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Hip bone marrow edema presenting as low back pain: a case report

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ABSTRACT

Background: Nonspecific low back pain (LBP) is frequently managed by physiotherapists. However, physiotherapists in a direct access setting may encounter patients with serious medical conditions, such as Bone Marrow Edema Syndrome (BMES) of the hip with symptoms mimicking LBP. To our knowledge, this is the first case to describe hip BMES presenting as LBP. Diagnosis was based on the patient's symptoms in conjunction with magnetic resonance imaging (MRI). In order to avoid misdiagnosing the patient, primary care clinicians should be aware that BMES can mimic nonspecific LBP. Objective: To present a rare clinical presentation of BMES of the hip mimicking nonspecific LBP. To the best of the author's knowledge, this is the first case to describe hip BMES presenting as mechanical nonspecific LBP. Case presentation: This case report describes the history, examination findings, and clinical reasoning used for a patient with LBP as a chief complaint. Furthermore, the clinical presentation (i.e. pain location and its changes related to load) and the symptoms behavior (i.e. immediate symptoms decrease after few hip treatment sessions and quick worsening of the hip pain related to loading activities) after two treatment sessions increased the suspicion of an underlying medical condition of the hip joint and lead to the decision for additional evaluation. A MRI showed a serious hip BMES. Conclusions: This case report highlights the importance of including a comprehensive and continuous differential diagnostic process throughout the treatment period, looking for those risk factors (i.e. red flags) that warrant further investigation and referral to the appropriate physician. Physiotherapy diagnosis should include clinical reasoning, clinical presentation, and symptom behavior in addition to appropriate referral for medical assessment and diagnostic imaging when appropriate. Physiotherapists working within a direct access environment have the competence and responsibility to participate with other health professionals in the differential diagnose process especially for patients presenting with serious pathology mimicking musculoskeletal disorders.

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Bone marrow edema Syndrome, differential diagnosis; direct access; hip pain; low back pain; physiotherapy

Background

Back pain is a health problem which affects many people around the world and has been recently classified as the most important cause of disability (Mesner, Foster, and French, 2016). In 85% of cases, the cause of the back pain is not specific (i.e. it cannot be attributed to either specific serious diseases or nerve root irritations) (Downie et al., 2013; Maselli et al., 2016; Petersen et al., 2003); therefore, its clinical presentation is considered to be benign and can be managed conservatively. However, a very low percentage of patients (15%) suffer from back pain due to more serious medical diseases (e.g. tumor, fractures, infections, and cauda equina syndrome) (Downie et al., 2013).

In clinical practice, physiotherapists may encounter patients that complain of low back pain (LBP) that is

actually originating from the hip joint, not the lumbopelvic region. Clinical presentations of both nonspecific LBP and referred pain to the lower limb are common presentations for patients suffering from an underlying medical disease (Gleberzon and Hyde, 2006; Lishchyna and Henderson, 2004). In the early stages of a serious medical pathology, the clinical presentation can mimic common musculoskeletal disorders and therefore make the differential diagnosis more challenging for clinicians working in direct access environments (Mourad et al., 2016). In fact, nonspecific LBP can present with more severe symptoms compared to specific LBP in the early stages. This scenario can cause a delay in the correct diagnosis of a serious pathology that may need medical or surgical management. Bone marrow edema (BME) is a radiological descriptive term used for the

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first time by Wilson, Murphy, Hardy, and Totty (1988) that broadly labels all those clinical conditions characterized by spreading signal hypointensity on T1 weighted images and spreading signal hyper-intensity on T2 weighted sections. In hip joint, BME magnetic resonance imaging (MRI) findings are typically revealed on the anatomical area between the femoral head to the greater trochanter on MRI associated with pain as the primary complaint (Aigner et al, 2008b; Bilgici, Sakarya, Selçuk, and Sakarya, 2010; Patel, 2014; Santoso, Ingale, Park, and Yoon, 2017; Yi, Lee, and Kim, 2015).

