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REVIEW ARTICLE

Piriformis pyomyositis, a cause of piriformis syndrome—a systematic search and review

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Abstract

Piriformis pyomyositis is a rare form of purulent skeletal myositis. As previous studies concerning piriformis pyomyositis had lower level of evidence and no systematic review has been published yet, we performed a systematic search to review and describe causes, symptoms, red flags, and available treatment options for piriformis pyomyositis. Using PubMed and PubMed Central databases, we found 21 articles describing 23 cases of piriformis pyomyositis. Based on the retrieved information, alongside acute sciatica like buttock and/or hip pain, high-grade fever, aggressive deep seated gluteal pain, neurological deficit of sciatic nerve distribution, positive straight leg raising test, and raised inflammatory biomarkers (erythrocyte sedimentation rate, ESR, C-reactive protein, CRP) provide clues for diagnosis of piriformis pyomyositis. Some cases were very ill but no death was documented. *Staphylococcus aureus* was the most common pathogen, but *Group A* as well as *Group β Streptococcus*, *Salmonella typhi*, *Proteus mirabilis*, *Brucella melitensis*, and *Escherichia coli* were also involved in the disorder. To treat the piriformis pyomyositis, broad-spectrum antibiotics were found to be useful; however, sometimes, antibiotic switching was warranted based on blood and tissue aspirate reports. Drainage and/or surgical exploration of the affected piriformis muscle were required in cases where antibiotics appeared ineffective. Piriformis pyomyositis is a rara avis and performing of prospective studies will hardly be feasible.

Keywords Myositis · Piriformis pyomyositis · Piriformis syndrome · *Staphylococcus aureus* · Systematic review

Introduction

Piriformis syndrome (PS) is an example of extra-spinal sciatica. It is also called deep gluteal syndrome, wallet neuritis, credit carditis, piriformis disorder, etc. [1]. Symptoms and signs of PS are due to piriformis muscle (PM) spasm and/or irritation of sciatic nerve vicinity or both [1, 2]. Features of PS can also be perpetuated by a fatty wallet compressing the adjacent sciatic nerve, called wallet neuritis; sometimes, the terminology is used interchangeably, though incorrectly with PS, as some wallet neuritis patients may not have unique features suggestive for PS [1, 2].

FAIR (flexion-adduction-internal rotation) test, Pace sign, and Freiberg maneuvers are often used in classifying PS individuals. Pace sign consists of pain and weakness by resisted abduction and external rotation of the hip when patients remain seated. Freiberg sign involves reproduction of pain and weakness on passive forced internal rotation of the hip in supine position [1, 2]. The Freiberg sign and Pace sign is found to be positive in 56.2 and 46.5% cases, respectively [1].

PS is an often overlooked cause of therapy-resistant sciatica in patients that is generally well treatable [3]. PS is an isolated disorder of piriformis muscle (primary) and/or irritation of sciatic nerve; yet, the condition has been documented in association with fibromyalgia, lumbar spinal canal stenosis, leg-length discrepancy, gluteal trauma, and repeated fall [2–4]. These conditions are generally benign and not associated with red flag signs. In some patients, PS may be caused by life-threatening conditions that deserve special attention like a developing pyomyositis. Though skeletal muscle infection most commonly involves the quadriceps muscle, gluteal

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muscles can also be affected [5]. It is important to diagnose and treat pyomyositis in a timely manner; otherwise, devastating consequences may threaten patients' life [5, 6]. We are yet to know details about piriformis pyomyositis including its prevalence among low back pain sufferers overall. Since in most occasions, these patients present with clinical features alike PS, diagnosis may be easily overlooked [3].

As of today, publications regarding piriformis pyomyositis include only a few case reports, case series, and all these can be categorized as level of evidence (LoE) 3 or 4 according to the Oxford Centre for Evidence-based Medicine (CEBM) [7]. There is no case control, cohort, randomized-controlled trial or systematic review concerning the condition.

So, we aimed to perform a systematic literature search and review, in order to collect and present all available information about piriformis pyomyositis, to describe causes, symptoms, red flags, and treatment for the condition and to increase the awareness of the physicians for this rare clinical scenario.

Materials and methods

To gather information available on piriformis pyomyositis, published works were searched and screened systematically using PubMed and PubMed Central databases from November 1, 2018 to February 28, 2019, and for this purpose, Preferred Reporting Items for Systematic Meta-Analysis (PRISMA) guidelines were used (Fig. 1) [8]. During this tenure using 'piriformis pyomyositis' strings, we found 21 and 39 published papers, respectively, in PubMed and PubMed Central databases (total 60 articles) for initial screening. Also, we looked for case reports mentioned in the reference section of these articles. Both authors were actively involved with analyzing retrieved articles. As it was a systematic search and review, there were no strict exclusion and inclusion criteria; however, articles falling in any of the following categories were excluded—(a) articles published in other than English or Dutch language (1-Spanish) and (b) articles that were not relevant to PM infection (33). Five articles were found to be overlapping in both searches. So, excluding 39 articles, finally, we found 21 articles including one Dutch article to be eligible for further screening. We found Dutch paper's abstract in English and Dutch co-author translated full-text English, hence included it in the review. We did not find any reports on proceedings of meetings or supplemental issues of journals of abstracts presented at congresses. All the screened articles are categorized as case series and review (2), letter to editor (3), case report (16). Based on the CEBM, they fall into either 3 or 4 level of evidence (C or D grade). We present retrieved information in four different (Table 1-4). Second, flowchart (Fig. 2) could guide reader how to diagnose, investigate, and manage piriformis pyomyositis as well.

