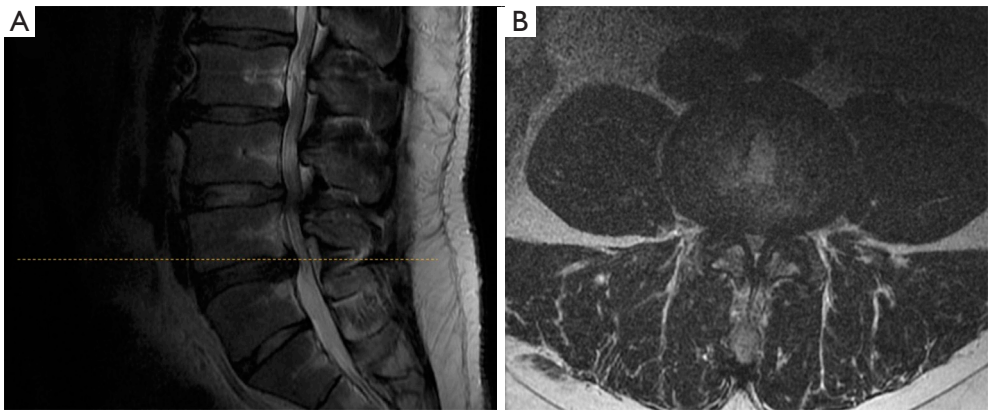

Figure 1 Preoperative MRI of the lumbar spine. (A) Sagittal MRI of the lumbar spine showing congenital lumbar stenosis with a narrowed spinal canal and multiple levels of the severe stenosis most notable at L2–3, L3–4, and L4–5; (B) axial MRI showing the most stenotic level, L3–4.

extremity pain radiated to both feet and was exacerbated by both standing and walking approximately 50 feet. Associated symptoms such as numbness and tingling persisted and remained unchanged. Furthermore, the patient noted uncontrolled erections while walking, which almost always occurred in conjunction with his worsening bilateral leg numbness. The patient exhibited slight gait instability but denied falls. The physical exam revealed normal muscle bulk and tone without weakness. MRI of his lumbar spine revealed severe stenosis at L2–3 and L3–4, as well as a L4–5 disc herniation (*Figure 1*). The patient's presentation was consistent with a diagnosis of LSS with neurogenic claudication. Given the progressive nature of symptoms, the patient elected to have decompression surgery.

Of note, the differential for priapism consists of, but is not limited to, blood disorders, adverse drug reactions, and pelvic trauma—all of which were of low clinical suspicion for this patient. Extensive urological evaluation was avoided given that episodes of priapism were not notable for penile pain and self-resolved within minutes following cessation of ambulation. These factors offered reassurance that the risk of long-term urologic damage was low.

Operative and postoperative course

After successfully intubation, the patient was placed in the prone position on a Jackson table using Wilson frame. The lumbar area was prepped and draped in the usual fashion. A 2 cm midline incision was made at L2–3; sequential dilators were used to gain access to the right hemi-lamina and an 18 mm tube was used to maintain exposure. The microscope was brought into the field and a right hemi-laminectomy was performed with high-speed burr. Bilateral decompression was achieved via the unilateral hemi-laminectomy using high-speed burr at right L2–3 by under-cutting the lamina on the left side. At this level, the ligamentum flavum, which demonstrated substantial hypertrophy, was removed using curettes and Kerrison rongeurs. Decompression was confirmed with Woodson probe. The same steps were repeated at the L3–4 and L4–5 levels to achieve decompression. At L4–5, a microdiscectomy was also performed to remove the herniated disc fragment. The wound was thoroughly irrigated and closed with vicryl sutures and skin glue. The patient was successfully extubated then transferred to PACU in stable condition.

Surgical and pathologic findings confirmed admission diagnosis of LSS with neurogenic claudication. The post-operative course was uncomplicated. The patient's pain was well controlled. The patient was discharged on post-operative day (POD) 1. At his 6-week follow-up visit, the patient revealed complete resolution of spontaneous priapism and postural instability. Furthermore, severity of claudication symptoms was markedly decreased. During this visit, the senior author requested and received verbal consent from the patient to publish these findings.