This clinical presentation follows a bone fracture which causes an alteration of trabeculae combined with both the emission of interstitial fluid and hemorrhages in the medullary cavity, causing hyperemia and/ or vascular congestion, along with increasing intraosseous pressure, decreasing perfusion, hypoxia, and, consequently, the replacement of the bone marrow with an aqueous material and a poor bone mineralization (Aigner et al, 2008b; Bilgici, Sakarya, Selçuk, and Sakarya, 2010; Patel, 2014; Santoso, Ingale, Park, and Yoon, 2017; Yi, Lee, and Kim, 2015). The injury itself can have many causes (i.e. trauma, degenerative inflammatory process, infection, metabolic/endocrine, vascular, iatrogenic, and neoplastic diseases) (Manara and Varenna, 2014) that define its classification. On the other hand, the term Bone Marrow Edema Syndrome (BMES) (Patel, 2014) is used when it is impossible to determine a specific cause. However, as BMES presents proper etiopathogenesis, bioimages and prognosis this term ought to be used to distinguish this syndrome from other clinical presentation (Korompilias, Karantanas, Lykissas, and Beris, 2009). That is, BMES is a clinical-radiological finding characterized by either transitory or chronic symptoms, mostly pain in the homo-lateral hip and/or knee region, without any specific signs of avascular necrosis, trauma, or previous infection (Patel, 2014). Normally, when the femoral head-neck junction is injured, it typically refers pain to the groin region, the gluteal area, and the anterior thigh. To date, three studies have demonstrated that severe pathologies of the hip joint (i.e. fractures, osteonecrosis, and dysplasia) may refer pain to the lumbar region (Ben-Galim et al., 2007; Gleberzon and Hyde, 2006; Greenwood, Erhard, and Jones, 1998). However, to the best of the authors' knowledge, referred pain from BMES of the hip to the lumbar spine, mimicking nonspecific mechanical LBP has not been previously described in the literature. The pain onset may be either sudden or gradual with a progressive worsening of the intensity. Typically, pain gets better with rest and loading management but gets worse after overload (i.e. related to the actual tissue load capacity), causing limping and functional limitation (Hofmann, 2005; Patel, 2014).

Physiotherapists routinely assess patients whose primary complaint is LBP. The current case report highlights the importance of including system review and clinical reasoning in assessing LBP patients instead of basing the diagnosis only on imaging findings (Brinjikji et al., 2015; Nakashima et al., 2015). Failure to correctly screen for associated disorders in patients presenting with complex disorders may lead to an inappropriate management. The purpose of this case report is to increase clinicians' awareness of the signs and symptoms of BMES that can manifest as nonspecific mechanical LBP and describe the assessment, screening, and diagnostic process.

Case presentation

Patient medical history

A 49-year-old Caucasian male, construction worker (used to work 8 hours per day), presented in a direct access physiotherapy clinic, with a previous acute LBP medical diagnosis. The patient complained of stiffness and a dull back pain (Numeric Pain Rating Scale (NPRS) 6/10) mostly right-sided since approximately last month, with an insidious onset that had progressively worsened. The patient also reported right anterior deep thigh pain (NPRS 5/10) associated with a feeling of heaviness, linked to LBP symptoms (Figure 1). The symptoms appeared to get worse during working, after walking for more than 10 minutes or

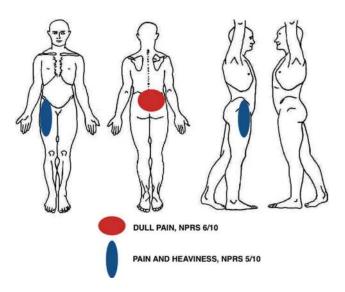


Figure 1. Symptoms at first visit. In red: LBP NPRS 6/10during working activities; in blue: hip NPRS 5/10 and heaviness.

after 30y minutes of sitting. The symptoms reportedly got better with 30 minutes of rest or during sleep.

The patient reported no previous LBP episodes. Moreover, at the review of the past medical history, the patient did not have any significant past or current medical problems or trauma; however, he had surgery for an inguinal hernia and hydrocele repair 10 years ago.

Notably, the patient reported an increase in work load during the last two months, which was temporary linked with the LBP onset. At the time, as the symptoms were progressively worsening, frequent breaks during the day were needed. The patient was unable to perform overtime work that he was normally used to. However, he refused to fully rest from work because he was worried that he may be laid off by his employer.

