

Pubic stress fracture presenting as a strain of adductor longus in a 16-year-old elite soccer player with Crohn's disease: a case report

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Background: *Adductor strains are the most commonly reported muscle injuries in adolescent soccer players and the second most common muscle injuries in adult players. Health practitioners should be aware of possible differential diagnoses, such as a pubic stress fracture or pubic apophysitis when athletes present with chronic groin pain.*

Purpose: *To present a rare case of a unilateral pelvic stress fracture of a 16-year old elite soccer player with a history of Crohn's disease.*

Study design: *Retrospective case report (n=1).*

Methods: *Case notes of two sports-based practitioners were reviewed and compiled retrospectively.*

Results: *Following activity restriction, a period of rest, conservative care, and progressive rehabilitation, this athlete was able to achieve a pain-free state with near equal iso-kinetic strength bilaterally as measured*

Contexte : *Les entorses des adducteurs sont les blessures musculaires les plus fréquemment signalées chez les joueurs de soccer adolescents et les deuxièmes blessures musculaires les plus fréquentes chez les joueurs adultes. Les professionnels de la santé devraient connaître les diagnostics différentiels possibles, comme une fracture de stress du pubis ou une apophysite pubienne lorsque les athlètes présentent des douleurs chroniques à l'aine.*

Objectif : *Présenter un cas rare de fracture de stress pelvien unilatéral chez un joueur de soccer d'élite de 16 ans ayant des antécédents de maladie de Crohn.*

Plan d'étude : *Cas clinique rétrospectif (n=1).*

Méthodologie : *Les notes de cas de deux praticiens du sport ont été examinées et compilées rétrospectivement.*

Résultats : *Après une période de restriction d'activité, une période de repos, des soins conservateurs et une rééducation progressive, cet athlète a pu atteindre un état indolore avec une force isocinétique bilatérale presque égale, mesurée par le test musculaire Cybex 6000 (Cybex International Inc., Medway,*

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by Cybex 6000 (Cybex International Inc., Medway, MA, USA) muscle testing. Full activity was resumed 10 months after initial presentation and the athlete was able to return to playing professional soccer.

Summary: *This case report presents a rare diagnosis of a unilateral pubic stress fracture presenting as a strain of adductor longus. Although quite rare, differential diagnoses such as a potential underlying stress fracture should be considered when presented with chronic or recurrent groin pain.*

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KEY WORDS: chiropractic, stress fracture, pubic bone, groin pain, soccer injury

Introduction

According to the Federation Internationale de Football Association (FIFA), soccer is the most popular sport in the world with over 265 million players worldwide. In FIFA's 2011 "Big Count" study, Canada ranked 22nd in terms of athlete participation in soccer with an estimated 2.7 million participants. The United States ranked second, behind China, in athlete participation with 24 million (fifa.com). The sport of soccer involves running, kicking, jumping, and cutting - resulting in a number of lower limb injuries.¹⁻³ Groin injuries account for 5% to 18% of all soccer injuries and frequently present diagnostic challenges for practitioners.^{2,4-5} Strains of any of the three adductor muscles represent the most common cause for soccer-related groin injury. A 10-year prospective cohort study of elite French adolescent (age 14 to 16) soccer teams, representing 237,600 hours of exposure time, demonstrated that muscle strains were the third most common injury behind contusions and sprains (mostly of the ankle). Of these muscle strains, the groin was the most common location of injury making it a very common and likely diagnosis for an athlete presenting with groin pain.^{3,6-8} Possible differential diagnoses for groin pain include osteitis pubis, inguinal disruption⁹, apophysitis and in rare cases pelvic stress fractures.

Stress fractures can be divided into the following types: fatigue fracture, insufficiency fracture, and a combined

MA, USA). *L'activité sportive complète a repris 10 mois après la présentation initiale et l'athlète a pu recommencer à jouer au soccer à un niveau professionnel.*

Résumé : *Ce cas clinique présente un diagnostic rare de fracture de stress pubien unilatéral se présentant sous la forme d'une lésion du moyen adducteur. Bien qu'ils soient assez rares, les diagnostics différentiels tels qu'une fracture de stress sous-jacente potentielle devraient être pris en considération dans le cas d'une douleur chronique ou récurrente à l'aîne.*

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MOTS CLÉS : chiropractie, fracture de stress, os pubien, douleur à l'aîne, blessure au soccer

type – fatigue and insufficiency fracture. Fatigue fractures are thought to develop through an accumulation of micro trauma and the inability of the bone to keep up with skeletal repair.¹ Insufficiency fractures develop when there is inadequate bony remodeling in response to normal stressors^{1,6-8} and these insufficiency fractures tend to occur with Relative Energy Deficiency in Sport (RED-S) previously called the Female Athlete Triad¹⁰, metabolic disease and osteoporosis. In sports medicine, stress fractures of the pelvis are far less common than lower extremity stress fractures, and only account for 1% to 7% of stress fractures.^{3,6-8} These fractures are considered low-risk and have been reported mainly in long-distance runners and female military recruits.^{3,6-8} There are many factors that contribute to the development of stress fractures. Extrinsic factors include training regimen, footwear, training surface, and type of sport.^{1,6-8} Intrinsic factors include biomechanics, anatomy, hormones, and nutrition.¹ The third type of stress fracture is a combined fatigue and insufficiency fracture, and pertains to the following case.

