

Four symptoms define the piriformis syndrome: an updated systematic review of its clinical features

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Abstract

Purpose To update the evidence on the clinical features of the piriformis syndrome since the first systematic review published in 2010.

Method A systematic review of all case, cross-sectional and prevalence studies.

Results The commonest features reported were: buttock pain, pain aggravated on sitting, external tenderness near the greater sciatic notch and pain on any maneuver that increases piriformis muscle tension, and limitation of straight leg raising. The quality of case reports since the previous review has not improved with considerable under-reporting of presumed negative tests. Three recent cross-sectional and prevalence studies have been reported, but the two larger studies are at high risk of bias.

Conclusions Piriformis syndrome can be defined by a quartet of symptoms and signs. Many physical tests have been described, but the accuracy of these tests and the symptoms cannot be concluded from studies to date. Straight leg raising does not rule out the diagnosis. Piriformis syndrome is at a stage previously encountered with herniated intervertebral disc: that piriformis muscle pathology can cause sciatica has been demonstrated, but its prevalence among low back pain and sciatica sufferers and the diagnostic accuracy of clinical features requires cross-sectional studies free of incorporation and verification biases. One small cross-sectional study provides an encouraging example of how such

studies could be conducted but would need replication in a broader population and better reporting.

Keywords Piriformis syndrome · Sciatica · Systematic review · Diagnosis

Introduction

Piriformis syndrome (PS) refers to pain caused by impingement of the sciatic nerve by the piriformis muscle, causing buttock pain, sciatica or both. The sciatic nerve has an intimate relationship to the piriformis muscle, exiting the pelvis usually inferior to the muscle at the greater sciatic notch (Fig. 1). The piriformis muscle originates on the anterior surface of the sacrum, and its tendon attaches to the medial aspect of the greater trochanter. Its principal action is to externally rotate the hip. It additionally acts as a weak abductor and flexor. It is innervated by the spinal nerves L5 to S2. Various causes of impingement have been reported: congenital anomalies where the nerve or one of its branches passes through the piriformis muscle; trauma to the muscle including hematoma; overuse reported in athletes; muscle hypertrophy; shortening of the muscle; infection within the muscle; and leg length discrepancy. Specific tests have been suggested that, when present, are said to distinguish it from sciatica caused by radiculopathy (Table 1). These tests, apart from tonic external rotation of the hip, all reproduce the patient's pain by increasing pressure on the sciatic nerve, through tensing the piriformis and related muscles, either passively or actively.

The syndrome is controversial, with claims that it is under-diagnosed [7] [8] or over-diagnosed [9]. In this situation, the clinical features of the syndrome need to be

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Fig. 1 Anatomic relations of the piriformis muscle and sciatic nerve