Examination and physiotherapy diagnosis

Neurological signs (i.e. frank motor weakness, altered reflexes, or sensory impairment) were excluded. During the observation, the patient showed a flattened lumbar lordosis associated with a kyphotic posture. The symptoms intensity increased during repeated squatting, especially during the returning phase to standing. In addition, repeated active lumbar flexion and extension movements reproduced the patient's symptoms. Palpation revealed trigger points (TrPs) over the paraspinals and right medius gluteus muscles. L1-L3 were found painful and stiff during the provocative posterior to anterior spring testing. Moreover, passive end range lumbar spine flexion revealed hypomobility and symptom reproduction. The passive internal rotation of the symptomatic hip was found to be restricted and painful; however, this finding is a common presentation in the LBP population (Sadeghisani et al., 2015). There were no signs of sacroiliac joint involvement. The patient was subsequently diagnosed as having acute LBP with referred pain to the thigh (Vleeming et al., 2008).

Physiotherapy intervention

On the first physiotherapy session, the patient was informed and reassured of his condition and prognosis and was involved in the management strategies choices. Manual therapy techniques were delivered with the goal of pain reduction and function restoration. Gentle accessory posterior-to-anterior grade III (i.e. into resistance) non-thrust mobilizations to the painful lumbar segments were performed followed by high-velocity low-amplitude thrust manipulation and mobilization with movement to the thoraco-lumbar junction. Trigger points were also manually (i.e. ischemic compression) treated in the same session. During the intrasession re-assessment, the patient reported an immediate reduction of the resting symptoms at the lumbar region (NPRS 2/10) but no changes to the lower limb symptoms.

The patient was advised to maintain an active lifestyle within pain limitations, paying attention to optimal loading strategies. Repeated active extension exercises were also prescribed with a dosage of 6-10repetition, 2-3 times per day.

On the second visit, after 3 days, the patient reported an almost complete regression of the lumbar symptoms (NPRS 1/10) that was felt only during working duties. However, no changes in the lower limb symptoms were noticed. Therefore, a more detailed examination to the lower limb was performed. Squatting, one leg load, and crossed leg were found to provoke the patient's symptoms. Active hip internal rotation and combined flexion and abduction were limited and painful. Passive hip flexion combined with internal or external rotation were both hypo-mobile (i.e. stiff) and painful. The Scour test (Sutlive et al., 2008) and Fadir test were also positive suggesting a femoroacetabular Impingement (FAI) (Zhang et al., 2015).

In order to improve range of motion and reduce pain intensity, TrPs of the medius gluteus were retreated by manual ischemic compression. Moreover, multidirectional end-range traction combined with techniques to the hip joint were performed.

The patient reported an intra-session 50% decrease of his symptoms during provocative activity. Loading management was also prescribed (i.e. avoiding overload activity such as lifting high weights and adding short rest periods during the day). However, as the symptoms seem to be driven by an arthrogenic hip issue and because of the limited improvement in pain and function following two sessions of physiotherapy, the patient was referred to the orthopedic surgeon who prescribed a pelvic X-ray.

At the third physiotherapy visit after 10 days, the patient complained of a worsening of the hip pain at rest (NPRS 3/10) and during his work duties (NPRS 8/10) (Figure 2). Moreover, the patient also described episodes of pain in the lumbar region concurrent with worsening hip pain. This worsen of symptoms may have resulted from an extra-load permitted by the temporary partial decrease of the pain level due to the previous treatment sessions. The X-ray of the pelvis revealed degenerative changes of the acetabular roof and a bilateral osteophytosis of the cotyloid notch. Moreover, on the right side, partial degenerative changes of the femoral head-neck junction were

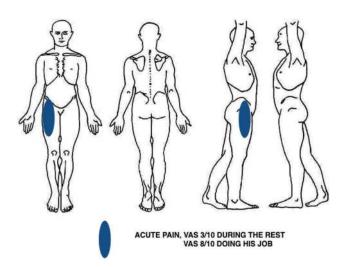


Figure 2. Symptoms at third visit. In blue: hip rest pain NPRS 3/ 10 and during loading/working activities NPRS 8/10; note the complete remission of LBP symptoms.



Figure 3. Plan X-ray of the pelvis showing degenerative changes of the femoral head–neck junction, confirming a CAM type FAI.

