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Case report and review of the potential role of the Type A piriformis muscle in dynamic sciatic nerve entrapment variant of piriformis syndrome

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Abstract
Piriformis syndrome (PS) is an underdiagnosed but common cause of chronic buttock pain and sciatica. Anatomical variants of the piriformis muscle and sciatic nerve have not been thought to be significant in the pathophysiology of PS however, recent description of the piriformis musculotendinous junction has identified a common variant that we believe frequently results in dynamic sciatic nerve entrapment at the infra-piriformis fossa. We performed ultrasound guided low-dose Botulinum Toxin-A (BTX-A) injection to the lower piriformis muscle belly in an elite Australian Rules football player with PS and Type A piriformis muscle to relieve symptomatic sciatic nerve compression. Positive response to targeted BTX-A piriformis muscle injections support the hypothesis that sciatic nerve compression by Type A piriformis muscles may contribute to the pathophysiology of neuropathic PS, along with other functional factors. Sciatic nerve compression due to Type A piriformis at the infra-piriformis fossa has not been described previously and is a potentially common cause of neuropathic PS, especially when combined with other functional factors such as piriformis muscle spasm/hypertrophy and sacroiliac joint counternutation.

Keywords Piriformis syndrome · Type A piriformis muscle · Sciatic neuritis · Botulinum Toxin-A

Introduction

Piriformis syndrome (PS) is a controversial and potentially common but underdiagnosed cause of buttock pain and 'non-discogenic' sciatica [2–5, 7, 11, 13, 18]. Recognition and diagnosis of PS has been affected by a number of factors, including controversy over what constitutes the clinical entity, its pathophysiology (particularly the role of variant piriformis and sciatic nerve anatomy), the absence of definitive diagnostic tests, and overlap with other causes of deep gluteal syndrome [2, 4, 5, 11, 13, 18].

Most of the medical literature regarding PS relates to refractory cases of PS requiring intervention, including injection therapy and surgery [5, 11, 15]. Whilst the aetiology is said to be unidentifiable in most cases, gluteal trauma, anatomical variants, myofascial syndrome, and piriformis spasm or hypertrophy have been commonly identified causes [11]. In the surgical and rehabilitation literature, the focus has primarily been on the anatomical relationship of the sciatic nerve to the piriformis muscle, and to a lesser extent, factors relating to the medial piriformis muscle including piriformis muscle belly hypertrophy, as the cause of symptoms [5, 11, 13, 15, 16, 20].

Beaton and Anson originally identified six anatomical variations between the piriformis muscle and sciatic nerve [1]. A larger review of multiple cadaveric studies of the piriformis muscle and sciatic nerve showed the most common relationship (Type I), seen in 83.1%, is the classically described undivided sciatic nerve passing below the piriformis muscle. The commonest variant to this relationship, seen in 13.7%, is a divided sciatic nerve with the common

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peroneal nerve passing through the piriformis muscle and the tibial nerve passing below the muscle. Other variants were considerably less common (1.3–0.08%). However, the authors cast doubt on the role of piriformis and sciatic nerve anomaly in the pathogenesis of PS, citing a similar prevalence of piriformis and sciatic nerve anomaly in PS patients and the normal population [17].

Despite the typical description of PS being a compression of the sciatic nerve at the infra-piriformis fossa, little consideration has been given to the lower piriformis muscle in the role of PS pathophysiology. In 2007, Windisch et al. more clearly defined the anatomy of the lateral piriformis muscle, including the musculotendinous junction and fusions with other pelvic girdle tendons, including the superior gemellus, obturator internus, and gluteus medius [20]. The commonest variant of the piriformis muscle musculotendinous junction occurred where the upper muscle belly had a greater distance from the musculotendinous junction to the insertion compared to the lower muscle belly (Type A), which was present in 63% of specimens. However, little was made of this finding by the authors, who noted that "it seems, that anatomical causes of piriformis syndrome are rare" [20].

Management of PS is centred around rehabilitation, which may be augmented by imaging guided injection of local anaesthetic and cortisone; in general, cases requiring surgical intervention, including nerve decompression and tenotomy of the piriformis muscle, are uncommon [5, 10, 11, 13]. More recently, botulinum toxin has also been utilised for the treatment of PS [6, 8, 9, 11, 21].