Discussion

LSS is one of the most commonly diagnosed spinal conditions and affects more than 200,000 people in the United States (5,6). Although symptoms and neurologic

Table 1 Reports of priapism associated with LSS

Author	Year	Age	Clinical presentation	Surgery	Outcome
Brish (11)	1964	36	Painful lower extremity paresthesia and weakness with spontaneous priapism. Micturition was difficult to initiate however sexual function remained intact	L2–5 laminectomy	Complete resolution of priapism, paresthesia, walking intolerance, and urinary disturbance. Sensory deficits remained unchanged at 6-week follow-up
Ravindran (12)	1979	61	Lower extremity numbness with spontaneous priapism provoked by 200 yards of walking. Symptoms resolved with rest	L4–5 disc excision and foraminotomy	Complete resolution of claudication and priapism
Maurice-Williams (13)	1985	60	Priapism and paresthesia radiating to buttock and genital area precipitated by walking 300 yards. Patient experienced relief of symptoms within 4 minutes after resting. Sexual performance and bladder function were unaffected	L3–5 laminectomy	Complete resolution of symptoms within 6 weeks
Ram (14)	1987	70	Spontaneous priapism, painful claudication, weakness, and numbness in lower extremities weakness after walking 200 meters. All symptoms would resolve with rest	L2–4 laminectomy and facetectomy; L2–3 and L3–4 discectomy	Complete resolution of claudication symptoms and priapism
Tubbs (15)	2005	15	Lower extremity weakness and priapism provoked by walking short distances. Relief of symptoms with lumber flexion	T8–L5 laminectomy with T7–S1 thoracolumbar fusion	Complete resolution of symptoms at 12-month follow-up
Cansever (2)	2007	74	Priapism, back pain, paresthesia, and sensory deficits	L4 partial hemilaminectomy with medial facetectomies and L4–5 discectomy with posterior interbody fusion	Complete resolution of symptoms at 6-month follow-up

First author, age of patient, symptoms associated with stenosis are noted. Summary of surgical intervention and postoperative outcomes are outlined; LSS, lumbar spinal stenosis.

findings vary, LSS is characterized by lower back and lower extremity pain, paresthesia, numbness, and weakness. Foraminal or lateral recess stenosis is associated with unilateral radiculopathy while central canal stenosis usually presents with bilateral symptoms (7,8). Neurogenic claudication is the cardinal sign of LSS, which is characterized as the posture-dependent progressive onset of lower extremity pain, paresthesia, and numbness while standing, walking, or performing any activity that requires extension of the lumbar spine. Weakness, manifesting as foot drop or knee buckling, can be experienced before the onset of pain (9). Patients will often relieve the debilitating pain by assuming a stooped posture, which increases central canal or lateral recess space and subsequently decreases pressure on neural tissue and vasculature (10).

Autonomic dysfunction is rarely observed in patients with LSS; however, a subset of male patients has been reported to experience intermittent priapism associated with the onset of neurogenic claudication symptoms (*Table 1*). Priapism is defined as an erection in the absence of sexual arousal and/or stimulation. An erection in a healthy patient is mediated by a reflex mechanism from the S2–S4 segments of the cord. Tactile stimuli are transmitted to the cord via dorsal roots of S2–S4 (15). The reflex loop is completed

with parasympathetic input from the perineal branches of the pudendal nerve, arising from the same levels, which stimulates the shunting of arterial blood in the corpus cavernosa erectile tissue of the penis (12). Sympathetic stimuli from the hypogastric nerve trigger vasoconstriction within the corpus cavernosa resulting in penile relaxation. In patients with LSS and intermittent priapism, mechanical compression of the thecal sac is thought to increase parasympathetic reflex activity (16).

The combination of priapism and neurogenic claudication in the context of LSS has been observed in a broad range of ages, coexisting conditions, and LSS etiologies (11). Cansever *et al.* describes the case of a 74-year-old man who presented with LSS neurogenic claudication and intermittent priapism (2). L4 partial hemilaminectomy with medial facetectomies were performed and the patient noted complete resolution of all symptoms at the 6-month follow-up. Similarly, Maurice-Williams *et al.* reported complete resolution of symptoms following decompression surgery in patient who also presented with neurogenic claudication and intermittent priapism (13). Furthermore, a 15-year-old male with achondroplasia reported the onset of bilateral lower extremity weakness provoked by walking. He received relief within 10 minutes after assuming the fetal position. The patient underwent extensive thoracolumbar fusion from T7–S1 and was completely asymptomatic at the 12-month follow-up (15).

Baba *et al.* contributed the most comprehensive examination of LSS with intermittent priapism to date (16). They report the results of electrophysiological studies as well as imaging studies for seven patients with LSS and spontaneous priapism with an average follow-up time of 3.3 years. Priapism was provoked by a range of anatomical positions including walking, standings, and squatting while defecating in the lavatory. Furthermore, the patients exhibited variable exercise intolerance. Some became symptomatic after walking 50 meters while others noted the onset of priapism after walking 300 meters. MRI revealed various degrees of arachnoiditis in three patients, highlighting a possible pathological role of inflammation of the meninges in addition to mechanical compression of the thecal sac.

Herein, we report the case of a 33-year-old male who presented with neurogenic claudication and intermittent priapism caused by LSS. Priapism is a rare clinical finding associated with LSS; however, our case and others in the literature demonstrate the utility of decompressive surgery in the resolution of these symptoms.

Acknowledgments

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Informed consent was obtained from the patient for publication of this case report and any accompanying images.

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