Skeletal Radiology (2019) 48:1119–1123
https://doi.org/10.1007/s00256-018-3117-z

CASE REPORT

Medial iliac stress fractures in athletes: report of two rare cases: review of literature and clinical recommendations

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Received: 24 September 2018 / Revised: 9 November 2018 / Accepted: 12 November 2018 / Published online: 28 November 2018

Abstract

Stress fractures are a common diagnosis in sports medicine and can result in significant loss of function, athlete playing time, and potentially lead to chronic symptoms. However, unusual locations of stress fractures may present with vague symptoms and a relatively benign physical exam, leading to difficulty in arriving at the correct diagnosis. Pelvic stress fractures are less common than lower-extremity stress fractures in athletes, occurring in only 1–5% of all stress fractures and typically occur in pubic rami. Furthermore, iliac bone stress fractures are even rarer, with only a few case reports in the literature. Their presentation can easily be missed on routine workup and imaging. We present two cases of the very rare superomedial iliac bone stress fracture in athletes, an unusual location for this uncommon stress fracture. We review the available literature on this condition and provide clinical commentary on workup and treatment recommendations.

Keywords Stress fractures · Athletes · Ilium · Pelvic bones · Leg injuries

Introduction

Stress fractures of the pelvis and lower extremity may be difficult to diagnose and can represent a challenging patient population to counsel and treat. With the growing participation in athletics and exercise, stress fractures must be high on the differential diagnosis for anyone presenting with exertional extremity pain that has not improved with conservative measures. Pelvic stress fractures are rare in athletes and represent only 1.3–5.6% of all stress fractures [1]. The superomedial iliac stress fracture is an exceedingly rare type of pelvic stress fracture in athletes; there are only a few cases reported in the literature [1–4]. We present two cases of superomedial iliac fatigue stress fractures occurring in runners and review the literature. Physicians need to be aware of this diagnosis and location as it may be missed on routine workup and can be mistaken for "nonspecific low back pain." Overlooking this diagnosis can lead to functional impairment, lost playing time, and chronic symptoms. We provide clarification on diagnosis of this type of stress fracture, radiological workup, and treatment options. Lastly, in the age of increasing interventional injection options for low back pain, we provide clinical commentary and precautions for anyone presenting with similar symptoms to our cases.

Case report #1

A 35-year-old female elite marathon runner (5′7″, 106 lb., BMI 17.6) presented with persistent left sacral and sacroiliac (SI) joint region pain in February of 2018. She was a professional marathon runner, competing for the last 10 years, and reported no prior change in training regimen (specifically no sudden increase in intensity or volume) or change in diet. She denied any major orthopedic injury or illness and has never had interruptions in training or competition for the last 10 years. For the last 1 month prior to presentation, she noted onset of mild lower back pain, initially diffuse in nature. She was able to continue

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training through the symptoms and commenced training for a major marathon. She decided to try running at altitude outdoors, however, due to winter weather, she was forced to run in snow and slippery conditions. Later she switched to a treadmill to log more hours, which was not her customary training style. The patient engaged in this new training format for approximately 2 weeks.

Approximately 2 weeks prior to presentation, she slipped without actually falling during a run and felt an increase in aching low back pain. She sought outside medical attention and treatment with a physical therapist and was diagnosed with left SI joint dysfunction. Her symptoms were tolerable at the time, and she decided to go on an overseas trip for an international race. After landing from a > 10-h plane flight, her lower back pain acutely worsened. She rested for a day, however symptoms persisted. The patient tried to compete in the race but withdrew due to pain. Upon returning home, she requested further evaluation, presented to clinic, and an MRI was obtained. The MRI demonstrated an incomplete stress fracture of the medial iliac bone with associated marrow edema (Fig. 1).

The patient took 2 weeks off entirely from any exercise; she was offered crutches to limit weight-bearing but declined. She began cross-training with bodyweight unloading in the pool. After 4–6 weeks of relative rest, she gradually restarted running again. For the next 3 months, she resumed formal training in a progressive fashion. At 4 months after the fracture was diagnosed, she was able to run competitively again.

Case discussion

This case demonstrates an unusual location for an iliac stress fracture. Iliac bone stress fractures are not common, accounting for only 4% of stress fractures in one study [5] and do not typically occur in the superomedial iliac region [5–7]. In any long-distance runner with a stress fracture, the signs and symptoms of the female athlete triad [8] must be investigated, but were not present in this case. It was believed that the abrupt change in training regimen (the patient was not accustomed to running in snow or treadmill running) was the causative factor. Treatment of stress fractures typically involves rest from the offending activity, time, addressing training errors, and assessment or potential risk factors [9]. Upon eliminating the training change and with appropriate rest, this patient was able to successfully recover and resume full training again.

