

Piriformis syndrome: Still unsolved issues

Piriformis Syndrome (PS) is an example of extra-spinal sciatica due to compressive neuropathy and can also be named as 'deep gluteal syndrome', 'pelvic outlet syndrome' or 'pseudo-sciatica'. Its prevalence among Low Back Pain (LBP) sufferers ranged between 0.3% and 36%; though the condition is prevalent in women, men also get the disorder [1]. In PS, resultant localized gluteal and radiating LBP is due to a spasmodic Piriformis Muscle (PM) and compressed, irritated and stretched Sciatic Nerve (SN) behind the PM, respectively [1]. In clinical practice, sometimes regional gluteal pain predominates in PS, whereas in some patients, pain alike sciatica can be the presenting complaint confusing treating physician, leading to incorrect treatment of the patients. There are two types of PS – primary and secondary; in case of primary PS, the pathology is intrinsic to PM and or SN, however; in secondary PS, clinical features of associated disorders, like Lumbar Spinal Stenosis (LSS), Leg-Length Discrepancy (LLD), fibromyalgia (FMS), fatty wallet, etc., may complicate the clinical picture [2]. Although 70 years have elapsed after the 1st clinical description of PS, we are yet to define precisely the PS pathophysiology, etiology, unique clinical features, confirmed diagnostic tests, best treatment modality, natural history of the disease, etc. [3]. Published studies with Lower level of Evidence (LoE) address various aspects of PS and most of the study outcomes are inconclusive. So, we take the privilege of addressing areas of PS that might interest both clinicians and researchers in the field.

Who is prone to get PS?

Piriformis syndrome may affect people irrespective of age, sex and occupation [1]. However, the disorder is apparently more common in women than in men because of the wider pelvis and Q-angle [4]. It appears the condition affects young people less frequently [1]. Since the piriformis muscle is a deep seated structure, overuse of the lower back could pose deep gluteal musculatures including PM under sustained strain evoking clinical manifestations suggestive for PS. In our previous studies, we reported higher PS frequency among Bangladeshi housewives; however day laborers, drivers and bankers also had the disorder [1]. PS may cause problems in dancers [5]. Despite these studies still little is known about the prevalence of PS in men and women and more research is required.

What is the cause of piriformis syndrome?

It is very difficult to make conclusive remarks about PS causation, as few publications are addressing the question what may cause PS to develop. Based on previous LoE articles, overuse of PM may be one of the most important reasons, though most of the published works didn't precisely define PM overuse, rather hypothesized how it could be [1]. In our previous research, we explained how the PM could get overused; however the way of overusing PM was not the same in all PS cases. So it is important to define exactly the PM overuse pathophysiology causing PS. Anomalous sciatic nerve and / or PM could contribute to PS features, though this idea is not confirmed by a recent large-scale collaborative radiologic study by Bertret and colleagues who didn't find significant PS prevalence in anomalous sciatic nerves compared to normal [6]. Primary PS features could be due to spreading of myofascial trigger points within the PM. Direct gluteal trauma and a previous fall also may contribute to PS induction [1].

Sometimes PS is synonymously, though incorrectly used with wallet neuritis; in the latter condition patients usually complain about sciatica-like pain upon persistent gluteal compression due to an external fatty wallet and they rarely have deep seated regional gluteal pain and in most cases they are negative for PS maneuvers and most importantly they improve dramatically following radical 'walletectomy' [2]. However, both PS and wallet neuritis may simultaneously be present in a single patient [7]. Leg-Length Discrepancy (LLD) may be associated with PS. In LLD, the resultant asymmetry of pelvis poses lower lumbosacral segment, pelvis and deep gluteal structures including the PM under intense stretch contributing to PS features. According to the ACR 1990 classification criteria for Fibromyalgia syndrome (FMS), one of the 18 tender areas resides at the insertion of PM tendon to the posterior aspect of greater trochanter of the femur that could get irritated depending on the risk factors causing more gluteal pain or persistent regional gluteal pain. Because of inappropriately treated PS could induce the development of central sensitization which may result in generalized body ache, for example FMS [8,9].

The double crush hypothesis postulates that axons compressed at one site could also get compressed at another site and first came into

Md Abu Bakar Siddiq^{*1} & Johannes Jacobus Rasker²

¹Department of Physical Medicine and Rheumatology, Brahmanbaria Medical College, Bangladesh

²Department of Rheumatology, Faculty of Behavioral Sciences, University of Twente, The Netherlands

*Author for correspondence: abussiddiq37@yahoo.com

light by Upton and McComas in 1973. Based on this hypothesis higher prevalence of cervical radiculopathy is observed in patients with Carpal Tunnel Syndrome (CTS) than that of non-CTS [10]. Piriformis syndrome especially in association with LSS could be an example of 'double crush syndrome', as SN nerve roots and SN get compressed at lumbar spine and extra-spinal levels (behind PM), respectively; however requires further exploration.