Table 1.	Types	of	femoroacetabular	impingement.
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	Description
PINCER	This type of impingement occurs because extra bone extends out over the normal rim of the acetabulum. The labrum can be crushed under the prominent rim of the acetabulum
CAM	In cam impingement the femoral head is not round and cannot rotate smoothly inside the acetabulum. A bump forms on the edge of the femoral head that grinds the cartilage inside the acetabulum
COMBINED	5

noticed, confirming a CAM type FAI (Figure 3 and Table 1). Given the irritability and quick worsening of the symptoms related to the increased load, the rehabilitation program was stopped and further evaluation was required using specific tests such as the Patellar

Pubic Percussion Test (Borgerding, Kikillus, and Boissonnault, 2007; Maselli, Giovannico, Cataldi, and Testa, 2014; Tiru, Goh, and Low, 2002), Percussion Test (Rahman et al., 2013), 128 Hz Tuning Fork Test (Jawad, Odumala, and Jones, 2012; Segat, Casonato, Margelli, and Pillon, 2016), and Single Leg Hop Test (Livingston, Deprey, and Hensley, 2015), which were all positive (Table 2). These tests increased concerns of a specific hip bone disease.

The patient was referred again to the orthopedic surgeon who prescribed an MRI (Figure 4) that showed a "severe alteration of the signal of the right femoral head which spreads all over the femur neck until the region of the intertrochanteric line. These findings suggest a severe BME associated with a homolateral effusion of the hip joint."

Medical management and subsequent physiotherapy

Several studies have evaluated the effectiveness of noninvasive therapies in the management of BMES (Capone et al., 2011; Flores-Robles et al., 2017; Gao et al., 2015; Suresh, 2010; Ting, Esha, and Manit, 2016). That is, the surgeon then prescribed a long period (i.e. 30 days) of full rest associated with tapentadol, clodronic acid, colecalciferol, aceclofenac, and bromelain pharmacological therapy. Pulsed electromagnetic field therapy was advised as well. At the follow-up visit, the patient's clinical presentation showed significant improvements. An additional 30 days of physiotherapy focused on progressive loading and tissue adaptation. A 60-day follow-up MRI was recommended. At the final visit, there was a complete regression of his symptoms (i.e. for both hip and back pain) with complete restoration of function. The follow-up MRI showed (Figure 5) "an almost complete regression of the BME compared with the previous MRI, although a focal edema still persists on the upper-external portion of the femur head". For a more detailed story management, see the timeline in Figure 6 (Table 3).

Discussion

BMES usually affects the hip joint, but may also involve the knees, ankles, or feet (Starr et al., 2008). As might be expected with non-weight-bearing articulations, BMES infrequently affects the upper extremities and is also rare among children (Starr et al., 2008). On the other hand, it generally affects pregnant individuals (20–40 years), mostly during the third trimester of

Table 2. Description of special tests performed.

Test	Description
Hip Scour test (Sutlive et al., 2008)	A test that screens for nonspecific hip pathology such as femoral acetabular impingement or labral tears. The patient lies supine on a table/plinth while the therapist passively flexes and adducts the hip. The therapist then applies a compressive force at the knee by applying an axial load along the longitudinal axis of the femur pushing the head of the femur into the acetabulum. The hip is then moved through an arc of flexion abduction. A positive test is resistance felt anywhere through the arc, asymmetry, and provocation of symptoms. The resistance may be caused by capsular tightness, adhesion, myofascial restriction, or labral lesion. Diagnostic accuracy for impingement/labral/intra-articular test: sensitivity = 50% (26–74), specificity = 29% (12–51) LR +/LR = .71/1.72
Fadir test (Zhang et al., 2015)	A test that screens for anterior–superior impingement syndrome, anterior labral tear and iliopsoas tendinitis. The patient lies supine on a table/plinth while the therapist passively brings the patients hip into full flexion, lateral rotation, and full abduction as a starting position. The therapist then brings the hip into extension while combining a medial rotation with adduction motion. A positive sign is provocation of symptoms with or without a click. Diagnostic accuracy for FAI, labral tear: sensitivity = 99%, specificity = 25%, LR+/LR-1.3/0.04 (labral tear)
Patellar Pubic Percussion Test (PPPT) (Borgerding, Kikillus, and Boissonnault, 2007; Maselli, Giovannico, Cataldi, and Testa, 2014; Tiru, Goh, and Low, 2002)	The test is a form of osteophony or auscultatory percussion which is used in the assessment of bone integrity by analyzing its vibrations through the use of a stethoscope and bony prominence percussion. The patient lies in supine and the therapist helps them to find their pubis symphsis and haves them hold the bell of the stethoscope on it. The therapist then makes sure the legs are in a symmetrical neutral position. While stabilizing the leg and patella in a neutral position the therapist then percusses the patella and listens for a change in pitch and loudness produced. A positive sign will be a dull and diminished sound compared to the unaffected side. Diagnostic accuracy: sensitivity = 94%, specificity = 95%, LR+/LR = 10.4/0.06.
Percussion Test (Rahman et al., 2013)	To assess the presence of fracture in the lower extremity. The patient lies in supine. After having found the great trochanter of the patient, the therapist strikes it with the second and third fingers of the hand. A positive test is reproduction of the patient's worst pain. Diagnostic accuracy: unknown
128 Hz Turning Fork Test (Jawad, Odumala, and Jones, 2012; Segat, Casonato, Margelli, and Pillon, 2016)	Test for the presence of a stress fracture. The patient lies in sitting or supine. The physical therapist places a 128 Hz tuning fork on the suspected site of the stress fracture. A positive test is reproduction of the patient's worst pain. Diagnostic accuracy: unknown
Single Leg Hop Test (Livingston, Deprey, and Hensley, 2015)	To assess for a fracture in the lower extremity. The patient lies in standing. The patient hops up and down on the affected limb several times barefoot. A large amount of pain in a localized area of the lower extremity is a positive test and may signify a fracture. Diagnostic accuracy: unknown