This is a case report of a sixteen-year-old national level male soccer player presenting with a seemingly recurrent episode of adductor longus strain. The player had been diagnosed with Crohn's disease six months prior to presentation. Advanced diagnostic imaging later revealed a stress fracture of the left superior pubic ramus with associated evidence of inguinal disruption. Our motivation for

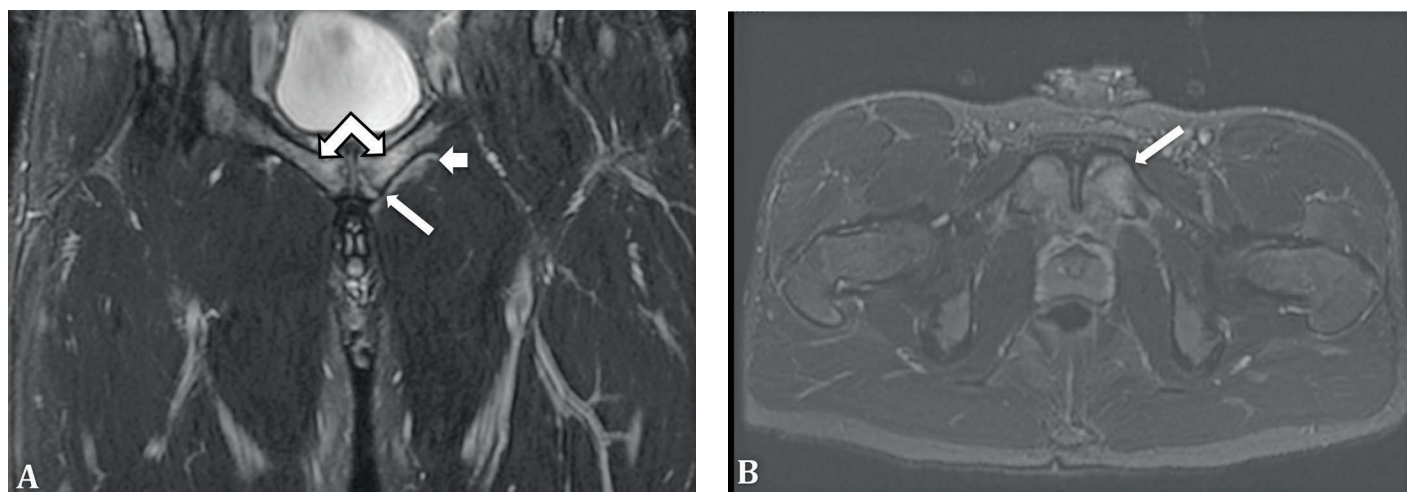


Figure 1.

Sagittal (A) and Axial (B) T2 weighted MR images displaying bilateral bone edema on both sides of the pubic symphysis (double arrow) with a small amount of fluid in the joint (Osteitis Pubis). There is a dark signal cleft indicating a non-displaced fracture of the left superior pubic ramus (thin arrow). There is also increased signal intensity within the left adductor longus tendon (thick arrow) and tendon thickening (Tendinopathy). MRI taken in December of 2011.

this study is to provide clinical awareness to healthcare providers of the rare, but possible differential diagnosis of pubic stress fractures in athletes with chronic groin pain.

Case presentation

A sixteen year-old male national level soccer player presented to a sports medicine clinic with left groin pain one week following an injury sustained during a soccer game in which the player was tackled. He was able to walk off the field without assistance following the tackle. The pain on presentation was located over the region of the left groin and radiated around to the gluteal region on the ipsilateral side. He also described paresthesia in the posterior left thigh. He had injured his left groin several times with repeated strains in the past. All resolved successfully with conservative management. The patient reported no pain with coughing, sneezing, or straining. His medical history included a diagnosis of Crohn's disease by his family physician six months earlier, for which he was taking low-dose oral methotrexate (once a week). He was in a period of remission at the time of presentation.