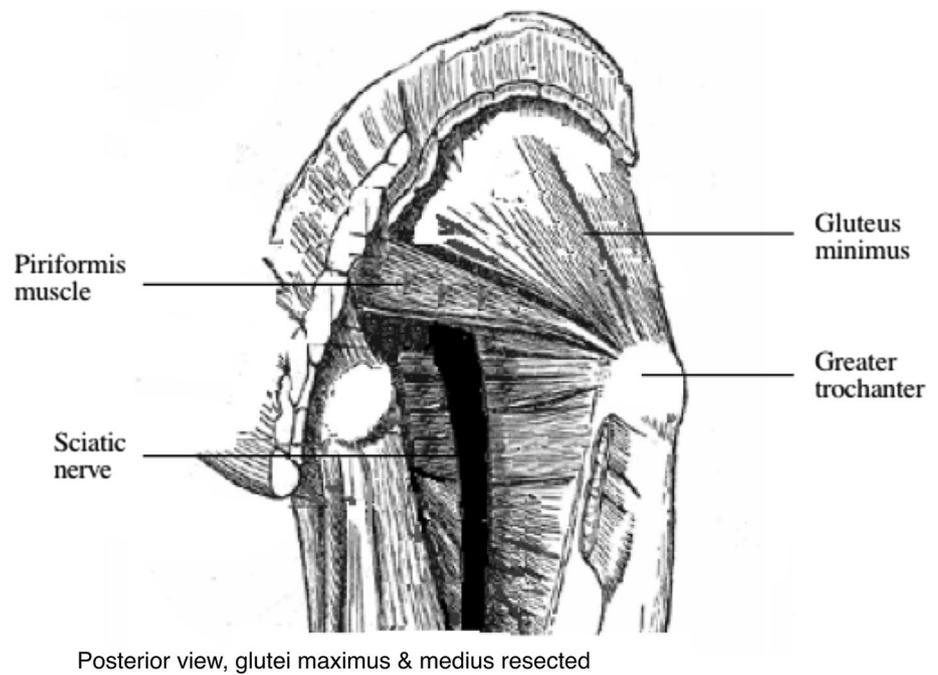


Table 1 Specific tests PS

Name of test	Date first described	Description	Attributed to
Freiberg	1934	The patient lies prone with knees flexed and then rotates the hip	Freiberg and Vinke [1]
Pace	1976	The clinician provides resistance to hip abduction by holding the sitting patient's knee	Pace and Nagle [2]
Tonic external rotation of hip	1981	Visible sign, hip externally rotated while patient at rest in supine position	Solheim [3]
FAIR = flexion abduction internal rotation of hip	1981	Maintaining the hip in flexion abduction and internal rotation.	Solheim [3]
Beatty	1994	The patient holds the flexed hip in abduction against gravity while lying on the unaffected side	Beatty [4]
Heel-contralateral knee maneuver (HCLK)	2013	The patient externally rotates, flexes the hip, and places the heel on the contralateral knee, and then, the examiner flexes the contralateral hip	Michel et al. [5]
Active piriformis	2013	The patients actively abducts and externally rotates the hip in the lateral position against resistance by the examiner	Martin et al. [6]
Seated piriformis	2013	The examiner internally rotates the hip and palpates the sciatic notch with the patient seated	Martin et al. [6]

These tests, apart from tonic external rotation of the hip, all reproduce the patient's pain by increasing pressure on the sciatic nerve, through tensing the piriformis and related muscles, either passively (forced by the examiner) or actively (performed by the patient). External tonic rotation is caused by shortening of the piriformis muscle

identified and be followed by studies of diagnostic accuracy and prevalence.

The first systematic review of the clinical features of the syndrome published in 2010 [10] estimated a range of frequencies for its clinical features. The evidence available was mainly in the form of case reports and series. The commonest features were: buttock pain, aggravation by sitting,

external tenderness over the greater sciatic notch and pain with maneuvers that increase tension in the piriformis muscle. The review raised five implications for research. First, whether PS-specific features are more frequent in PS than in patients with radiculopathy. Second, whether a combination of the quartet occurs more commonly in PS than other causes of sciatica. Third, whether the quartet is accompanied

by objective tests of nerve trunk compression, such as imaging or nerve conduction studies. Fourth, that further case reports and series will not improve our understanding of PS but if submitted, should report comprehensively. Fifth, the aforementioned research questions are best answered by a cross-sectional study with objective investigations for nerve compression. Since then, the relevant literature has been added to. Apart from more case reports and series, there have been large cohort and cross-sectional studies. Furthermore, nine reviews of PS [11–18] have appeared since 2010, but all were literature reviews rather than systematic reviews. Therefore, an updated systematic review is justified. The aims of this review were to update the evidence on the frequencies of the clinical features of PS and to evaluate the progress made against the above recommendations.

Methods

The review was registered with PROSPERO (registration CRD42014014706). The methods were in accord with the PRISMA statement on the conduct of systematic reviews [19].

Terminology

The terms used in PS research are inconsistent. While PS is usually reserved for compression of the sciatic nerve by the piriformis muscle, some use it for compression by any musculoskeletal structures in the pelvis and some even for space occupying lesions outside the PM such as endometriotic cysts [20–22]. Alternatives have been suggested to cover all conditions, such as pelvic outlet syndrome [23], deep gluteal syndrome [24] or nonspinal/extra-spinal sciatica [25]. This review takes PS to mean primary PS as described by Papadopoulos and Khan, namely pathology within the piriformis muscle [26].