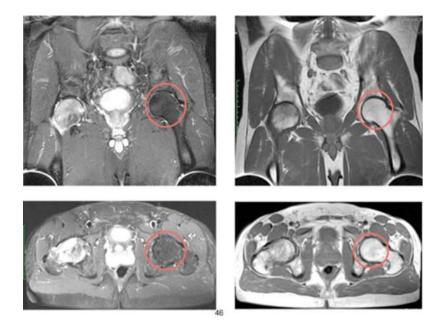


Figure 4. Frontal and transverse T1 and T2-weighted MRI of the pelvic region showing a severe bone marrow edema of the right femoral head.

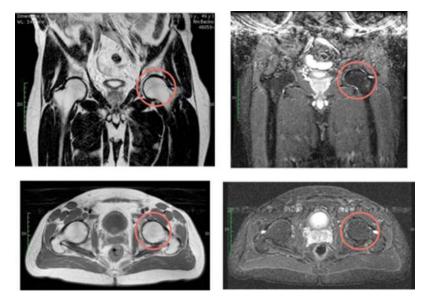
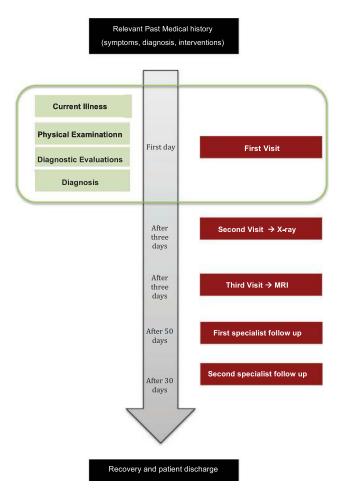


Figure 5. Frontal and transverse T1 and T2-weighted MRI of the pelvic region showing a complete regression of the bone marrow edema compared with the previous MRI.





pregnancy, and male adults over 30 or 60 years (Starr et al., 2008).

When the hip joint is affected by pathology or dysfunction, it normally refers pain to the groin, gluteal, or anterior thigh region and in severe presentations is associated with limping and functional limitations. Often the tapping test is a positive finding in these patients (Aigner et al., 2008a).

The most common clinical presentation is spontaneous mechanical pain and symptoms are strongly related to load. Complete rest is usually the best strategy in order to reduce pain and disability. The onset of BMES can be sudden or insidious with a progressive worsening if not properly managed. Frequently BMES causes joint stiffness, and in severe presentations, patients experience a hospitalization as the symptoms can be extremely disabling.