In this report we describe a case of PS in a young male Australian Rules footballer with Type A piriformis muscle morphology and compressive neuropathy of the sciatic nerve at imaging, who was treated with ultrasound (US) guided low-dose Botulinum Toxin-A (BTX-A) injection targeted to the lower belly of piriformis. Whilst the role of variant anatomy at the musculotendinous junction has been downplayed by the authors of the original paper describing the anatomy of the lateral piriformis muscle, we believe the Type A piriformis variant defines a common compression point, that along with co-existing functional factors may be a significant contributor to pathophysiology in dynamic sciatic nerve entrapment [4].

Case report

18-Year-old male right-footed State level Australian Rules football player with right buttock pain and referred below-knee right lower limb pain refractory to rehabilitation limiting training and game-day running volumes, including sprint distances, as well as kicking power and distance. Clinical assessment demonstrated reduced right passive straight leg raise by 25° compared to baseline and left leg, increased tone in the biceps femoris muscle, anterior pelvic tilt and increased right counternutation.

Initial lumbar spine MRI showed a chronic right L5 pars defect and disc degeneration at L4/5 without focal disc herniation. A clinical diagnosis of PS was made and piriformis rehabilitation was instituted including addressing functional pelvic and hamstring issues without substantial improvement. After failed rehabilitation, diagnostic US of the right buttock was performed which demonstrated a prominent lower piriformis muscle belly extending out of the greater sciatic notch, resulting in compression of the sciatic nerve against the posterior column of the acetabulum (Fig. 1a). US guided local anaesthetic and cortisone injection to the right piriformis muscle confirmed the clinical diagnosis

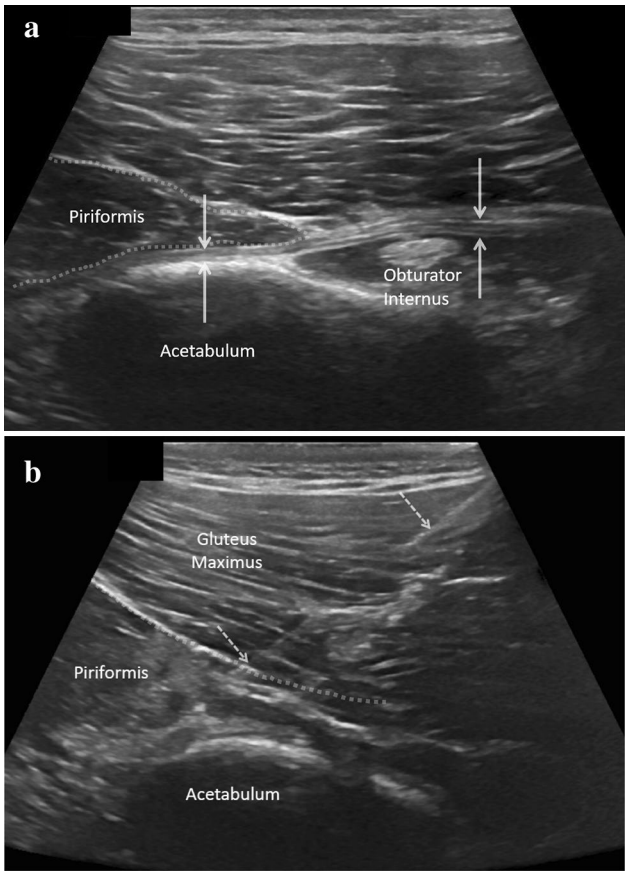

Fig. 1 **a** 18-Year-old elite Australian Rules footballer with buttock and referred below-knee pain. Longitudinal ultrasound of right buttock at the infra-piriformis fossa. A prominent lower piriformis belly (dashed line) compresses the sciatic nerve against the acetabulum (left arrows) with loss of normal intraneural fascicular architectural pattern. Note mild fascicle oedema and increased calibre of the sciatic nerve (right arrows) below the lower piriformis muscle belly. **b** Transverse ultrasound of right buttock at the infra-piriformis fossa during targeted piriformis injection. Needle (dashed arrows) shown, with the tip within the lower piriformis muscle belly (below dashed line) at the acetabulum

with positive response to local anaesthetic injection however, there was no response to cortisone injection at clinical follow-up.

At 6 weeks, the player returned for US guided low-dose BTX-A injection where targeted injection of lower muscle belly of the right piriformis muscle adjacent to the sciatic nerve at the posterior column of acetabulum was performed with 50 U BTX-A to selectively reduce compression of the sciatic nerve by the lower piriformis muscle belly (Fig. 1b).