Results

Among the 21 articles, nine, seven, and five articles describe piriformis pyomyositis in girls/women (Table 1), male adolescent and men (Table 2), and pediatric patients' (Table 3), respectively. We structured the findings including red flag signs (Table 4) into three subheadings: piriformis pyomyositis in girls and women, male adolescent and men, and pediatric patients.

Piriformis pyomyositis in girls and women

Chong and Tay described piriformis pyomyositis in a 30-year-old Thai woman presenting with features resembling sciatica. However, persistent and progressive gluteal pain, fever, and inflammatory biochemical (ESR, CRP) markers increased physicians' suspicion that there could something more than true sciatica, and further analysis of abdomen and pelvis using computed tomography (CT) unveiled a space occupying lesion involving the ipsilateral gluteus medius and PM that responded with parenteral vancomycin therapy. Previous dilatation and curettage for missed abortion might have induced the pathology [9].

In another report, *Colmegna* et al. described piriformis pyomyositis in a 18-year-old woman who had undergone unsafe induced abortion and presented with complaints of fever and clinical manifestations like right PS. With MRI, a space occupying lesion in the respective PM was revealed, and blood cultures and CT guided fine needle aspiration yielded *Staphylococcus aureus* as the causal pathogen; she was successfully treated with antibiotics [10].

Piriformis pyomyositis has also been described postpartum. *Kinaham* and *Douglas* described the condition in a 20-year-old woman, a few days after forceps delivery, with clinical, biochemical, and advanced imaging (CT/MRI) features suggestive of piriformis pyomyositis. With conventional NSAIDs and intravenous antibiotics, she recovered completely within 1.5 months [11]. *Gaughan* et al. described PM-pyomyositis in a 34-year-old Caucasian primiparous woman following vaginal delivery. Alongside progressive left gluteal, thigh, and leg pain, she reported lower limb weakness and spreading rash over the thigh; laboratory tests showed leukocytosis, raised ESR, CRP, and an MRI of the pelvis revealed edematous gluteal and piriformis muscles. As further scrutiny revealed a forearm cellulitis at the site of intravenous cannula, the authors concluded that this could have been the source of bacterial seeding to the left gluteus, piriformis, and adductor muscles from the infected cannula causing bacteremia; on the other hand, the organism could have been introduced through the traumatized genital structures during the delivery and transported into the cannula. Unfortunately, no cultures of blood or aspirate from the involved structures were done.

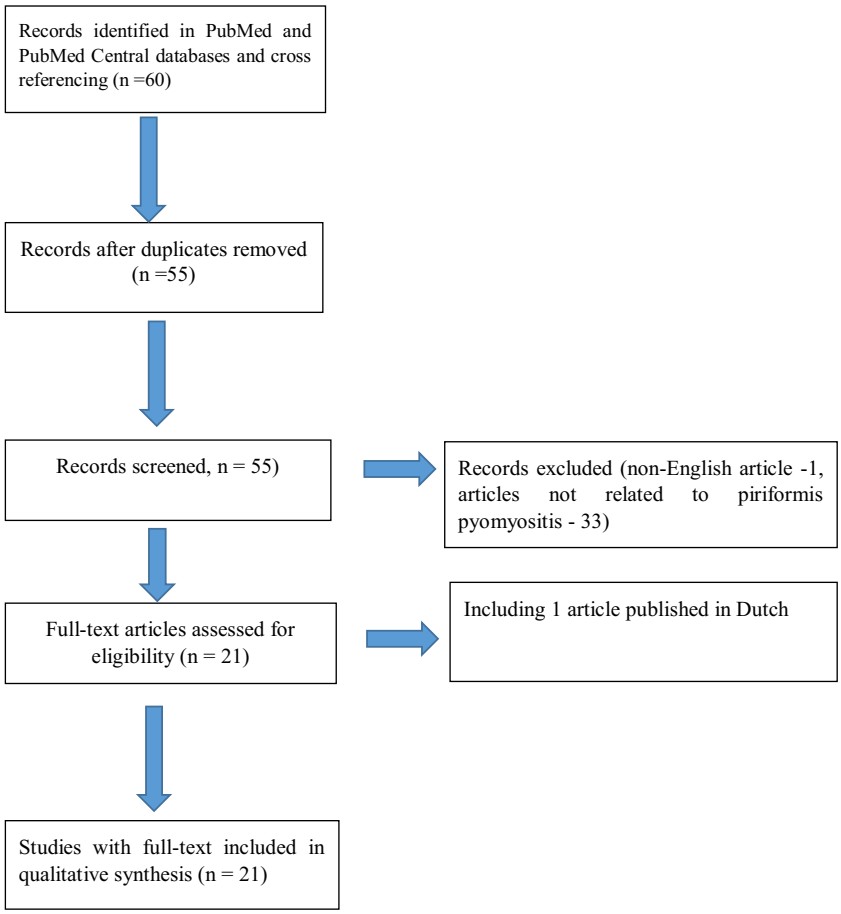

Fig. 1 Flow chart of screening and selecting articles for review

The patients' symptoms and radio-imaging signs improved after few weeks antibiotics [12].