Nonspecific back pain is probably the most frequent musculoskeletal disorder that physical therapists may encounter (Mesner, Foster, and French, 2016). Although the natural history of LBP tends to be favorable, as primary care clinicians, physiotherapists need to be able to screen for bone disorders that may need urgent orthopedic medical management rather than exercise or manual therapy interventions. Musculoskeletal symptoms that mimic nonspecific LBP could be either the initial phase of a more serious pathology or secondary symptoms of an even more serious disease (Downie et al., 2013; Mesner, Foster, and French, 2016; Negrini et al., 2006). Failure by the physiotherapist to correctly screen for conditions outside physiotherapist's scope of practice could cause a delay in proper management.

In this case report, the persistence of hip pain following a failing physiotherapy management to the lumbar spine, the behavior of the symptoms in response to weight bearing movements (e.g. return from squatting

Evolution		Time 0	Time 1	Time 2	Time 3	Time 4	Time 5	Time 6	Time 7
Day		Day 1: initial physiotherapy visit	Day 1: re-assessment after first physiotherapy	Day 4: second physiotherapy session	Day 4: re-assessment after second physiotherapy session	Day 14: third visit that leads to refer the patient	Day 20: orthopedic surgeon visit	Day 50: progressive load	Day 80: final visit 60 days MRI
NPRS (0-10)	Back pain		2/10	1/10	I	I	I		0/10
	Thigh pain	5/10	No change	No change	2–3/10	8/10	I	Progressive symptoms reduction	0/10
TrPs		<i>paraspinals</i> and right medius	I	I	I	I	I	I	I
PA		gluteus muscles L1 to L3 were found painful and	I	I	I	I	I	I	I
Passive ROM	Lumbar	Hypomobility and symptom	I	I	I	I	I	I	Painfree
	Right hip	reproduction Restricted and painful hip internal rotation	I	Flexion combined with internal or external rotation were both hypo- mobile (i.e. stiff) and painful	I	I	I	Painfree	Painfree
Right hip active ROM		I	I	Internal rotation, combined flexion and abduction were limited and painful	I	I	I	I	Painfree
Special hip tests	Scour test Eadir test	I	I	Positive	I	I	I	I	I
Differential diagnosis	PPPT		I		I		1	I	
tests	Percussion test	I	I	I	I	Positive	I	I	I
	128 Hz tuning fork test	I	I	1	I	Positive	I	I	I
	Single leg hop test	I	I	I	I	Positive	I	I	I
MRI	<u>-</u>	I	1	I	I	I	BME	I	Almost complete regression of

to standing), and the onset and the complaints of LBP related to hip pain informed the physical therapist's clinical reasoning to specifically examine the hip region as a probable source of the pain. After a first suspicion of FAI, the symptom behavior (i.e. hip pain persists despite the improvement of LBP symptoms) led to contacting the referring physician for additional imaging examination that revealed a serious hip BMES. Although X-ray documented a FAI, this condition was not the primary pain generator related to the patient's symptoms. The FAI was still present on X-ray imaging after hip symptom resolution due to a successful management of the BMES. It is well documented in the literature that altered findings on imaging cannot be reliably interpreted as the primary pain generator if they do not directly correlate to the findings on clinical examination of the patient (Brinjikji et al., 2015; Nakashima et al., 2015). The proper referral to an orthopedic physician for additional diagnostic imaging and examination was essential in order to identify the correct diagnosis and select the best management of this complex hip disorder that was mimicking LBP.

Conclusions

The aim of this case report was to describe the physiotherapist's perspective of the relevant findings from history and physical examination in a patient with a serious pathology that was mimicking LBP. The relevant aspects of BMES screening, pathophysiology and differential diagnosis were discussed and the unpredicted symptom pattern behaviors, that led to a proper second referral, were outlined. This case report describes the clinical presentation and the clinical decision-making process that led a physiotherapist to suspect a different pain source than that diagnosed by a physician based on imaging interpretation. This case also supports that physiotherapists are able to screen pathologic medical conditions in direct access settings and are able to identify when a patient's clinical presentation is outside of their scope and in need of additional medical referral for further investigation (Ojha, Snyder, and Davenport, 2014; Pendergast, Kliethermes, Freburger, and Duffy, 2012). The inconsistency of the clinical presentation to the physician's diagnosis led the physical therapist to contact the referring physician twice to suggest the need for additional diagnostic imaging examination in order to guide therapeutic treatment and prognosis. Thus, the collaboration between health-care professionals is recommended for patients presenting with complex pathologies.

Declaration of Interest

The authors declare no conflict of interest.

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