The research definition of low back pain (LBP) is pain felt between the subcostal margin and the lower natal fold [27]. Some studies into PS use LBP to highlight pain in the lumbar area in opposition to the buttock [28, 29]. LBP and buttock pain are therefore reported separately.

Inclusion and exclusion criteria

All observational studies in peer-reviewed journals of patients presenting with back pain or sciatica in whom the diagnosis of PS was made and that reported clinical features were eligible. Studies were excluded if: they were not about PS; the patient did not present with back pain or sciatica; did not report clinical features; the language was not English, French, Spanish, Greek or Russian; the publication was not

a print or internet biomedical journal; or the condition was a complication of hip surgery or fracture.

Search

The NHS Evidence Healthcare Databases Advanced Search was used to search four databases: AMED, CINAHL, Embase and Medline. A broad search strategy was used to maximize returns (Table 2). References of retrieved articles were searched for further articles. The period of the search was from March 1, 2008, to December 31, 2016.

Screening

Two reviewers screened titles and abstracts for retrieval of reports. The references of retrieved articles were screened for further references. Both reviewers read the retrieved articles to decide on inclusion/exclusion. Disagreements were settled through discussion between the two reviewers until consensus was reached.

Data extraction

Data were extracted independently by two reviewers according to the previous protocol [10]. Two features not counted previously were added in this review: radiation of pain to the leg and paresthesia in the leg. Studies in the former review were scrutinized again for these additional features. Studies were divided into ‘individual data studies’ (case reports and case series reporting data for each patient) and cross-sectional studies.

Features stated as present or absent were recorded as positive or negative, respectively. If the absence of a feature was not explicitly stated or vaguely stated, it was recorded as ‘not reported.’

Quality assessment

The quality of case reports was assessed according to the previous protocol [10]. The history was given a categorical grading according to the number of items missing in the report: good ≤ 2 items missing; satisfactory = 3 or 4;

Table 2 Search terms

1. exp/sciatic*
2. sciatic*.ti,ab
3. 1 OR 2
4. Piriformis or pyriformis
5. syndrome
6. 3 AND 4
7 4 and 5
8 6 OR 7

poor ≥ 5 . Items essential in a pain history are age and sex, onset, site, radiation, relieving and aggravating factors, duration, evolution of the condition and past medical history. Examination was graded into three categories according to range of signs reported: PS specific, usual sciatica signs or both. Cross-sectional studies were assessed for risk of bias (ROB) according to QUADAS-2 [30].

Data synthesis

Data from individual data studies were combined with those of the 2010 review to calculate estimates of the frequencies. In the former review, analyses were undertaken for different potential denominators: whether studies had corroborated evidence (such as perioperative) or whether they explicitly reported the absence of features. Different analyses produced a range of frequencies, but the ranking of clinical features was consistent. In this review, only the denominator of all cases was planned because the primary aim of the current review was to identify the most common features. Data in the cross-sectional studies were unsuitable for pooling so a descriptive synthesis was planned.

Sensitivity analyses

Two sensitivity analyses were undertaken. First the subgroup of corroborated cases was analyzed separately to check whether this altered the ranking of clinical features. Second, the abstracts of excluded studies were scrutinized to evaluate the potential impact of their content on the results.

Results

Flow of studies

The flow of studies is shown in Fig. 2 and the planned analysis in Fig. 3. Twenty-five eligible studies were found: 22 case studies with data on 36 individuals, two cross-sectional studies and one prevalence study (Table 3).

Particular mention is due to one excluded study, Siddiq et al. [54]. It was excluded because the presence of a PS-specific sign was a criterion for recruitment so that the prevalence of these signs would automatically be 100% making pooling impossible. Nevertheless, the study is valuable because it replicates the findings on etiology in smaller case studies.

