



Piriformis syndrome as an overlooked cause of pain in a patient with axial spondyloarthritis: a case report

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Piriformis syndrome is a neuromuscular disorder characterized by hip, buttock, and leg pain. Axial spondyloarthritis is a rheumatic disease primarily affecting the sacroiliac joint and the spine. Due to their anatomical proximity, the potential relationship between piriformis syndrome and sacroiliitis has been discussed for some time. However, literature review revealed that there is no study on piriformis syndrome in individuals with axial spondyloarthritis. Here, we present the case of a 30-year-old female with axial spondyloarthritis who developed severe low back, hip, and buttock pain that persisted despite initial treatment for axial spondyloarthritis. We first re-evaluated her condition through physical examination, magnetic resonance imaging, and an injection test for piriformis syndrome. Following a comprehensive assessment, the patient was diagnosed with both axial spondyloarthritis and piriformis syndrome. Subsequently, a tailored treatment plan was devised, addressing both conditions, and after a 3-month course of treatment, we obtained significant reduction in pain of the patient. This is the first case report in literature, where we used injection test to confirm the diagnosis of the piriformis syndrome in a patient with axial spondyloarthritis. We therefore strongly advocate considering piriformis syndrome as a potential etiology for pain in individuals with axial spondyloarthritis consistently. This recognition is important as piriformis syndrome does not respond adequately to non-steroidal anti-inflammatory drugs and may lead to unnecessary use of biological disease-modifying antirheumatic drugs. Timely identification and intervention are imperative in ensuring optimal patient care.

Keywords: Axial spondyloarthritis, Piriformis muscle syndrome, Sacroiliitis, Ultrasonography, Pain

INTRODUCTION

Piriformis syndrome is a neuromuscular disorder characterized by the compression or irritation of the sciatic nerve by the piriformis muscle [1]. The piriformis muscle originates from the anterior surface of the sacrum and inserts onto the superior border of the greater trochanter. Notably, it shares a close anatomical proximity with the sciatic nerve. Piriformis syndrome usually manifests itself with hip and buttock pain, sometimes radiates to the leg [1]. Due to pain, individuals with this syndrome

may experience limitations in hip range of motion, discomfort when sitting, and postural changes. These symptoms can significantly impact an individual's quality of life, underscoring the importance of accurate diagnosis.

Piriformis syndrome is often overlooked in clinical practise because its presenting can be similar to that of lumbar radiculopathy, sacroiliitis [2]. Historically, the diagnosis of piriformis syndrome has been challenging, primarily due to the absence of consistent objective findings and the lack of a definitive single diagnostic test [3]. However, nowadays, the local anaesthetic

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injection into the piriformis muscle is accepted as a reference test [4]. There are many etiological factors in the pathogenesis of piriformis syndrome, the focus of our current discussion is on sacroiliitis, which is considered one of the potential etiological factors due to the proximity of the sacroiliac joint and the piriformis muscle [5-7].

Axial spondyloarthritis (axSpA) is a chronic inflammatory disorder predominantly affecting the axial skeleton [8]. The condition is characterized by a spectrum of clinical features, encompassing inflammatory back pain, arthritis, enthesitis, uveitis, and dactylitis [8]. Additional clinical features include a familial predisposition to spondyloarthritis, a personal history of psoriasis or colitis, radiographic evidence of sacroiliitis, and a notable prevalence of human leukocyte antigen (HLA)-B27 [8]. In our comprehensive literature review, no study was identified that specifically investigated piriformis syndrome in individuals diagnosed with axSpA. Considering that sacroiliitis is involved in the etiology of piriformis syndrome, it is surprising that there is no study on the diagnosis and treatment of piriformis syndrome in patients with axSpA.

Accurate diagnosis is crucial given the distinct treatment approaches for piriformis syndrome and axSpA. The first pivotal step is to consider piriformis syndrome in the differential diagnosis, particularly when sacroiliitis is implicated in its etiology, necessitating consideration in patients with axSpA. This awareness prompts a comprehensive patient examination, incorporating appropriate diagnostic methods, resulting in reduced complaints through targeted treatment. The initial diagnostic steps involve discerning the patient's pain pattern and identifying specific differences indicative of piriformis syndrome during the physical examination. Subsequent therapeutic measures encompass the injection test, serving both diagnostic and treatment purposes, alongside treatment planning involving exercises and the regulation of daily living activities. Failure to ascertain the pain's cause may lead to the mismanagement of a potentially easily resolvable issue, contributing to the progression of piriformis syndrome and associated morbidities such as pain, limitations in daily living activities, and even foot drop.