Physical examination revealed full range of motion of the left hip with pain only at the end ranges of abduction as well as hip extension. There was also pain with manual

resisted testing of adduction as well as internal rotation of the left hip. Examination of the low back was unremarkable as was neurological evaluation of the lower limb. Palpation revealed tenderness over the pectineus and adductor longus muscles. Isokinetic muscle testing was performed using the Cybex 6000 (Cybex International Inc., Medway, MA, USA). The patient reported being right leg dominant. Testing was performed at 30 degrees/second. Five repetitions were performed and the average of these five repetitions was recorded as 110 foot pounds of force on the right for adduction and 98 foot pounds of force on the left for adduction. In comparing the force generated from the right to the left leg a 22% strength deficit on the left with adductor strength testing during the initial assessment was identified. Isokinetic testing of the hamstrings and quadriceps using the Cybex 6000 did not reveal strength deficits comparing right to left.

The patient was diagnosed with a suspected strain of the left adductor longus and conservative treatment was initiated consisting of relative rest, interferential current (IFC), cluster laser, and ice. He was also referred to a general surgeon who specializes in sports injury management two days later for a second opinion and a diagnostic ultrasound. Following his examination, the surgeon concurred

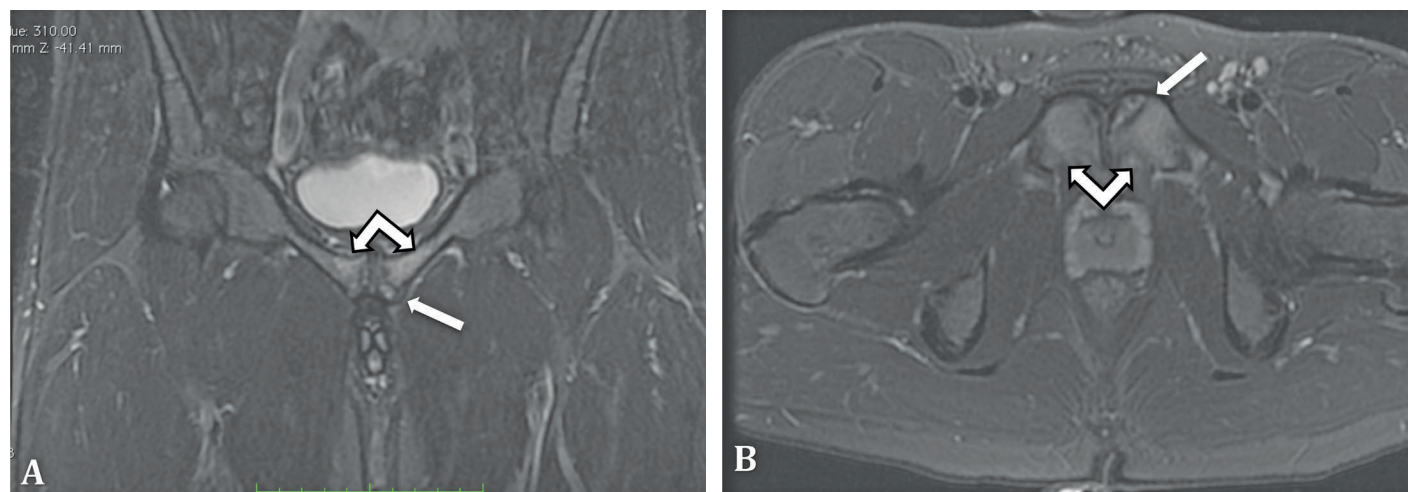


Figure 2.

Second MRI taken March 2012. Image A is the sagittal view and Image B is the axial view. There is again prominent T2 signal involving the pubic symphysis on bilaterally (double arrow). There is a dark signal cleft identified in the left aspect of the pubic symphysis (single arrow) which the radiologist considered suspicious for an apophysitis. There is no evidence of significant tendinopathy or tears within either groin.

with the suspected diagnosis and ordered a bilateral diagnostic ultrasound of the groin region. Ultrasound examination revealed elongated lymph nodes in the left groin region with no sign of muscle tear or hernia. Due to operator dependency of diagnostic ultrasound as an imaging modality, the patient was still suspected to have a minor tear of the adductor longus and was referred back to the sports specialist chiropractor for continued therapy.

Treatment continued consisting of IFC, laser, ice, as well as active rehabilitation of the pelvic musculature for approximately three weeks at which point the patient reported approximately 75% improvement in symptoms, but was still having trouble with full speed running and kicking the ball. A follow-up appointment was scheduled with the general surgeon who then ordered a follow-up diagnostic musculoskeletal ultrasound. Once again, results showed no evidence of a tear or hernia. MR imaging was subsequently ordered and scheduled for one month later. Despite continued therapy and rehabilitation, the patient plateaued at 75% improvement and saw no change over the following month.