In a case report, *Wong* et al. described the disorder at the 8th day post-partum of a 31-year-old woman following ventouse delivery of a 4000-g female infant. Eight days after delivery, the patient presented with a sudden onset left sciatica-like pain, with limited straight leg raise (SLR) test and fever, laboratory showed raised CRP and ESR and thrombocytosis. Blood cultures, vaginal swabs, and mid-stream urine were obtained. Her blood culture revealed heavy growth of *Group β-streptococcus*; however, high vaginal swab revealed heavy growth of *Escherichia coli*, moderate growth of *Group β-streptococcus*, and mixed anaerobes. Mixed growth was also found in mid-stream urine. An MRI scan of the pelvis revealed a space occupying lesion encroaching the left sciatic nerve and PM. She was treated with Tazobactam 4.5 g (three times a day—5 days), Vancomycin 1 g (three times a day—2 days), and Metronidazole (1 day), later replaced by oral amoxicillin-clavulanic acid 500/625 mg (three times a day) for a total 10 days. As the patient started improving with antibiotic treatment, aspiration was not attempted. She made an uneventful recovery and was discharged at day 10 post-presentation [13].

Koda et al. described piriformis pyomyositis in a 42-year-old female nurse complaining of severe right-sided sciatica with limited daily activities. PS maneuvers were not possible because of pain. Laboratory examination showed leukocytosis, raised ESR, and CRP. An MRI of the lumbar spine was normal, but CT/MRI scanning of the pelvis revealed high signal intensity suggestive of edematous, swollen PM, compatible with purulent myositis. As the patient was not improving with the antibiotic, her gluteal region was explored under general anesthesia to release PM and sciatic nerve, followed by improved gluteal pain and hip range of motion (ROM), although no apparent pus discharge was observed during the procedure. The patient got complete pain relief with normalization of PM morphology as shown by CT/MRI scanning with satisfactory job return within 2 months after the surgery [14]. *Sharma* et al. reported purulent PM in a 45-year-old type 2 diabetic woman, who presented to the emergency department complaining of right hip pain, limping, raised body temperature, and features resembling septic shock. On physical examination, there was antalgic gait, the right hip was externally rotated with limited ROM and right-sided gluteal tenderness. Pelvic CT scanning revealed edematous, necrotic right obturator internus, and PM. After surgical exploration, the muscles were debrided and irrigated resulting in improvement of the patient's condition and normalization of her body temperature. Histology confirmed the diagnosis of necrotizing

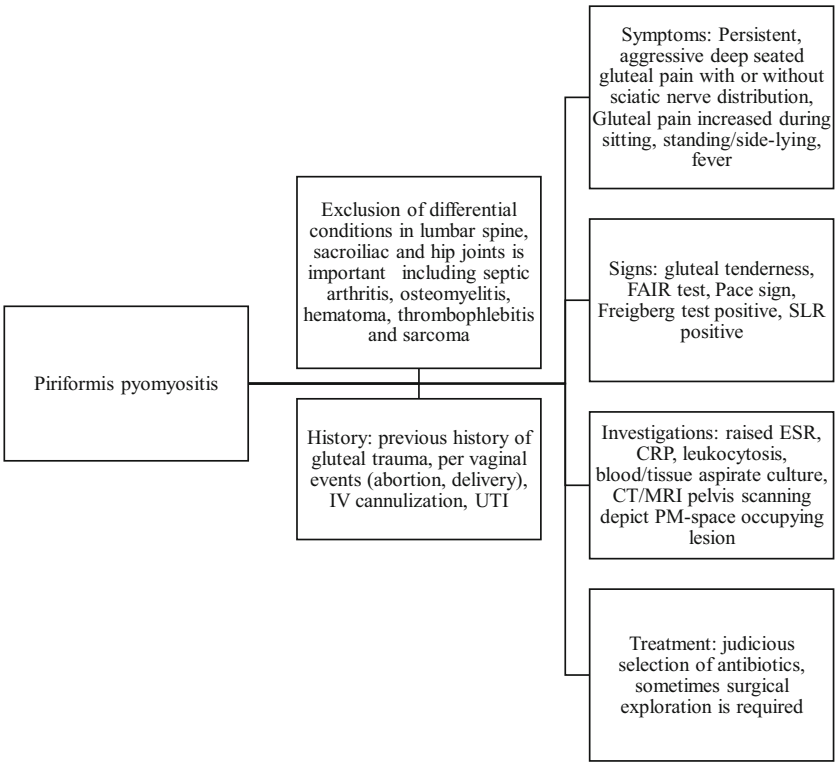

Fig. 2 Flow chart for diagnosing, investigating, and managing piriformis pyomyositis. Note: IV, intravenous, UTI, urinary tract infection, MRI, magnetic resonance scanning, CT, computed tomography, CRP, C-reactive protein, ESR, erythrocyte sedimentation rate, SLR, straight leg raise, FAIR, flexion-adduction-internal rotation, PM, piriformis muscle

myositis, and tissue culture revealed *Group A Streptococcal* bacteria. The patient was treated successfully with benzyl penicillin and clindamycin [15].