Case report #2

A 36-year-old female (5′ 7″, 139 lbs, BMI 21.9) presented to a university pain clinic in January 2015; she was an avid runner, doing numerous 5K, 10K, and half-marathons a year, with the desire to "keep running no matter what to keep the weight off." She first noted right hip symptoms and was told she had trochanteric bursitis. She was training for a 10K at the time, proceeded to run through the pain and complete the

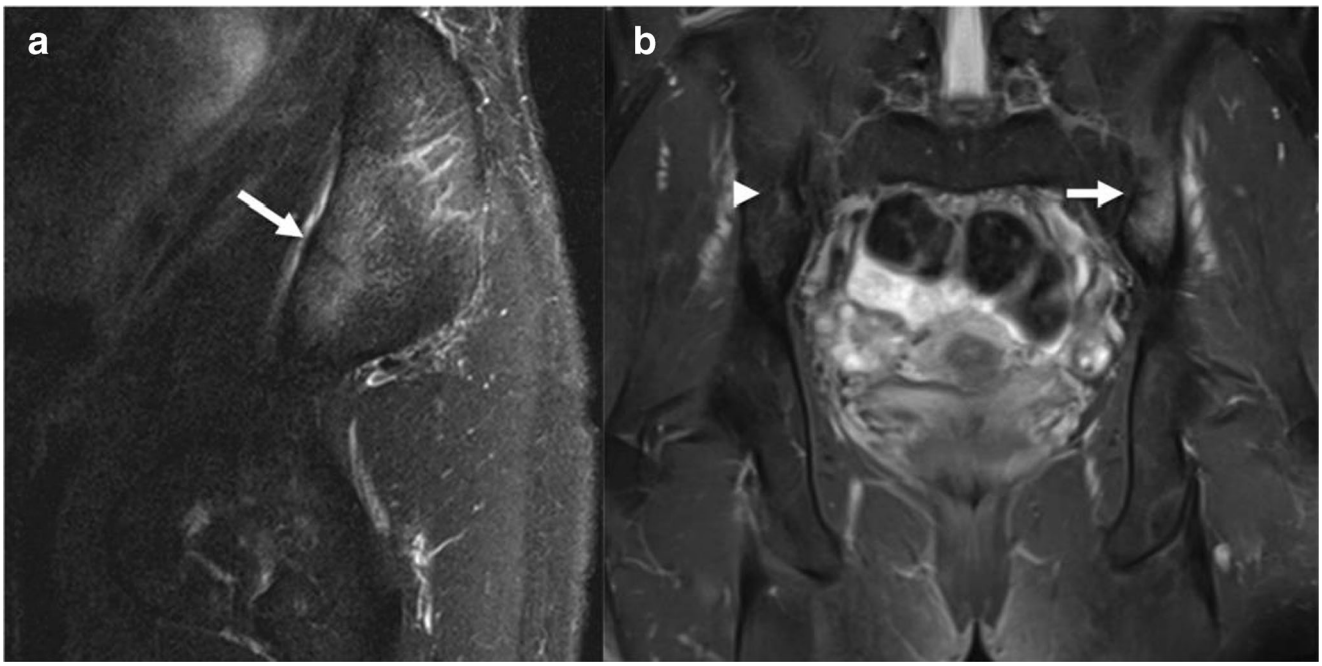

Fig. 1 Case #1 MRI. **a** Sagittal T2-weighted fat-suppressed sequence through the medial aspect of the left iliac bone demonstrates an incomplete, low-signal fracture line (*arrow*), with regional bone marrow edema. **b** Coronal proton-density fat-suppressed (fluid-sensitive) image through the entire pelvis shows asymmetric bone marrow edema in the left iliac bone, and shows the relationship of the fracture line with the SI joint (*arrow*). The patient had minimal bone marrow edema on the contralateral side, however without a fracture line (*arrowhead*)

10K race but then experienced severe right lower back pain radiating into her right thigh to the knee. In March 2015, her symptoms progressed to right lower back pain and she was diagnosed with right SI joint dysfunction.

Over the next 2 years, she was given several diagnoses, from lumbar disc herniation, radiculopathy, "sacral disorder" and spinal stenosis in 2015 to lumbar spondylosis and chronic pain syndrome in 2016. The patient was adamant about continuing to train for marathons. Throughout this period, she took several pain medications including hydrocodone, tapentadol, tramadol, amitriptyline, and duloxetine, and proceeded with (in order) the following procedures over 2015–2016: right L3-L5 medial branch block, right SI joint steroid injection, bilateral lumbar radiofrequency ablation, left SI joint injection, repeat left bilateral lumbar radiofrequency ablation, left ischial bursa injection, and two more repeat bilateral lumbar radiofrequency ablation procedures. Imaging evaluation consisted of a lumbar spine MRI that showed minor disc disease at the L5-S1 and radiographs, which showed stigmata of remote healed bilateral sacral stress fractures with some residual sclerosis. At some point, the patient was eventually lost to follow-up but continued with further spinal procedures, undergoing a total of 13 procedures in 20 months.