In light of these considerations, we find it pertinent to present a case involving piriformis syndrome and axSpA. Our intention is to underscore this relatively underexplored clinical association and advocate for further investigation in this area.

CASE REPORT

We present the case of a 30-year-old female who sought care at the rheumatology outpatient clinic of the university medical faculty with a history of complaining about low back and hip-buttock pain. The patient was first admitted to a general hospital 2 years ago due to inflammatory back pain, and she received a diagnosis of axSpA. She was prescribed a full dose of a non-steroidal anti-inflammatory drug (acemetacin, 120 mg/day). She reported that her pain and morning stiffness decreased with exercise and medical therapy initially, nevertheless, hip- but-tock pain of herself has intensified last 2 months. Subsequently in general hospital by medical doctor, patient was prescribed another non-steroidal anti-inflammatory drug (meloxicam 15 mg/day), but her symptoms were not improved then the patient was referred to our clinic with a biologic disease-modifying antirheumatic drug recommendation. No history of systemic disease (such as psoriasis or colitis) or familial predisposition to symptoms (familial spondyloarthritis) was identified. Additionally, there were no records of recent trauma or intramuscular injection. Furthermore, the patient had no history of arthritis or dactylitis. The patient reported that her pain gradually worsened with prolonged sitting and walking. For the case of severity of pain, she pointed out her pain level is an 8 out of 10 on the numerical rating scale. Disease activity was evaluated with Bath Ankylosing Spondylitis Disease Activity Index (BASDAI), a patient-based scoring system, and was found to be 4.3 [9,10]. On physical examination, tenderness was elicited over the left gluteal region and sacroiliac joints. The pain was provoked by applying flexion, adduction, and internal rotation of the hip (FAIR) and Freiberg test. The lumbar range of motion was within the normal range. No specific findings were observed during the inspection. There was no tenderness noted upon evaluation of the spinous processes or paravertebral muscles. Additionally, no stiffness was detected in the paravertebral muscles. Nervous system examined carefully, could not find any abnormalities. Considering the laboratory values; hemoglobin 13.1 g/dL (normal range: 11~15.4), white blood cells $5.83 \times 10^9/L$ (normal range: $4.01\sim 9.75$), platelet $234 \times 10^9/L$ (normal range: $151\sim 387$), erythrocyte sedimentation rate 9 mm/h (normal range: $0\sim 20$), c-reactive protein 3.8 mg/L (normal range: $0\sim 8$), creatinine 0.67 mg/dL (normal range: $0.6\sim 1.1$), alanine transaminase 11.00 U/L (normal range: $5\sim 34$). All values were within the normal range. Brucella agglutination test and HLA-B27 were negative. Radio-

graph of the sacroiliac joint revealed grade 1 sacroiliitis for the right sacroiliac joint and grade 2 sacroiliitis for the left sacroiliac joint. Radiograph of the foot and ankle revealed bilateral achilles enthesitis and calcaneal spur. Magnetic resonance imaging (MRI) of sacroiliac joints provided bilateral active inflammatory and structural lesions of sacroiliitis. The patient's sacroiliac joint radiography and MRI can be seen in Figure 1. MRI of the lumbar spine revealed no compression or irritation of nerve roots. Based on the aforementioned findings, we have substantiated the diagnosis of axSpA. This conclusion is supported by the patient's reported history of inflammatory back pain persisting for more than 3 months, radiographic evidence of sacroiliitis, presence of enthesitis and a positive response to non-steroidal anti-inflammatory drugs. In addition, we also pre-diagnosed piriformis syndrome based on her history (patient's pain which is provoked by sitting) and physical examination findings (tenderness in the piriformis muscle positivity in FAIR and Freiberg tests). Immediately thereafter, local anesthetic and steroid injection was meticulously injected into the piriformis muscle. The patient was positioned prone, ensuring neutral rotation of the lower extremities. Sterile technique was meticulously upheld during the entire injection process, guided by ultrasound. Prior to the procedure, a preliminary scan incorporated Doppler imaging to discern and avoid vessels. Special attention was devoted to visualizing the sciatic nerve, ensuring stringent measures to prevent any inadvertent contact with the needle. The transducer was firmly placed along the long axis over the piriformis muscle, ensuring precise alignment of the needle in-plane with the transducer. The procedure involved a 2 mL injection comprising lidocaine 2% combined with 1 mL of betamethasone, meticulously injected under ultrasound guidance. This utilized a 22-gauge, 90-mm spinal needle (Egemen International, Izmir, Turkey), complemented by a 1~8-MHz convex probe (Esaote

Co., Genova, Italy). After the injection, she rated her pain 3/10, this marked relief of pain confirmed the diagnosis. The patient was educated how to avoid activities so that increase pain. An exercise program including stretching movements was suggested. Meloxicam 15 mg was prescribed. At a 3-month follow-up, the patient reported that the pain was significantly improved, now rated as 0~1/10, and therefore, she returned back to her normal life. The last BASDAI score was 1.9.