Piriformis pyomyositis due to an invading infectious agent, such as *Staphylococcus aureus*, group A. *Streptococcus*, *B. melitensis* could cause severe gluteal pain with fever and require different treatment approach than usual for PS [11,12]. As of today, published papers regarding etiology and association of PS are scarce and mostly based on poor study samples, warranting more research.

What are the common diagnostic features of PS?

Regional and or radiating gluteal pain according to the sciatic nerve distribution is the mostly seen pattern in PS, often mimicking true lumbar spinal sciatica. In PS, gluteal pain may aggravate during longstanding sitting, especially on hard surface and is associated with tingling, numbness or burning sensations on the outer aspect of the ipsilateral leg. Pain also aggravates with walking, lying on the affected side, during attempted standing from sitting position or squatting, however, in chronic cases ambulation may provide some pain relief [1]. The most important peculiarity of PS diagnosis are its 'inconsistent clinical manifestations' and one of the most important reasons of why PS is underdiagnosed and remains undiagnosed for a long time; In some cases PS features are completely inseparable from those of spinal sciatica, especially during an acute attack, a reason why PS can be over-diagnosed [1]. In an excellent literature review regarding PS clinical manifestations, Hopayian and colleagues mentioned the following four features that are common in PS –

- Buttock pain,
- External tenderness over the greater sciatic notch,
- Aggravation of the pain through sitting and
- Augmentation of the pain with maneuvers that increase PM tension [3].

Gluteal atrophy can be a complication in chronic cases [1]. Even though SN gets compressed behind PM, SN deficit is not common in the disorder, but lower limbs weakness because of SN neuropathy could be seen in bilateral PS as reported in a case report by Moon et al. [13]. In

infective piriformis myositis fever is one of the most common presenting feature and unique for this purulent PS [11]. However, in finding more accurate diagnostic features we need to do more prospective, multicenter, longitudinal studies.

What are confirmed diagnostic tests for PS?

There is no single confirmatory test for PS diagnosis. It's a disorder of exclusion of other clinical mimickers, namely LSS, cyclical sciatica, FMS, deep seated gluteal abscess, wallet neuritis, pelvis tumor, Pott's disease, hip arthritis, superior cluneal nerve disorder, osteitis condensans illii, sacroiliitis, etc. [2,11-12,14]. Clinical maneuvers with variable sensitivity and specificity may identify whether PM is involved in the presenting gluteal pain; examples of these maneuvers are the Piriformis sign, Pace sign, FAIR (Flexion, Adduction, Internal Rotation) test, Freiberg test, and Beatty test. Digital per-rectal examination revealing pain, especially when the index finger glides over the affected side of the pelvic wall is suggestive of PS [1]. The positive Straight Leg Raising (SLR) test signifies lumbar nerve roots pathology and usually is negative in PS, however in acute PS, SLR could be positive, though based on lower LoE. Besides this clinical information, MRI/CT scanning of pelvis could approximate PM pathology and its extension. MRI also could exclude associated pelvis and lumbar spine pathology. Moreover, nerve conduction velocity and electromyogram of the affected gluteal region and ipsilateral lower limb can differentiate between myopathy and neuropathy - true spinal sciatica from extra-spinal sciatica [1,7]. Taheri et al. describe that the musculoskeletal ultrasonography has higher sensitivity in diagnosing hypoechoic myofascial trigger points and appears to be useful in diagnosing piriformis myofascial pain syndrome, however its reliability should be tested in a longitudinal study with large number study participants [15]. So in terms of confirmed diagnostic test for PS we are yet to have the most appropriate one.

Is PS life threatening?

It is very difficult to provide a straightforward answer of this question as studies addressing the fact are lacking. Most published works did not highlight this part of PS and rather mentioned it as a benign neuromuscular condition without any significant comorbid consequences, although it may be very painful and incapacitating. As of now, no published report document mortality issues associated with PS. However, recently, piriformis pyomyositis is being considered as a life threatening cause of PS and may be fatal when proper attention has not been given in terms of timely diagnosis and treatment [11,12]. In a

published case report Gaughan and colleagues hypothesized that transient intravenous cannula induced septicemia could complicate piriformis pyomyositis due to seeding of the infectious agent into the PM and generated gluteal pain with fever, treated successfully with antibiotics [11]. Jeon et al. documented PS features, gluteal atrophy and ipsilateral lower limb weakness due to radiotherapy induced scarring of the respective PM compressing SN vicinity in cervical cancer [16]. Lower limb weakness is reportedly possible in cases of bilateral PS [13]. However, all these study outcomes based on lower LoE and further research with large number of PS could provide further valuable information.