Results: case studies

Quality of case studies

The quality of case studies published after publication of the previous review shows a similar distribution between poor and good studies (Table 4). The difference between papers before and after 2010 is not statistically significant.

Frequencies from case studies

The most commonly reported features were in the order of decreasing frequency: radiation to the ipsilateral leg, external tenderness around the greater sciatic notch,

Fig. 2 Flow of records

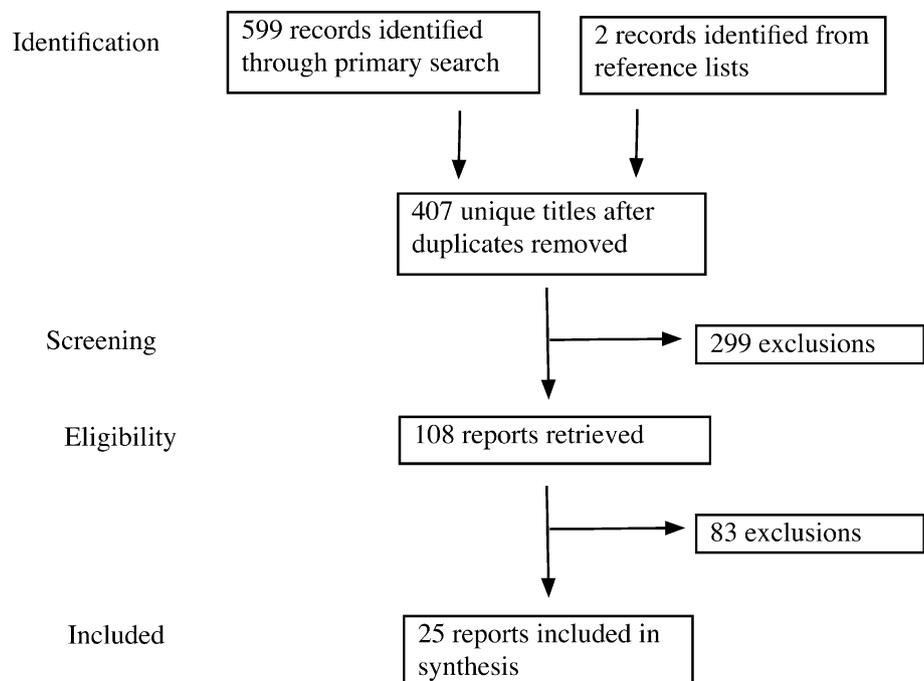


Fig. 3 Plan of analysis

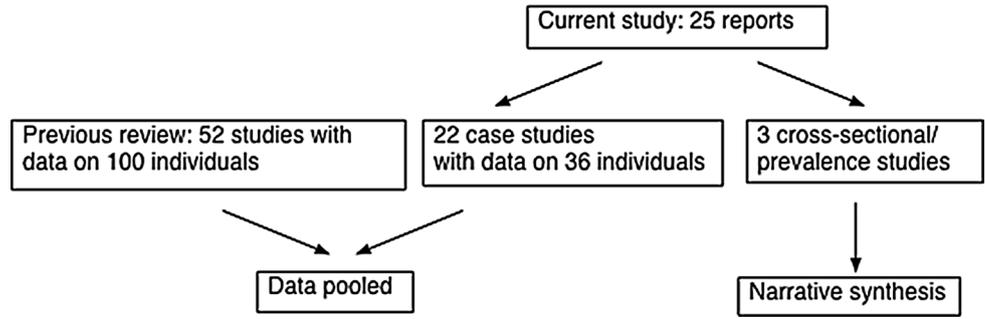


Table 3 Included studies

Study type			
Case studies	Study: first author, year	Number in study	Number included in review
	Aydemir K, 2010 [31]	1	1
	Chapman C, 2011 [32]	1	1
	de la Peña Parra, E, 2013 [33]	1	1
	Dere K, 2009 [34]	2	2
	Drampalos E, 2014 [35]	1	1
	Giebaly DE, 2012 [36]	1	1
	Hamdi W, 2013 [37]	1	1
	Kabatas S, 2008 [38]	1	1
	Koda 2013 [39]	1	1
	Kraus E, 2015 [40]	1	
	Kulkarni R, 2015 [41]	1	1
	Misirlioglu TO, 2015 [42]	1	
	Parlak A, 2014 [43]	1	1
	Ruiz-Arranz JL, 2008 [44]	14	14
	Siddiq MAB, 2014 [45]	1	1
	Sivrioglu AKI, 2013 [46]	1	1
	Sun CH, 2012 [29]	1	1
	Toda T, 2013 [47]	1	1
	Tonley JC, 2010 [48]	1	1
	Yildirim P, 2015 [49]	1	1
	Yoshimoto M, 2009 [50]	3	1
	Younger DS, 2015 [51]	1	1
	Villano EQ, 2015 [52]	1	
Cross-sectional studies	Study: first author, year	Number in PS group	Number in control group
	Martin MD, 2014 [6]	23	10