In a case series, *Wong* et al. documented piriformis pyomyositis in three elderly women [16]. The first case was a 45-year old woman, presenting with radiating low back pain suggestive of sciatica, though she later developed high-grade fever and impaired daily activities, limited hip joint movements. Alongside raised inflammatory biomarkers, CT scanning showed abscess formation in the left pelvis, with extension to the iliacus and piriformis muscles; she was treated empirically with intravenous cloxacillin with gradual resolution of the pain. The second case was a 58-year-old woman, following a 2-week history of progressive lower back and buttock pain and a 1-week history of fever, chills, and rigors. The patient was not improving with intravenous meropenem 500 mg every 8 h and metronidazole 500 mg every 8 h, followed by intravenous Co-amoxiclav (1.2 g every 8 h) and oral augmentin (375 mg 3 times daily). In this case, CT-guided drainage of the piriformis abscess was required as the symptoms persisted even after introducing the parenteral antibiotics. As the aspirate yielded negative cultures, leaving only numerous neutrophils suggestive of acute inflammatory exudate, the patient was treated with antibiotics empirically. Case 3, a 71-year-old woman presented with the complaint of 1-day fever and pain in the left buttock, lower abdomen, and raised inflammatory biomarkers were found. Ultrasonography of the abdomen revealed bilateral hydronephrosis, not improving with transurethral Foley catheterization; further evaluation with retrograde pyelography revealed obstruction at the pelvi-ureteric junction that could be the contributing factor for the urinary tract infection that further spread to the left PM causing pyomyositis as revealed under pelvis CT scan. The patient was empirically treated with intravenous cefuroxime 750 mg every 8 h, but as urine and blood culture yielded *Staphylococcus aureus*, this treatment was changed to cloxacillin (intravenous cloxacillin 1 g every 8 h of intravenous cloxacillin 1 g every 8 h for 2 weeks followed by 4 weeks of oral cloxacillin—1 g 4 times daily) with improvement of pain, fever, and myositis features as a result.

Hu et al. described piriformis pyomyositis in a 16-year-old girl [17]. She presented complaining of radiating gluteal pain impairing her daily activities, later complicated by fever and urinary retention which eventually was relieved with catheterization. There was right gluteal tenderness, reduced pinprick sensation according to right S3 and S4 dermatomes, and reduced right ankle jerk. Inflammatory biomarkers (ESR/CRP) were raised. Gadolinium-enhanced MRI scan of the pelvis revealed a '4 × 5 × 3'-cm space occupying lesion within the PM. Blood and CT-guided PM aspirate yielded *Staphylococcus aureus*; she was treated with intravenous flucloxacillin followed by oral flucloxacillin and fusidic acid over 2 months providing complete relief without any recurrence within next 1 year [17].

Table 1 Piriformis pyomyositis in girls and women

Study	Patients' demography	Clinical findings	Inflammatory biomarkers	Blood or aspirate cultures	Imaging	Treatment
Chong and Tay 2004 [8]	30-year-old Thai woman	Severe gluteal pain, fever	Raised ESR, CRP, leukocytosis		CT scan of pelvis	Parenteral and oral antibiotics (vancomycine)
Colmegna et al. 2007 [9]	18-year-old woman	Unsafe abortion 1 month ago	Raised ESR, CRP, leukocytosis	Blood and pus culture yield *S. aureus*	CT and MRI pelvis	4-week broad spectrum antibiotic, but didn't specify
Kinahan and Douglas 1995 [10]	20-year-old woman, 40-week gestation (early labour) (complicated pregnancy, thrombocytopenia	5th post-partum day develop severe left trochanteric pain, knife-like buttock, low back/hip pain, external rotated leg, buttock tender, PR exam. Painful, SLR positive	Raised ESR, CRP, leukocytosis	Not done	CT and MRI pelvis revealed iliacus and PM swelling	Parenteral and oral antibiotics (6 weeks) (not specified)
Gaughan et al. 2011 [11]	34-year-old Caucasian primiparous woman	Progressive left thigh pain 2nd postpartum day onwards, buttock pain, thigh rash on 8th day,	Raised ESR, CRP, leukocytosis	Not done	MRI of pelvis	Benzylpenicillin, parenteral flucoxacillin, metronidazole
Wong et al. 2012 [12]	31-year-old nulliparous woman with eventful ventouse delivery	Excruciating sciatica like pain, low back pain, left lower leg pain, fever	Raised ESR/CRP, alkaline phosphatase,	Blood and high vaginal swab culture revealed isolated heavy growth of Group β-streptococcus and *E. coli*, respectively	CT abdomen/pelvis, MRI pelvis	Tazobactam (5 days), Vancomycin 1 g (2 days) and Metronidazole (1 day)) followed by to oral co-amoxyclav (625 mg) and amoxicillin (500 mg) for a total duration of 10 days
Koda et al. 2013 [13]	42-year-old female nurse	Radiating low back, right buttock pain, impaired daily activities	Raised ESR, CRP, leukocytosis	Not done	CT and MRI pelvis	Surgical exploration of tendinous portion of PM, parenteral antibiotics (not specified)
Sharma et al. 2011 [14]	45-year-old type 2 diabetic woman	Fever, septic shock, sever localizing right hip pain, limited movement	Raised ESR, CRP, leukocytosis	Group A Streptococcus cultured from tissue debris	MRI and CT scan pelvis	Surgical exploration, PM debridement and drainage, benzylpenicillin and clindamycin
Wong et al. 2008 [15]	45-year-old women	Radiating low back pain, later high fever impaired daily activities, limited hip joint movements.	Raised ESR, CRP, leukocytosis	Blood culture yields *S. aureus*	CT scan pelvis	Cloxacillin
Wong 2008 [15]	58-year-old woman	14 days progressive lower back and buttock pain and 1-week fever, chills, rigor	Raised ESR CRP	Aspirate negative culture	CT scan of pelvis	Meropenem (no effect) Augmentin IV and orally and CT-guided drainage of the piriformis abscess
Wong 2008 [15]	71-year-old woman	fever and pain in the left buttock, lower abdomen	Raised ESR CRP	urine and blood culture positive for *S. aureus*	CT scan pelvis and abdomen	IV cloxacillin for 2 weeks and 4 weeks of oral cloxacillin