DISCUSSION

In this case report, confirmed piriformis syndrome as the underlying source of pain in a patient diagnosed with axSpA. And with effective treatment of this syndrome, unnecessary biological disease-modifying antiheumatic drug treatment was prevented. The patient's diagnosis of axSpA was evaluated according to the Assessment of Spondyloarthritis International Society criteria for axSpA [11]. Piriformis syndrome was considered in addition to the diagnosis of axSpA, due to the characteristic feature of the pain, the FAIR and Freiberg tests, which we saw as positive in piriformis syndrome during physical examination, and the presence of tenderness on the piriformis muscle; this diagnosis was also confirmed by injection testing. Following the confirmation of both conditions, a customized treatment approach was instituted, leading to a notable reduction in the patient's pain.

The relationship between sacroiliitis and piriformis syndrome has been the subject of medical studies for years. Yeoman [5] were among the pioneers to explore the interplay between the sacroiliac joint and the piriformis muscle. In this study, the neighbourhood of the anterior sacroiliac ligament, piriformis muscle and sciatic nerve was taken into account, following this they concluded the inflammation in this region may be the



Figure 1. Radiograph and magnetic resonance imaging (MRI) of sacroiliac joints. This figure presents a radiograph and MRI of the sacroiliac joints taken at the time of diagnosis. The radiograph shows grade 1 sacroiliitis in the right sacroiliac joint and grade 2 sacroiliitis in the left sacroiliac joint. Additionally, the MRI, conducted using a short tau inversion recovery sequence in a semi-coronal orientation, reveals bilateral active inflammatory lesions characteristic of sacroiliitis.

cause of sciatica. Again approximately one century ago, Freiberg and Vinke [6] also stated that sacroiliac joint inflammation may cause sciatica. Despite these early insights, the association between piriformis syndrome and axSpA remained largely unexplored for many years. Nonetheless, emerging research is beginning to shed light on this connection. In a study by Vassalou et al. [7], using MRI on 116 patients with clinically suspected piriformis syndrome, it was found that three patients had piriformis syndrome secondary to ankylosing spondylitis-related sacroiliitis. Furthermore, there are case reports of sacroiliac osteomyelitis presenting as piriformis syndrome in the literature, some of them can be seen in [12,13].

Additionally, some report also suggest that sacroiliac joint dysfunction may contribute to piriformis syndrome [14], as it was found that a spasm in the muscle can trigger the sacroiliac joint strain, and at the same time, a strain in the sacroiliac joint can trigger spasm in the muscle. However, on the contrary, in literature review, we found some studies suggesting that there is no relationship between sacroiliac joint syndrome and piriformis syndrome [15]. For example, in a study undertaken by Bernard and Kirkaldy-Willis on nonspecific low back pain, only 19 of 336 patients with sacroiliac joint syndrome had painful muscle syndrome [16]. Therefore, based on the literature review above, it is evident that there is conflicting data among research studies, and there is a lack of sufficient research reports on piriformis syndrome in patients with axSpA. Our case report contributes to the existing literature by illustrating this association, as it identifies piriformis syndrome as the underlying cause of pain in a patient diagnosed with axSpA.

It is crucial to share this report to emphasize the significance of accurately treating piriformis syndrome in individuals with axSpA. If not treated appropriately and in a timely fashion, the disease can worsen, the etiology of pain may associate with other causes and then patient can face inappropriate medical treatments like biological disease-modifying antirheumatic drugs, and also, health insurance institutions might be under unnecessary financial burden. In our case involving axSpA and piriformis syndrome, both conditions were effectively managed through a combination of exercise, lifestyle modifications, and non-steroidal anti-inflammatory drugs. Consequently, there was no need to proceed to the next stage of axSpA treatment.

This is the first case report of a patient with axSpA diagnosed with piriformis syndrome using the reference test, the injection test. We suggest keeping piriformis syndrome in mind as a cause

of pain in patients with axSpA. Furthermore, there is a need for more studies evaluating presence of piriformis syndrome in patients with axSpA. Such a study already under consideration.