	Michel F, 2013 [5]	250	30 radiculopathy 30 asymptomatic
	Singh US, 2013 [53]	182	None

buttock pain, any PS-specific sign, limited straight leg raising (SLR) and reproduction of pain on prolonged sitting (Fig. 4). The female-to-male ratio was 75:61. The mean age was 43 years.

Sensitivity analyses

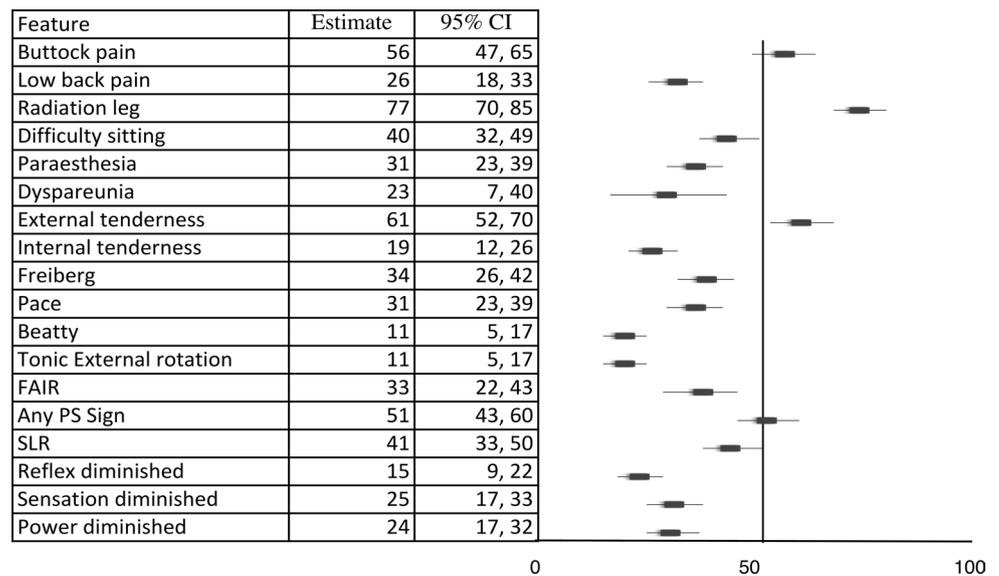
Of the 36 new cases in this review, fifteen had some form of corroboration, such as hypertrophy of the PM on MRI [31,

Table 4 Quality scores of case reports, before and after 2010

Signs	Case reports up to 2010			Case reports up to 2016		
	History			History		
	Poor	Satisfactory	Good	Poor	Satisfactory	Good
None	1	0	0	3	0	0
Routine sciatica signs only	2	1	2	0	1	2
PS signs only	2	4	0	1	1	2
Sciatica and PS signs	8	10	22	3	2	7

The quality of history is graded according to the number of items missing in the report: good ≤ 2 items missing; satisfactory = 3 or 4; poor ≥ 5 . Overall quality is represented by position in the grid so that the best studies are in lower right hand cell and the worst in the upper left hand cell

χ^2 using Yates correction $p = 0.138$

Fig. 4 Frequencies of clinical features from all case reports

38, 41, 43, 46, 49, 50], relief after injection of the piriformis muscle with corticosteroid [42], or relief after division of the piriformis muscle [40]. The ranking of clinical features was virtually the same in the subgroup analysis of 82 corroborated cases (Fig. 5).