Table 1 (continued)

Study	Patients' demography	Clinical findings	Inflammatory biomarkers	Blood or aspirate cultures	Imaging	Treatment
Hu and colleagues 1998 [16]	16-year-old girl	Radiating low back pain, fever, urinary retention, gluteal tenderness, reduced sensation S3 and S4 dermatomes	Raised ESR CRP, leukocytosis	Blood and aspirate culture grew *S. aureus*	Gadolinium-enhanced MRI-pelvis revealed PM mass	IV flucloxacillin followed by oral flucloxacillin and fusidic acid for two months.

IV intravenous, *CRP* C-reactive protein, *ESR* erythrocyte sedimentation rate, *MRI* magnetic resonance scanning, *CT* computed tomography, *PM* piriformis muscle, *S* sacral

Piriformis pyomyositis in male adolescent and men

Chusid et al. describe a 17-year-old young man, a competitive swimmer who presented with complaints mimicking piriformis pyomyositis 2 days after vigorous swimming [18]. Besides unique features for PS, he had positive bilateral SLR test, raised ESR/CRP, and neutrophilia and blood cultures yielded *Proteus mirabilis*. MRI/CT abdomen and pelvis revealed PM swelling. Directly after diagnosis, IV vancomycin and cefotaxime were started, later switched to cefotaxime and tobramycin providing clinical and biochemical relief within 2 weeks [18].

Patients with piriformis pyomyositis may also present at the emergency department with severe hip pain, positive straight leg raise sign, fever with rigor, and sweating as depicted by *Kraniotis* et al. in a 19-year-old Caucasian man with no immune-compromised conditions. Besides PS symptoms and fever, there were increased common inflammatory biomarkers; pelvic MRI T1-weighted image and STIR scanning revealed edematous right PM and abnormally high signal intensity, respectively. After gadolinium administration, widespread extension of the lesion to soft adjacent tissues was revealed, suggestive of myositis. Agglutination test revealed a high titer (> 1/1280) for *Brucella melitensis*, and blood culture was also positive for *Brucella melitensis*. Antibiotic treatment for brucellosis was launched with the following regimen: doxycycline 100 mg 2 times/day, rifampin 900 mg once/day, and ciprofloxacin 500 mg 2 times/day. After 6 months of treatment, the patient was completely recovered regarding both clinical and MRI findings [19].

In an interesting case report, *Oh* et al. describe piriformis pyomyositis complicated with extensive epidural abscess as found by MRI scanning, involving all the spine segments in a 19-year-old male who presented with history of 2-week fever, low back pain, and nuchal rigidity [20]. The epidural abscess symptomatically improved following surgical decompression with regaining of limbs muscle strength. Cultures from the epidural abscess aspirate yielded methicillin-sensitive *Staphylococcus aureus* that was treated successfully with a 6-week course of intravenous nafcillin [20].