The patient again presented to the sports medicine clinic in 2017 when she "threw out her back" and came to clinic in severe pain and asking for a hip joint cortisone injection, as she thought she had a labral tear. She had done numerous races over the last year and her pain was increasing; her symptoms were now interfering with daily activities. On exam, she had normal hip range of motion but pain with end-range flexion and internal rotation suggestive of intra-articular pathology. She also reported symptoms over the right posterior iliac crest region and had positive right SI joint tenderness. Lumbosacral and hip radiographs that day showed no acute osseous abnormality (and no new abnormal sacral sclerosis). An MRI arthrogram of the right hip was obtained, and while it did show an anterosuperior labral tear, there was bone marrow edema in the right iliac bone anteroinferior to the SI joint, with a linear hypointensity from the inferior iliac bone adjacent to the SI joint extending to the superior iliac wing, compatible with a nondisplaced superomedial iliac stress fracture (Fig. 2).

The patient was counseled to limit weight bearing, avoid impact exercise, start physical therapy, and warned of potential fracture progression; also, female athlete triad symptomatology was discussed. She did obtain a DXA scan, which showed lumbar spine Z-score −2.0 (borderline abnormal), right femur −1.6, and left femur −1.1, which progressed compared to a prior DXA (Z-scores were used because patient was premenopausal). The patient kept training despite counseling, and in fact began full marathon-distance training.

Once again, she was lost to follow-up. She saw an orthopedic surgeon the following month with continued symptoms, and was reluctant to stop running since she had an upcoming marathon in 11 weeks. The surgeon ordered repeat X-rays showing a vague band of sclerosis along the central aspect of the right iliac bone adjacent to the SI joint (not visible on the prior study 2 months ago) corresponding to the stress fracture (Fig. 2); repeat MRI showed there was continued bony edema. She was not permitted to run, and after the second MRI, she did adhere to this. With a clear diagnosis, she also weaned off pain medications she was taking for her "chronic low back pain" diagnosis. The patient moved out of the region the following month and was ultimately lost to follow-up.

Case discussion

This case represents an unusual location for a pelvic stress fracture and highlights the challenging nature of the endurance

Fig. 2 Case #2 MRI and radiograph. **a** Coronal proton density image of the right hip demonstrates an incomplete, low-signal fracture line (*arrow*). **b** Corresponding coronal T2-weighted fat-suppressed image shows the regional bone marrow edema surrounding the fracture line. Fluid in the hip joint is from a contrast injection (*asterisk*). The patient also had a labral tear (not shown). **c** Pelvic radiograph performed 1 month after the MRI shows a vague band of sclerosis in the medial aspect of the right iliac bone (*arrow*). There is no sclerosis in the contralateral iliac bone, or on radiographs performed approximately 2 months earlier (not shown)

athlete mindset. The case also raises several questions. Multiple chronic issues and baseline chronic pain can confound the diagnosis if the stress fracture is the current pain generator; however, we believe that due to the acute and new pain, in the sacroiliac region, in the clinical context of numerous recent races, the stress fracture was indeed the cause of her new pain. We question if repeated spinal procedures (14 procedures in less than 2 years) could have played a role in her stress fracture, with postulated mechanisms including local cortisone effect on regional bone, and/or reduced nociception from repeated radiofrequency ablations, and the impaired ability to sense the bony pain of an impending/developing stress fracture. We also question if her new posteromedial iliac crest region pain from the stress fracture was simply missed, and the pain was misinterpreted as SI joint pain or lumbar spondylosis and further spinal procedures were directed at this. Furthermore, the patient had previously been diagnosed with a sacral stress fracture that should have alerted both patient and provider of potential risks for another fracture. A final point is that the patient's bone mass was slowly decreasing over time; however, the patient had not been further counseled on osteopenia/osteoporosis, evaluated by endocrinology, or tried any other potential interventions. Although her Z-score was considered within normal limits, at − 2.0, it was borderline for her age and sex.

Discussion

Superomedial iliac stress fractures are a relatively rare type of stress fracture. In review of the literature, it has been associated with long distance running, and appears more common in females [1–4] as in our cases. The classic female athlete triad of amenorrhea, osteoporosis, and eating disorders is suggested when this stress fracture presents in clinic [8]. It is usually treated conservatively with rest and activity modification. Patients may restart exercise gradually after several asymptomatic weeks. NSAIDs may be taken for this, but they do raise potential risk of delayed bone healing in animal models [10–12].