The impact that might have been made by papers excluded on language grounds was minimal. Eight would have had no impact because they were either not about PS, were reviews rather than original papers or did not have clinical data (cadaveric studies). Eight were unlikely to have an impact because they did not appear to report data relevant to this review and they had reported on few cases. In three studies, the impact could not be assessed because no abstract was available.

Results: cross-sectional/prevalence studies

All three studies reported a full history and physical signs as defined above. This contrasts with the four large series

reported in the previous review none of which did so. The quality of cross-sectional studies expressed as ROB (lower ROB = higher quality) is shown in Table 5. Their estimates of frequencies are shown in Table 6.

Michel et al. [5]

Michel et al's cross-sectional study was the largest to date: 250 patients with PS seen at a single center. Patients received a structured examination including several PS signs. It included a new test, the heel-contralateral knee maneuver (HCLK): The patient externally rotates and flexes the ipsilateral hip with the heel placed on the contralateral knee, and then, the examiner flexes the contralateral hip, thus tautening the piriformis muscle and compressing the sciatic notch. The findings were compared with two control groups, 30 patients with intervertebral disk herniation and radiculopathy and 30 healthy subjects. Clinical features were combined to develop a clinical scoring system. One hundred

Fig. 5 Frequencies of clinical features from case reports with corroboration

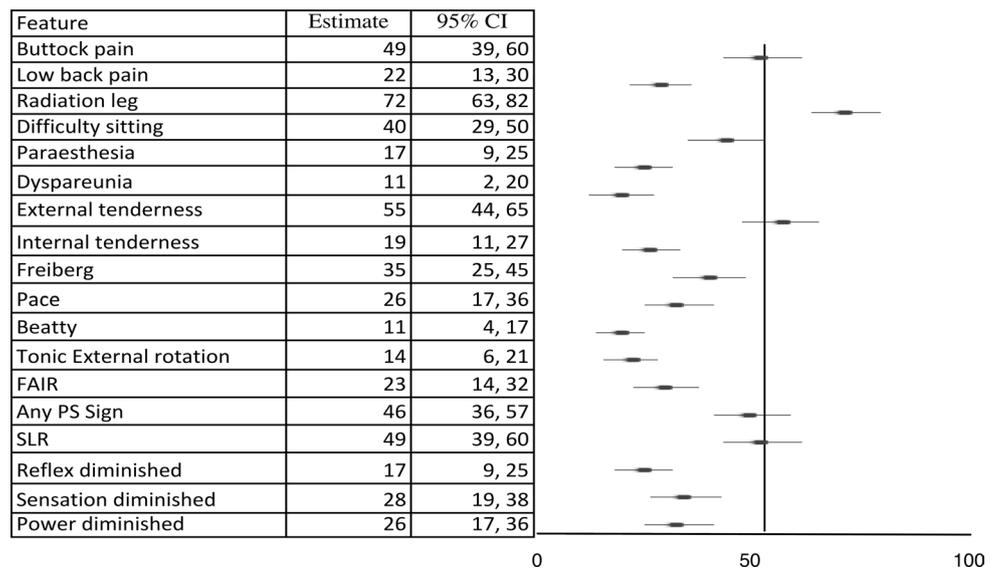


Table 5 Risk of bias in cross-sectional studies

Study	Patient Selection	Index Test	Reference Standard	Flow And Timing	Overall
Martin HD 2014	●	○	○	●	●
Michel F 2013	●	○	●	●	●
Singh US 2013	●	○	●	●	●