Post-injection the player was near symptom-free on return to pre-season training and during the following football season, exceeding team average running and sprinting volumes on in-season GPS assessment, with some residual symptom 'awareness' only. Due to these residual right buttock and leg symptoms, MRI of the sciatic nerves was performed the following pre-season. This demonstrated right sciatic neuritis due to compression of the nerve by the lower belly of piriformis against the posterior column of the acetabulum (Fig. 2a). Bilateral Type A piriformis muscles were present, with the right lower muscle belly musculotendinous junction extending to within 1.5 cm of its greater trochanteric insertion (Fig. 2b). The right piriformis muscle was asymmetrically larger at the infra-piriformis fossa compared to the left (Fig. 2a). Anatomical variation of the biceps femoris proximal tendon was also observed, with the lateral tendon contributing to the sacrotuberous ligament, attaching to the sacrum rather than the ischial tuberosity. Repeat low-dose (50 U) US guided BTX-A injection of the lower right piriformis muscle was performed. Post-injection, complete resolution of residual PS symptoms was noted and no symptom recurrence or impairment of athletic function over the following pre-season and football season.

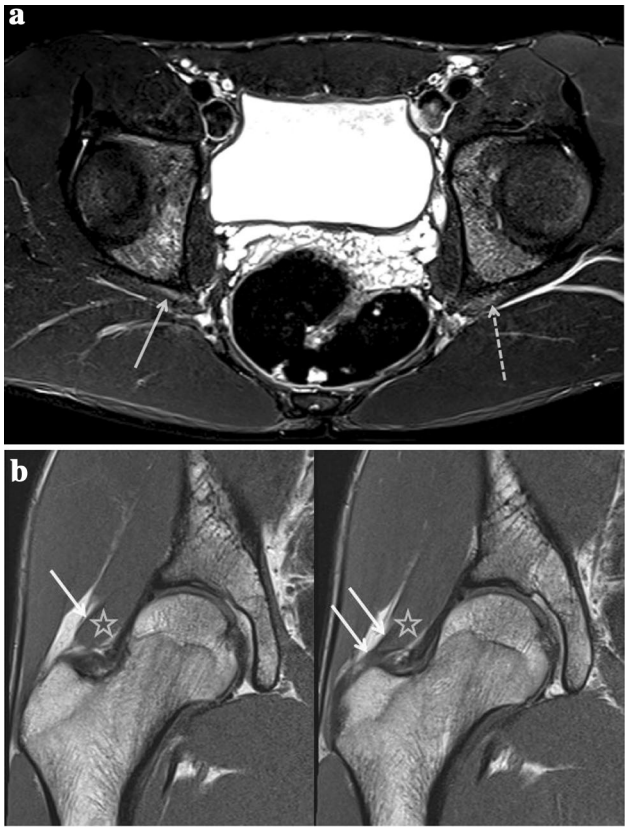

Fig. 2 **a** 18-Year-old elite Australian Rules footballer with buttock and referred below-knee pain. Axial T2-weighted fat saturated MR image of pelvis. Asymmetric sciatic nerve T2 signal is noted with confluent right sciatic nerve hyperintensity due to compressive neuritis (arrow), compared to normal left sciatic nerve (dashed arrow) demonstrating mildly hyperintense fascicles with typical dot-like morphology posterior to the acetabulum. **b** Coronal proton density (PD) MR images of the right hip. Type A piriformis musculotendinous junction with the lower muscle belly (star) MTJ extending to within 1 cm of the greater trochanter. Piriformis tendon position is demonstrated (arrows)

Discussion

Whilst the original description of PS by Robinson in 1947 referred to a specific clinical constellation of findings relating to the piriformis muscle following trauma, subsequent discussion of the diagnosis of PS has been controversial [7, 18]. This relates in part to the descriptions of other pathologies in and around the piriformis muscle also accounting for this clinical entity and the absence of diagnostic tests, including imaging, that are reliably able to confirm the diagnosis [18]. Additionally, whilst it is more widely recognised as a functional entrapment of the sciatic nerve as it leaves the pelvis, typically with buttock pain and lower limb radiculopathy ('neuropathic' presentations), descriptions of PS also refer to a myofascial pain syndrome associated with an hypertrophic, tender piriformis muscle, without associated neural entrapment ('somatic' presentations) [4, 7, 11, 13, 15].