What are the most effective treatments for PS?

Before treating PS, it is of paramount importance to classify it. In case of primary PS, treatment should focus on intrinsic PM pathology only, however secondary PS deserves treatment for associated conditions. Commonly used pharmacological and non-pharmacological modalities that are found to be useful in PS: Non-Steroidal Anti-Inflammatory Drugs (NSAIDs), analgesics (including adjuvants), piriformis stretching exercise, activities of daily living modification; however, intra-lesional steroid-lidocaine combination and botulinum toxin-A injections are reportedly most effective in the disorder, but we are yet in the dark to know exactly how frequent and how long patients with PM should receive these sort of interventions, in order to experience complete pain relief [1]. PS secondary to PM infection doesn't suit above modalities, judicious selection of antibiotics is indicated a [11,12]. Sometimes exploration of PM for surgical drainage, PM tendon resection or sciatic nerve neurolysis appears appropriate. So, best treatment approach for PS depends on myriad factors and there is no rule of thumb, for each individual a personal approach should be applied [1].

In conclusion, piriformis syndrome is an example of pseudo-sciatica with varying clinical manifestations. Our current knowledge about its pathophysiology, epidemiology, causation, risks association, clinical presentations, diagnosis, treatment, and consequences is insufficient and most importantly they are based on inconclusive study outcomes. So, future researchers could explore them and could provide more consistent information concerning piriformis syndrome.

References

- Siddiq AB, Hossain S, Khasru MR *et al.* Piriformis syndrome: a case series of 31 Bangladeshi people. *Europ. J. Orthop. Surg. Traumatol.* 27(2), 193–203 (2016).
- Siddiq AB. Piriformis syndrome and wallet neuritis: Are They the same? *Cureus.* 10(5), e2606 (2018).
- Hopayian K, Song F, Riera R *et al.* The clinical features of the piriformis syndrome: a systemic review. *Eur. Spine. J.* 19(12), 2095–2101 (2010).
- Boyajian-O'Neil LA, McClain RL, Coleman MK *et al.* Diagnosis and management of piriformis syndrome: an osteopathic approach. *J. Am. Osteopath. Assoc.* 108(11), 657–664 (2008).
- Martinez N, Mandel S, Peterson JR. Neurologic causes of hip pain in dancers. *J. Dance. Med. Sci.* 15,157–159 (2011).
- Bartret AL, Beaulieu CF, Lutz AM. Is it painful to be different? Sciatic nerve anatomical variants on MRI and their relationship to piriformis syndrome. *Eur. Radiol.* 28(11), 4681 (2018).
- Siddiq AB, Jahan I, Alpha M. Wallet neuritis – An example of peripheral sensitization. *Curr. Rheum. Rev.* 13, 999 (2017).
- Wolfe F, Smythe HA, Yunus MB *et al.* The American College of Rheumatology 1990 criteria for the classification of fibromyalgia. Report of the Multicenter Criteria Committee. *Arthritis. Rheum.* 33, 160–172 (1990).
- Siddiq MA, Khasru MR, Rasker JJ. Piriformis syndrome in fibromyalgia- clinical diagnosis and successful treatment. *Case Rep. Rheumatol.* 2014, 893836 (2014).
- Schmid AB, Coppieters MW. The double crush syndrome revisited - A Delphi study to reveal current expert views on mechanisms underlying dual nerve disorders. *Manual. Therapy.* 16, 557–562 (2011).
- Gaughan E, Eogan M, Holohan M. Pyomyositis after vaginal delivery. *BMJ. Case Rep.* 2011, bcr0420114109 (2011).
- Kraniotis P, Marangos M, Lekkou A *et al.* Brucellosis presenting as piriformis myositis: a case report. *J. Med. Case. Rep.* 5(1), 125 (2011).
- Moon HB, Nam KY, Kwon BS *et al.* Leg weakness caused by bilateral piriformis syndrome: a case report. *Ann. Rehabil. Med.* 39(6), 1042–1046 (2015).
- Siddiq AB. Superior cluneal nerve disorder: how often do we consider? *J. Recent. Adv. Pain.* 2, 4–5 (2016).
- Taheri N, Okhovatian F, Rezasoltani A *et al.* Ultrasonography in diagnosis of myofascial pain syndrome and reliability of novel ultrasonic indexes of upper trapezius muscle. *Ortopedia. Traumatologia. Rehabilitacja.* 18(2), 149–154 (2016).
- Jeon SY, Moon HS, Jung Han YJ *et al.* Post-radiation piriformis syndrome in a cervical cancer patient -a case report. *Korean. J. Pain.* 23(1), 88–91 (2010).