High risk of bias ● Moderate risk of bias ● Low risk of bias ○

percent of PS patients had positive PS tests. The difference between them and the controls was statistically significant, $p < 0.0001$. Two hundred and forty-one PS patients scored $\geq 8/12$ on the clinical scoring system compared to none in the controls, giving sensitivity and specificity of 96.4 and 100%, respectively. The study’s strengths are that it was prospective and recruitment was consecutive, but there were potentials for serious bias in the design. First, classification as PS depended on PS-specific tests which were themselves the outcomes under investigation (incorporation bias), so their prevalence of 100% was inevitable. Second, the comparison against healthy subjects, rather than symptomatic individuals, which is the accepted practice in diagnostic accuracy studies, ensured statistically significant differences. Third, the presence of ‘signs of lumbar radicular compression’ was an exclusion criterion which might explain why no PS patients had limited SLR, further biasing the scoring system.

Singh et al. [53]

This prevalence study of PS in patients presenting with LBP and sciatica to secondary care was conducted over 2 years. Out of 2910 patients, 182 were diagnosed with PS. The study

had potentially serious biases. Inclusion depended on pain reduction in response to an injection of corticosteroid and local anesthetic into the PM. This would misclassify some cases of PS as non-PS (misclassification bias) because PS, corroborated by imaging or surgery, that did not respond to local injection has been reported [10]. It is unclear whether participants and observers were blinded (reporting and observer biases). The authors did not report how patients were selected from the 2910 for injection or how many failed to respond to the injection (selection bias).

Martin et al. [6]

Martin et al. investigated patients who had undergone endoscopic evaluation of the sciatic nerve. They compared the clinical features of patients with and without sciatic nerve compression. Their clinic kept structured records of examination enabling such comparison retrospectively. Martin et al. introduced two variations on previous tests. In the active piriformis test, the patient actively abducts and externally rotates the hip in the lateral position against resistance by the examiner. In the seated piriformis test, the examiner internally rotates the hip and palpates the sciatic notch with the patient seated. The sensitivity and specificity for any

Table 6 Frequencies of clinical features (%) from cross-sectional/prevalence studies

Feature	Martin 2014	Michel, 2013 [5]	Singh, 2013 [53]
N	23	250	182
Radiation	35	100	100
Pain sitting	78	NR ^a	100
Paresthesiae	74	NR	2
External tenderness	NR	NR	84
Freiberg	52 ^b	100	84
Pace	NR	NR	55
Beatty	78 ^b	100	52
Tonic external rotation	NR	NR	52
FAIR ^d	NR	100	93
SLR	17	0 ^c	100
HCLK ^e	NA	100	NA

^a Pain on sitting or prolonged standing was, however, an inclusion criterion

^b Martin et al., the reported variants of these tests, Beatty = active piriformis, Freiberg = seated piriformis

^c A positive SLR was an exclusion criterion in this study

^d Flexion adduction internal rotation (of hip)

^e Heel-contralateral knee test, NR = not reported NA = not applicable (referring to a sign introduced by Michel et al.)

PS test (either active or seated piriformis test) were 0.91 (0.73–0.98) and 0.80 (0.49–0.94), respectively. The sensitivity and specificity of limited SLR were 0.15 (0.05–0.33) and 0.95 (0.68–1.00), respectively. The robust reference standard gives this study a lower risk of bias than Michel et al. and Singh et al. However, the study's generalizability and internal validity are uncertain. The sample, drawn from a tertiary clinic with strict criteria for endoscopy, did not contain the spectrum of patients seen across clinical settings (spectrum bias). Patients were selected for the study in reverse order to accepted practice, on the basis of receiving the reference standard rather than presenting symptoms (verification bias). Outcomes were incompletely reported as the authors did not state what proportion of those eligible for endoscopy actually had it. It is unclear whether the endoscopist was blind to the results of the piriformis tests at the time of endoscopy (observer bias).

Discussion

Study strengths

The current review's main strength is that unlike all other recent reviews it employed a systematic strategy concordant with internationally accepted reporting guidelines. Study

inclusion was comprehensive, and study quality was evaluated. The diagnosis of PM in the most recent studies was corroborated by tests such as MRI and endoscopic visualization. Sensitivity analysis of case studies where the diagnosis was corroborated was employed to test the findings.