The true incidence and prevalence of PS is unknown, largely due to the historical lack of clear diagnostic criteria and insensitivity of clinical and diagnostic tests, as well as it's overlap with other causes of deep gluteal syndrome that may confound diagnosis [4, 5]. The first two issues in particular contribute to the widely held perception that PS is under-diagnosed and why it is historically referred to as a 'diagnosis of exclusion' [12]. However, PS is thought by some to be a common condition, with experienced authors suggesting that it may be as frequent as herniated discs as a cause for sciatica [5]. Many cases of PS are 'somatic', relating to a myofascial syndrome of piriformis muscle over-activity or spasm [11]. Diagnostic tests including electrophysiological studies and imaging may be negative, especially in early presentations, as changes are functional and there is no neural compression, although piriformis

enlargement may be present [5]. These cases may be diagnosed clinically and effectively treated with rehabilitation, although local anaesthetic without or with cortisone injection is also widely used to establish the diagnosis [4, 11, 13]. In those with 'neuropathic PS', the authors believe that diagnostic studies including imaging and EMG are more likely to become positive due to the development of sciatic neuritis [4].

Imaging of PS has historically been performed to exclude alternative diagnoses including lumbar disc lesions and pelvic masses, amongst other differential diagnoses. US may visualise the piriformis muscle lateral to the sacrum however, assessment is generally limited to thin, young patients with sonolucent muscles, as in this case. Sonographic assessment is also severely hampered in middle-aged patients, the commonest PS demographic, by fatty replacement of gluteus maximus, which results in beam attenuation and limited piriformis muscle visualisation. When imaging is required, MRI is preferred as it able to more comprehensively evaluate the piriformis muscle and sciatic nerve, in addition to evaluating the pelvic contents and sacroiliac joints to exclude other pelvic diagnoses [4]. With more recent recognition of deep gluteal syndrome as a cause of buttock and referred lower limb pain, detailed assessment for the presence of sciatic neuritis and identification of perineural adhesions by MRI may also be necessary [4]. Otherwise, with regard to the imaging of PS in the existing literature, MRI imaging findings primarily relate to piriformis muscle asymmetry and sciatic nerve hyperintensity at the sciatic notch on conventional MR sequences and MR neurography [5, 16]. When combined, these two findings have a high specificity (93%) for prediction of good-to-excellent outcome for surgery, although the sensitivity (64%) is modest [5].

In clinically refractory cases of PS that have failed conventional rehabilitation, injection of a combination of local anaesthetic and cortisone or botulinum toxin have been demonstrated to be of benefit [6–8, 11, 12, 21]. There is now an increasing body of literature supporting the use of both botulinum toxin A and B in the management of PS [6, 8, 9, 11, 12, 21]. Typically doses administered range from 100 to 200 U BTX-A, although one study used a low-dose technique of 150 U Dysport, equivalent to 50 U BTX-A [21]. Surgical interventions for PS have consequently reduced since the introduction of botulinum toxin therapy [11].

Low-dose (50 U) US guided targeted injection of variant muscle anatomy with BTX-A has been used with clinical benefit in an athlete with functional popliteal artery entrapment syndrome [14]. Low-dose injection to the lower piriformis muscle belly was performed in this case to physiologically decompress the sciatic nerve at the infra-piriformis fossa whilst aiming to preserve piriformis muscle function. The positive clinical response to targeted injections of the lower piriformis muscle belly adjacent to the site of sciatic nerve compression in this case supports the hypothesis that Type A piriformis muscle MTJ morphology may play a significant role in the pathophysiology of PS. As the Type A piriformis is common, this variant may potentially contribute to a significant proportion of neuropathic PS presentations. Additionally, the presence of a Type A piriformis may explain the diagnostic utility of the FAIR test in PS, as a larger lower piriformis muscle belly increases compromise of the sciatic nerve at the infra-piriformis fossa with hip flexion, adduction and internal rotation (FAIR) [6]. We believe that this variant also explains the pathophysiology of piriformis-related dynamic sciatic nerve entrapment seen at endoscopy [4].

In this case, PS appears to be secondary to both functional and anatomical factors. Load-related compromise of the piriformis muscle in the dominant leg of a kicking and sprinting athlete is likely to contribute to piriformis muscle hypertrophy and/or spasm. Counternutation at the right sacroiliac joint, possibly associated with right L5 spondylolysis, rotates the lower sacrum and coccyx anteriorly, reducing the craniocaudal dimensions of the greater sciatic notch. Identification of a common anatomical variant of biceps femoris with a sacral attachment, seen in 50% of individuals, in the setting of increased biceps muscle tone, may also exacerbate counternutation [19]. When associated with a Type A piriformis muscle, these anatomical and functional factors may combine to compromise the infra-piriformis fossa and result in compression of the sciatic nerve by the lower belly of piriformis against the posterior acetabulum.