A 21-year-old Asian male complained about radiating right sciatica complicated with fever as described by *Phadka* et al. [21]. There was gluteal tenderness, leukocytosis, predominantly neutrophils (81.7%), raised CRP, and ESR. Pelvis MRI imaging showed high signal intensity in the right PM, with subtle changes in the ipsilateral right sacroiliac joint. Blood culture yielded *Salmonella typhi*. As the lesion was small and located deeper, surgical exploration was deferred. Initial lower ceftriaxone dose was found ineffective, but later up-titrating intravenous ceftriaxone (2 g 12 hourly) for 6 weeks improved the

Table 2 Piriformis pyomyositis in adolescent and adult male

Study	Patients' demography	Clinical findings	Biochemical markers	Culture report	Imaging	Treatment
Chusid 1998 [17]	17-year-old boy	High-grade fever, back pain, severe leg pain	Raised ESR and CRP	Blood culture yields *P. mirabilis*	MRI/CT abdomen and pelvis	Cefotaxime, tobramycin
Kraniotis et al. 2011 [18]	19-year-old Caucasian man	Fever with rigor/sweating, right hip pain	Raised ESR, CRP, leukocytosis	Blood culture positive for *B. melitensis*	MRI scan pelvis	doxycycline 100 mg twice daily, rifampin 900 mg daily and ciprofloxacin 500 mg twice daily
Oh et al. 2016 [19]	19-year-old male	Two-week fever, low back, nuchal rigidity	Raised ESR, CRP, leukocytosis	Epidural abscess culture revealsmethicillin-sensitive *S. aureus*	MRI whole spine, CT abdomen/pelvis	Surgical decompression, IV nafcillin.
Phadke et al. 2017 [20]	21-year-old male	Right gluteal pain, sciatica, fever	Raised ESR, CRP, leukocytosis	Blood culture grows *S. typhi*	MRI pelvis	Intravenous ceftriaxone dose was stepped up to 2 g 12 hourly for 4 weeks
Chen 1992 [21]	42-year-old man	Radiating left gluteal pain gluteal pain on deep palpation, externally rotated left leg, positive SLR, sensory and motor loss	leukocytosis, elevated ESR	pus culture yields *S. aureus*	CT pelvis revealed PM swelling	Surgical intervention was done cephalosporine for 3 weeks with complete pain and weakness relief within 3 months
Burkhart and Hamson 2003 [22]	69-year-old male		Raised ESR, CRP, leukocytosis			Parenteral and oral antibiotics
Giebaly et al. 2012 [23]	18-year-old boy, rugby player	Low back pain, left leg pain increased with straining, fever	Raised ESR, CRP, leukocytosis	Blood culture positive for *S. aureus*	MRI pelvis revealed piriformis muscle abscess	intravenous vancomycin and meropenim, later replaced with flucloxacillin and rifampicin

IV intravenous, *CRP* C-reactive protein, *ESR* erythrocyte sedimentation rate, *MRI* magnetic resonance scanning, *CT* computed tomography, *PM* piriformis muscle

Table 3 Piriformis pyomyositis in pediatric patients

Study	Patients' demography	Clinical findings	Inflammatory biomarkers	Blood and aspirate culture	Imaging	Treatment
Burton et al. 2005 [24]	14-year-old Caucasian boy	Intermittent fever, worsening right buttock and thigh pain	Raised ESR, CRP, leukocytosis	Not done	CT and MRI pelvis	Parenteral (flucloxacillin and ampicillin) and oral antibiotics
Schots et al. 2016 [25]	9-year old boy	Hip pain, limited daily activities and fever Pin point very localized pain	Raised ESR and leukocytosis	Blood- and pus culture positive for *S. aureus*	MRI pelvis	IV amoxicilline/clavulanic acid for 14 days followed by 4 weeks Clindamycin 25/kg/day po
Toda et al. 2013 [26]	6-year-old boy	right buttock, thigh pain, mild fever, sore throat	Raised ESR, CRP, leukocytosis	Not done	MRI pelvis	Oral antibiotics (10 mg/kg per day of cefdinir) were administered.
Miller et al. 2011 [27]	1 boy aged 2.5 years	Pain, fever for 5 days	Raised ESR more than CRP	blood culture negative	MRI pelvis	IV Co-amoxiclav and metronidazole for 4 days followed by 4 weeks Co-amoxiclav p.o.
Bhattad et al. 2016 [28]	4-year-old boy	Left buttock and thigh pain, fever	Raised ESR, CRP, leukocytosis	Blood culture positive for methicilin sensitive *S. aureus*	MRI pelvis	IV cloxacillin (2 weeks) and amikacin, oral cloxacilin for 4 weeks

ESR erythrocyte sedimentation rate, *CRP* C-reactive protein, *CT* computed tomography, *MRI* magnetic resonance scanning, *IV* intravenous

Table 4 Clinical and laboratory red flag features for piriformis pyomyositis

Characteristics	Description
Pain	Persistent, aggressive gluteal pain according to sciatic nerve distribution
Fever	Fever with or without rigor (high-grade) in association with gluteal pain
Past history	Past history of vaginal procedures, urinary tract infection, eventful IV cannulization
Signs	Positive FAIR test, Freiberg test, Pace sign and SLR
Laboratory markers	Inflammatory markers (ESR, CRP, total leucocyte count) are raised.
Blood or tissue aspirate culture	Could reveal inciting agents, in most cases *Staphylococcus aureus* is the main organism
Imaging	Pelvis CT and MRI scanning could reveal mass lesion in gluteal structures including piriformis muscle and their extension as well

IV intravenous, *FAIR* flexion-adduction-internal rotation, *SLR* straight leg raise, *CRP* C-reactive protein, *ESR* erythrocyte sedimentation rate, *MRI* magnetic resonance scanning, *CT* computed tomography

condition with normalization of inflammatory biomarkers and MRI-evidenced reduction of PM abscess size and reduced marrow edema of the involved sacroiliac joint [21].