While originally reported in military recruits, fatigue stress fractures are very common in sports medicine, accounting for up to 20% of all sports medicine clinic injuries [13]. The most common reported sites include the tibia, fibula, and metatarsals, and runners (especially track and field athletes) have the highest reported overall incidence [14]. Liong and Whitehouse studied lower limb stress fractures in the athlete and concluded that pelvic stress fractures represent only 1.3–5.6% of all stress fractures [1]. Common sports associated with pelvic stress fracture were mainly long-distance runners and gymnasts, with cases also reported in soccer players.

Most fatigue fractures occur in the lower extremities. Sites can be classified as high risk (e.g., femoral head, anterior cortex [tensile side] of tibia, navicular [tarsal], and proximal 5th metatarsal shaft) or low risk (e.g., pubic rami, sacrum, fibula, and 2nd to 4th metatarsal shafts) reflecting potential for fracture propagation, displacement, or non-union [15–18].

Fatigue stress fractures of the pelvis, as in the two cases discussed here, are considered low risk. Pelvic stress fractures are rare and may be missed on routine imaging; they represent 1.3–5.6% of all stress fractures [1], up to 7.1% in another study [9]. They have been reported in athletes and military recruits, and among women [2–4]. They usually occur in the pubic rami, and present with inguinal, pelvic, or groin pain.

The rare superomedial iliac stress fracture

There are only a few cases of superomedial iliac stress fractures reported in the literature. In a study by Battaglia, a 58-year-old female amateur marathon runner (BMI unknown) presented with a 1-month history of worsening pain in the right buttock [2]. MRI performed 1 month after the onset of symptoms and CT performed 2 months after the onset of the symptoms showed a fracture line in the right superomedial ilium, extending to the SI joint, surrounded by a sclerotic area, pathognomonic of fracture, with bone reparative changes. A DXA scan was reportedly normal. The patient was treated for 4 weeks with rest and nonsteroidal anti-inflammatory drugs (NSAID), and calcium supplementation to favor the healing process. Once she had been asymptomatic for three additional weeks, she gradually restarted her running activity.

Touhy et al. reported a 17-year-old Caucasian male collegiate track athlete (BMI 19.5), nationally ranked in high school, presenting with persistent left gluteal and sacral pain for 2 months and exercise-associated right groin tightness [4]. The intensity of his left-sided pain was rated 6 out of 10 in severity. A DXA showed a Z-score of − 1.43, within normal limits for his age. However, it was later found that the patient was HLA B27-positive and developed bilateral sacroiliitis 5 months after the fracture healed, and he was ultimately diagnosed with ankylosing spondylitis.

Finally, Amorosa reported a 24-year-old female (BMI 23) with history of a healed tibial shaft stress fracture 2.5 years prior and a sacral stress fracture 1.5 year prior, both previously treated with rest, presenting 1 day after completing a marathon with reports of right-sided lateral hip pain [3]. The patient reported moderate right hip pain several days before the race and self-medicated with NSAIDS. The pain became severe during the race, but she finished the raced walking. She had a history of oligomenorrhea and a lifestyle pattern consistent with exercise bulimia. An MRI was ordered due to concern for a labral tear, which showed an isolated stress fracture of the right iliac wing, confirmed on a subsequent CT examination. This fracture was unusual in that it did not involve the SI joint or the acetabulum, but involved the iliac wing and fossa, resulting in a complete extra-articular fracture. A DXA was

never performed in this patient, despite referral to a metabolic bone disease specialist.

Stress fractures typically present with a history of progressive focal or nonspecific extremity pain, in the absence of major trauma. Physical exam is classically nonspecific, with findings limited to vague tenderness, possible focal swelling, but often the exam is benign. Imaging is critical in the diagnosis of stress fractures. The initial imaging workup should start with radiographs of the area of interest because of its low cost and availability [19–22]; however, the sensitivity of fracture detection—particularly subtle or early fractures—is low and more sensitive imaging should be considered [15, 19, 21–23]. The American College of Radiology (ACR) recommends MRI without contrast as the imaging study of choice after negative radiographs [20]. MRI allows the detection of stress fractures at very early stages and will, at times, demonstrate a T1 and T2 hypointense fracture line within an area of marrow edema [2, 17, 21, 23].

Conclusions

We present two cases of a rare superomedial iliac fatigue stress fracture in female marathon runners. They both presented with vague low back pain complaints and the diagnosis was not initially clear. As a result, they both continued to run through the symptoms, potentially worsening the extent of their injuries. We recommend having a low threshold of initiating imaging with MRI for patients with a similar presentation. In the modern era of easy access to pain clinics and numerous spinal procedures, interventional physicians performing injections in the low back and SI joint region must also be aware of the possibility of this rare fracture and its presentation.

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