As with the imaging findings of sciatic neuritis in deep gluteal syndrome, neural fascicular oedema is present on MRI, associated with effacement of intra-neural and perineural fat [4] (Figs. 2a, 3). Fascicular oedema is not reliably assessed by US, except in suitable cases where there is adequate US tissue penetration (Fig. 1a). Additionally, compression of the nerve against the medial posterior acetabulum at the infra-piriformis fossa by a developmentally prominent and/or hypertrophic lower piriformis muscle belly is present (Figs. 2, 3). Neural compression and oedema (sciatic neuritis) are best observed on axial MR images (both T1 and T2-fat saturated) and Type A morphology of the piriformis MTJ may be appreciated on coronal non-fat saturated images (Fig. 2b). As noted above, sonographic assessment of PS is typically limited to young, athletic and/or slim patients with sonolucent muscles where the lateral aspect of the piriformis muscle, including a prominent lower muscle belly and sciatic neuritis due to compression, may be seen (Fig. 1a).

In conclusion, we believe that the positive response to targeted low-dose BTX-A injections of the lower piriformis muscle belly in this case supports the hypothesis that the Type A piriformis muscle variant plays a significant role in the pathophysiology of patients with neuropathic PS. However, development of PS is also likely to be associated

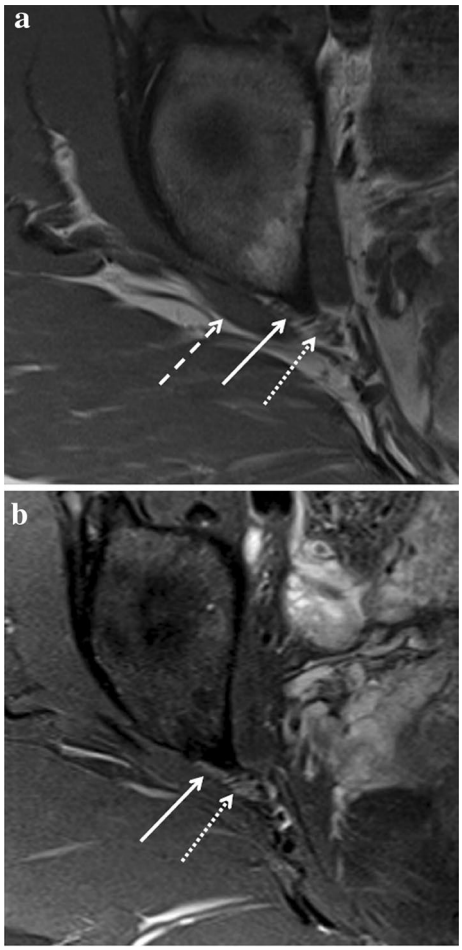

Fig. 3 **a** 37-Year-old female with buttock pain and clinical piriformis syndrome. Axial T1-weighted MR image of right hip showing compression of the lateral or common peroneal nerve (arrow) division of the sciatic nerve by the lower piriformis muscle belly (dashed arrow) compared to the tibial nerve (dotted arrow). Neural fascicles (dots and short lines on axial MR images) are of similar signal to muscle. Note effacement of peri- and intra-neural fat planes associated with common peroneal nerve compression when compared to tibial nerve. **b** Axial T2-fat saturated image of the right hip showing oedematous (high T2 signal) compressed common peroneal nerve fascicles of the sciatic nerve resulting in confluent neural high signal (arrow) compared to oedematous non-compressed tibial nerve fascicles, which have a white short linear appearance (dotted arrow) separated by black intraneural fat

with other functional changes, including piriformis muscle hypertrophy/spasm, pelvic instability (particularly sacroiliac joint counternutation) and anatomical variation with hamstring muscle overactivity that may also require treatment. We believe that imaging is an important part of the diagnostic pathway in neuropathic PS presentations as it may identify a common site of sciatic nerve compression at the infra-piriformis fossa that results in sciatic neuritis due to dynamic sciatic nerve entrapment previously reported by endoscopists. Further study into the role of the Type A piriformis in PS and the potential role of targeted BTX-A therapy to the inferior piriformis muscle in patients with this anatomical variant is recommended as imaging assisted diagnosis and management may preclude unnecessary surgical intervention.

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Author contributions ESCK: study conception and design, manuscript writing and critical revision. DW: manuscript writing, data collection and critical revision. JB: manuscript writing and critical revision.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Informed consent Informed consent was obtained from all individual participants included in the study. Additional informed consent was obtained from all individual participants for whom identifying information is included in this article.

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