A 42-year-old man with sudden pain, paresthesia involving left gluteal area, thigh and leg according to sciatic nerve distribution was described by *Chen* [22]. At examination, gluteal pain on deep palpation was found, externally rotated left leg, positive SLR, sensory, and motor loss, though without muscle atrophy. Further examination revealing pelvic wall tenderness, positive Freiberg sign, and the Pace sign favored PS diagnosis. However, associated leukocytosis, elevated ESR, later appearance of fever with aggravated gluteal pain, and CT revealing PM swelling provided further clue for the diagnosis of piriformis pyomyositis. The patient's gluteal area was explored surgically and aspirated pus was sent for culture-yielded *Staphylococcus aureus* that was managed successfully with cephalosporine for 3 weeks with complete relief of pain and weakness within 3 months [22].

Piriformis pyomyositis may also develop in an otherwise healthy individual as revealed in a 69-year-old tennis player and in an 18-year old rugby player [23, 24]. In both cases, patients developed sudden severe radiating low back pain within a very short duration mimicking prolapsed lumbar intervertebral disk; however, associated fever, inflammatory biochemical markers made the treating physicians wonder that there might be something else behind the scenario. Further scrutiny under MRI scanning revealed disk degeneration at L5/S1, fluid accumulation in the left iliopsoas, sacroiliac joint, and a space occupying lesion (abscess) within the left PM, compressing adjacent nerve roots with resultant sciatica features. Blood culture was positive for *Staphylococcus aureus*, and the patient was successfully treated with intravenous flucloxacillin and rifampicin with complete symptomatic relief after 24 weeks [24].

Piriformis pyomyositis in pediatric patients

Piriformis pyomyositis in pediatric patients is rare. In a 14-year-old Caucasian boy with 8-day history of intermittent fever, right buttock and thigh pain, and impaired daily activities, Burton et al. suspected piriformis pyomyositis [25]. Though the blood culture was negative, inflammatory markers were raised. The CT and MRI (T1 and T2) of the pelvis revealed ill-defined right greater sciatic notch mass and high signaled images of the piriformis muscle favoring the diagnosis of PM pyomyositis. MRI also showed an extension of the lesion up to the erector spinae muscles and around the right lumbar nerve roots, probably was contributing to the radicular symptoms. Despite negative cultures of blood culture and aspiration from the lesion, intravenous flucloxacillin and ampicillin for 7 days were given empirically resolving pyrexia and normalizing inflammatory markers and of MRI findings at 3-months [25].

Schots et al. describe a 9-year-old-boy who presented at the emergency clinic with a painful right hip that existed since 2 days. His active hip movement was painful. On passive movement and when walking, he could pinpoint the pain on one spot. Laboratory study showed elevated CRP and mild leucocytosis. Echography and conventional X-ray of the hip were normal. He was discharged after 1 day, but 2 days later, he returned to the clinic with increased pain and fever 38.9 °C, even higher CRP (156, $n < 1$) and the pinpoint gluteal pain. On MRI, a pyomyositis involved right piriformis and iliacus muscles with several small abscesses. Blood culture as well as echo-guided aspiration yielded *Staphylococcus aureus*. Detailed history revealed that an open blister on his right heel 3 weeks ago could have contaminated the pelvic muscles. He was successfully treated with intravenous amoxicillin/clavulanic acid for 14 days followed by 4 weeks clindamycin 25/kg/day orally [26].

Toda et al. describe features suggestive of piriformis pyomyositis in a 6-year-old boy presenting with severe right buttock and thigh pain with mild fever and raised inflammatory markers. Positive Freiberg sign was observed; T2-phase MRI images of the pelvis revealed high signal intensity as of the PM and corresponding sacroiliac joint. The patient improved following oral antibiotic, cefdinir (10 mg/kg per day), and experienced complete relief at 6-month follow-up with normal PM on MRI. In this patient, PM could get infected through the endo-pelvic fascia providing a route for infection from the pelvis and also from the pyoarthritis of the sacroiliac joint [27].

Miller et al. analyzed their 20-year database of over 16,000 pediatric orthopedic admissions and identified only three boys (2–12 years) with MRI-confirmed pyomyositis abscess, affecting the gluteal, piriformis, and adductor muscles. In one case of a 2.5-year-old boy, PM was involved with pain during 5 days and fever > 38 °C. Laboratory tests revealed high ESR and CRP, and MRI evidenced a small abscess in PM; yet, blood culture yielded no organism. Intravenous Co-amoxiclav and metronidazole for 4 days followed by 4 weeks oral Co-amoxiclav provided full recovery. Miller concludes that in children, pyomyositis features usually improve with a short course of intravenous antibiotics followed by 2–4-week oral therapy, though, sometimes open or percutaneous drainage is warranted [28].