SUMMARY

In this study, we present a case report of a 30-year-old female patient diagnosed with piriformis syndrome alongside axSpA. The patient initially presented with symptoms of axSpA, including inflammatory back pain and heel pain. Despite receiving treatment for axSpA, the patient continued to experience intense low back, hip, and buttock pain. Based on the patient's medical history and physical examination, the patient was pre-diagnosed with piriformis syndrome in addition to axSpA. A diagnostic injection test confirmed the diagnosis, leading to significant pain relief for the patient. This case report highlights the importance of considering piriformis syndrome as a potential cause of pain in patients with axSpA and calls for additional research in this area. Correctly diagnosing and treating piriformis syndrome in such patients is crucial to prevent unnecessary medical treatments and alleviate financial burdens on healthcare systems.

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CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

AUTHOR CONTRIBUTIONS

All authors contributed to the concept. K.E. examined, analysed the patient and prescribed the treatment for axial spondyloarthritis. I.A.G. examined the patient, performed piriformis injection and arranged the piriformis syndrome treatment. E.A.T. was a major contributor in data analysis, reviewed the literature and wrote the manuscript. All authors read and approved the final manuscript.

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REFERENCES

1. Kirschner JS, Foye PM, Cole JL. Piriformis syndrome, diagnosis and treatment. *Muscle Nerve* 2009;40:10-8.
2. Kean Chen C, Nizar AJ. Prevalence of piriformis syndrome in chronic low back pain patients. A clinical diagnosis with modified FAIR test. *Pain Pract* 2013;13:276-81.
3. Scott DD, Anderson LC. The piriformis syndrome: a review. *J Back Musculoskelet Rehabil* 1994;4:248-54.
4. Misirlioglu TO, Akgun K, Palamar D, Erden MG, Erbilir T. Piriformis syndrome: comparison of the effectiveness of local anesthetic and corticosteroid injections: a double-blinded, randomized controlled study. *Pain Physician* 2015;18:163-71.
5. Yeoman W. THE relation of arthritis of the sacro-iliac joint to sciatica, with an analysis of 100 cases. *Lancet* 1928;212:1119-23.
6. Freiberg AH, Vinke TH. Sciatica and the sacro-iliac joint. *J Bone Joint Surg* 1934;16:126-36.
7. Vassalou EE, Katonis P, Karantanas AH. Piriformis muscle syndrome: a cross-sectional imaging study in 116 patients and evaluation of therapeutic outcome. *Eur Radiol* 2018;28:447-58.
8. Sieper J, Poddubnyy D. Axial spondyloarthritis. *Lancet* 2017;390:73-84.
9. Garrett S, Jenkinson T, Kennedy LG, Whitelock H, Gaisford P, Calin A. A new approach to defining disease status in ankylosing spondylitis: the Bath Ankylosing Spondylitis Disease Activity Index. *J Rheumatol* 1994;21:2286-91.
10. Akkoc Y, Karatepe AG, Akar S, Kirazli Y, Akkoc N. A Turkish version of the Bath Ankylosing Spondylitis Disease Activity Index: reliability and validity. *Rheumatol Int* 2005;25:280-4.
11. Rudwaleit M, van der Heijde D, Landewé R, Listing J, Akkoc N, Brandt J, et al. The development of Assessment of SpondyloArthritis international Society classification criteria for axial spondyloarthritis (part II): validation and final selection. *Ann Rheum Dis* 2009;68:777-83. Erratum in: *Ann Rheum Dis* 2019;78:e59.
12. Leong MK, Huang P. Piriformis syndrome as the only initial manifestation of septic sacroiliac osteomyelitis. *Clin Med (Lond)* 2020;20:e18-9.
13. Phadke PS, Gandhi AR, More SA, Joshirao RP. *Salmonella* pyomyositis with concurrent sacroiliac osteomyelitis presenting as piriformis syndrome: a rare case. *J Postgrad Med* 2017;63:44-6.
14. Kirkaldy-Willis WH, Hill RJ. A more precise diagnosis for low-back pain. *Spine (Phila Pa 1976)* 1979;4:102-9.
15. Papadopoulos EC, Khan SN. Piriformis syndrome and low back pain: a new classification and review of the literature. *Orthop Clin North Am* 2004;35:65-71.
16. Bernard TN Jr, Kirkaldy-Willis WH. Recognizing specific characteristics of nonspecific low back pain. *Clin Orthop Relat Res* 1987;(217):266-80.