Study limitations

The main limitation arises from the bias inherent in case reports. Case reports are not the best evidence possible, but sometimes they are the best evidence available [55] and they make up the bulk of the literature in PS. The aggregation of data from case reports can lead to refinement for further research [56] as this review has done. Language restrictions were applied. However, very few reports were excluded on these grounds and they were unlikely to impact on the conclusions. Part of the analysis was on data from case studies of poor quality. However, these were not used to suggest the actual frequencies of clinical features but to identify the commonest that might constitute a syndrome. Recent observational studies were included, two of which were large series, were available, but only one was of moderate quality.

The quality of studies

The quality of case reports has not improved in the past 5 years. Incomplete reporting remains a substantial shortcoming. Reporting of clinical features was more comprehensive in the cross-sectional and prevalence studies showing improvement over previous large series. However, two of these studies were at high risk of bias.

The clinical features of PS

This update confirms the findings of the previous review and provides new insights. It confirms that a quartet of symptoms constitute a syndrome and furthermore that the syndrome is associated with changes in the piriformis muscle/sciatic nerve relations. The quartet is composed of buttock pain, pain aggravated on sitting, external tenderness near the greater sciatic notch and any PS sign, that is, pain with increased PM tension. A new insight is the presence of SLR limitation, rebutting the implied concept of Michel et al. that SLR limitation excludes the syndrome. PS can exist without radiation to the leg so that it should be in the differential diagnosis of LBP and not considered only in the presence of sciatica. None of the cross-sectional or prevalence studies provide reliable data regarding diagnostic accuracy of symptoms and signs. Singh [53] may have underestimated the prevalence of clinical features, whereas Martin et al. [6] may have overestimated their diagnostic accuracy. However, Martin et al's outcomes are so

large that future studies are unlikely to contradict the direction of the effects although they would probably reduce the size.

Implications for clinicians

Singh's study suggests that the prevalence of PS is likely to be clinically important, 6.3% (95% CI 5.4, 7.2). As before, pain in the buttock and worsening of pain on sitting should prompt a look for external tenderness and PS-specific signs. The presence of limited SLR should not exclude the diagnosis.

Implications for research

Further case studies cannot advance our knowledge of PS. The previous recommendation remains: future research should be designed as a cross-sectional study of consecutive patients suffering LBP or sciatica. The recording of symptoms and signs must include *all* those, which the evidence so far suggests are indicative of PS. The symptoms and signs should be compared against objective investigations that include looking for *both* nerve root *and* nerve trunk compression. The study by Siddiq et al. demonstrates the need for improved data on diagnostic accuracy for large-scale prevalence studies to take place [54]. PS in relation to sciatic nerve impingement resembles the status had disk herniation in relation to radiculopathy in the twentieth century: a link between clinical features and pathology has been established, but the research evidence for prevalence and diagnostic accuracy of clinical features is weak. The model of disk herniation as the sole cause of sciatica was overturned as later research revealed other causes, notably lumbar canal stenosis. Similarly other causes of sciatic nerve impingement are being reported [24, 25, 57, 58] so that the concept of PS needs reshaping as part of wider extra-spinal conditions causing LBP and sciatica. The proportion of sciatica patients without demonstrable radiculopathy lies between ten percent [50] and 35% [59]. Greater knowledge of extra-spinal sciatic nerve impingement offers a potential explanation for such cases. Two strategies are needed to exploit this potential. First, cross-sectional studies as described above, preferably in primary rather than secondary and tertiary centers. Second, future community surveys of back pain and sciatica should include PS/extra-spinal sciatica features. Eighty years after Freiberg and Vinke [1] first postulated a relationship between the piriformis muscle, the sciatic nerve and sciatica, the time for high quality research is long overdue.

Supplementary material

Background information and links to videos and images of physical tests can be found at <http://www.angliangp.org/piriformis-syndrome>.

Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interests.

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