Bhattad et al. described piriformis pyomyositis in a 4-year-old boy, where presenting complaints were high-grade fever, left buttock and posterior thigh pain, limited internal and external rotation of hip. Alongside point tenderness over the respective buttock, Freiberg sign was positive. Biochemistry revealed leukocytosis and raised CRP, and blood culture yielded methicillin sensitive *Staphylococcus aureus*. MRI of pelvis revealed hyper-intense signal in left PM and sciatic nerve sheath vicinity. After treating with intravenous cloxacillin (2 weeks) and amikacin, the patient started

becoming afebrile by the third day. However, the patient took oral antibiotics for another 4 weeks [29].

Discussion

The current review is the first review in medical literature regarding PS due to pyomyositis. Primary pyomyositis is also called tropical myositis and is rare in moderate climates, although the incidence may be rising [28, 30]. It is seen more frequently in children and in the elderly, especially when immune compromised, and slightly prevalent in males [31]. In a review, Bickels et al. found in 676 cases of primary pyomyositis of all ages, that the quadriceps (26%), iliopsoas (14%), and gluteal (11%) muscles were most often affected, and in 17% cases multiple sites were involved [31]. The etiology of pyomyositis differs and may be due to direct invasion of an infection into the muscle or due to a transient bacteremia, as also shown in our series. Pyomyositis can closely mimic numerous entities including septic arthritis, osteomyelitis, hematoma, thrombophlebitis, and sarcoma, and this should always be taken into account before diagnosis can be made [6].

Chiedozi et al. mentioned that skeletal pyomyositis follows three distinct phases—invasive stage (myositis without abscess formation), suppurative stage (myositis with abscess formation), and late stage with sepsis (patients are truly sick with consistent fever, septicemia, toxic and/or coma, could require intensive management) [5]; and in their study with 112 pyomyositis patients, performed in a tertiary hospital in Benin, suppurative stage was documented in highest 90% cases [5]. When identified, the causative organism is generally *Staphylococcus aureus*, like was the case in our series, although *Group A Streptococci*, *Escherichia coli*, and numerous others have also been isolated [6, 31].

The treatment of pyomyositis may include non-surgical approaches (non-steroidal anti-inflammatory drugs as well as antibiotics, depending on blood and drainage aspirate culture outcome) and surgical drainage, especially in the suppurative stage [4, 5]. The duration of antibiotic treatment depends on the clinical situation of the individual patients, but most authors use a regimen of intravenous followed by oral therapy for a total of 3–8 weeks, whether the abscess is drained or not [31]. Though intra-lesional (IL) steroid is one of the mainstays of PS treatment, here avoidance of such intervention with either corticosteroid or botox is warranted.

Twenty-three piriformis pyomyositis patients' described in our review were very ill and all these treatment modalities were applied depending on the clinical situation of the patient. None of these patients died, although in some cases, treatment was very complicated. We cannot exclude that other patients have died due to piriformis pyomyositis, but one can imagine that such cases may not be published by the treating clinicians. In most cases, diagnosis was made after clinical, laboratory, and MRI and/or CT scanning findings. Persistent gluteal pain is the frequent complaint of the disorder regardless of age and sex; however, pin point localized gluteal pain is characteristic for pediatric piriformis pyomyositis. Sometimes, a CT-guided aspiration and culture is warranted to confirm the diagnosis. In this review, *Staphylococcus aureus* ($n = 9$) was reported to be the single most important causative agent for piriformis pyomyositis ($n = 9$), but *Proteus mirabilis*, *Brucella melitensis*, *Salmonella typhi*, *E. coli*, and *Streptococcus beta and A* could also contribute in the disease process [10, 13, 15–19]. In nine cases, no causative agent was found or mentioned. In most cases, the judicial selection of antibiotics was enough for healing the pathology; yet, in five cases, surgical drainage was required.

Conclusions

all cases of piriformis pyomyositis have piriformis syndrome, but obviously not all piriformis syndrome is due to piriformis pyomyositis. In both conditions, deep-seated gluteal pain is common, but in case of pyomyositis, nonspecific systemic fever, raised inflammatory biomarkers, and typical pelvis CT or MRI scanning findings are characteristic; MRI is highly sensitive for muscle inflammation and fluid collection, and the investigation of choice. The treatment should be based on blood or PM aspiration culture findings, if available, and treated with appropriate antibiotics and guided by clinical results and CRP and/or ESR. If aspirate and blood cultures are negative or not available, patients should be treated with antibiotics empirically. In some cases, drainage and/or surgical exploration of the affected muscle may be required especially in patients are not responding to antibiotics. So, in a patient with piriformis syndrome aggressive buttock pain, fever with or without rigor and raised biochemical markers like ESR and/or CRP are red flags and the clinician should suspect pyomyositis and treat with appropriate antibiotics. However, we are yet to have an answer on the following questions: how prevalent is piriformis pyomyositis among piriformis syndrome? How does a piriformis pyomyositis develop? What is its pathogenesis? What are risk factors for developing piriformis pyomyositis? What about the natural history of the disease? If blood or aspirate cultures are negative or not available, what could be the antibiotic of choice for treating the condition? We expect that in the near future, researchers will find an answer on these questions.

Compliance with ethical standards

Disclosures None.

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