MR imaging of the pelvic and groin regions revealed a non-displaced fracture of the left superior pubic ramus extending to the symphysis pubis. This was associated

with bone edema of both pubic rami and a small amount of fluid in the symphysis pubis joint which was consistent with osteitis pubis. There was also increased signal within the left adductor longus tendon and tendon thickening in keeping with a tendinopathy. There was no definite tear identified in the adductor or abdominal musculature (Figure 1). Upon receiving the results from the radiologist, all attempts at sport activity and rehabilitative exercises were ceased. The patient was placed on limited weight bearing with crutches to ambulate and continued receiving conservative treatment for his injury.

Follow-up

Following the MRI diagnosis of stress fracture the patient was removed from all sports activity. The general surgeon opted for a non-surgical method of care and the patient continued with conservative care from the sports chiropractor and a certified kinesiologist. Conservative management consisted of rest, electromagnetic therapy (Theramend, Nortek Medical Devices) using a setting for osteoporosis and bone healing, interferential current (IFC), clustered infrared laser, and home icing. Treatment was delivered at a frequency of three times per week for twelve weeks, at which point the patient was reporting

significant improvements. Follow-up MR imaging was done at this time and compared to the previous MRI.

Multiplanar multisequence MR imaging of the pelvis revealed a high T2 signal within the pubic symphysis bilaterally. There was a dark signal cleft identified in the left aspect of the pubic symphysis which was considered suspicious for an apophysitis at the pubic symphysis by the reporting radiologist. There was no evidence of tendinopathy or tear within the groin or abdominal musculature (Figure 2).

Due to incomplete healing at this time, the patient continued with conservative care under the advice of the general surgeon. Cybex strength testing was performed one month later and the patient was showing significant improvements having only minimal deficits comparing right to left in adductor strength. Using the same parameters as on the initial Isokinetic Cybex assessment the values were 138 foot pounds generated on the right and 124 foot pounds generated on the left showing a residual 10% deficit on the left. Treatment and rehabilitation continued for the next month at which point the patient had no pain and no deficits in strength and was permitted to fully return to play. The full recovery for this patient from presentation to full return to play was approximately ten months. This patient currently just finished his first full season of professional soccer in Europe with no re-aggravation of his condition and no reports of groin pain.

Discussion

The groin is a common source of pain in rotational type sports that require large amounts of force transmission through the abdominal and adductor musculature via the pubic symphysis. The symphysis pubis is an amphiarthrodial, fibrocartilaginous joint charged with the task of dissipating tensile, shear, and compressive forces during the gait cycle.^{5,11} Sports in which players twist, turn, and kick, such as soccer, magnify these types of forces across the symphysis potentially leading to injury.¹² Common differential diagnoses for groin pain in the athlete are muscle strain, osteitis pubis, and inguinal disruption. Inguinal disruption is a commonly misunderstood term in the sports medicine literature and is often misdiagnosed as osteitis pubis, sports hernia, adductor strain, or abdominal wall strain. Inguinal disruption⁹ is considered to be a syndrome resulting from muscular imbalances across the symphysis pubis causing exertional pubic or groin

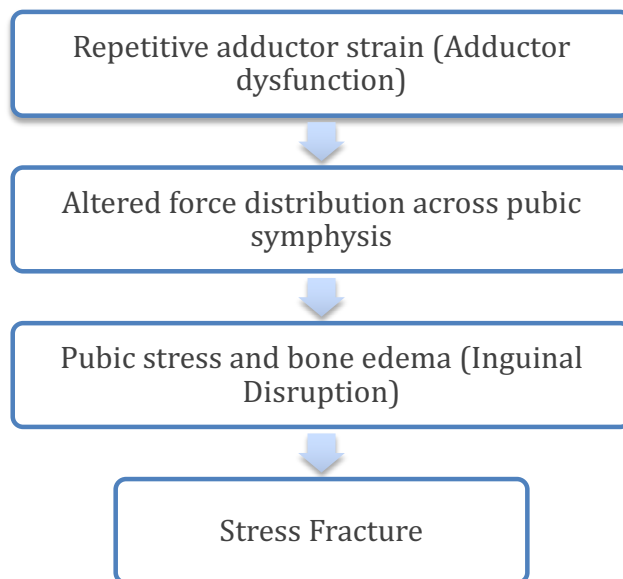


Figure 3.
Proposed progression of injury.

pain^{5,13}. The sequelae of inguinal disruption⁹ includes adductor dysfunction, and/or rectus abdominis dysfunction, along with osteitis pubis.¹³⁻¹⁵ ‘Sports hernia’, a term often used synonymously with inguinal disruption⁹, is actually a tear of the transversalis fascia of the abdomen, resulting in eventual incompetency of the posterior inguinal wall, and may, or may not, involve osteitis pubis¹⁶. The athlete in this particular case showed evidence of having both osteitis pubis and adductor longus tendinopathy. It is therefore assumed that this particular athlete represents a case of inguinal disruption⁹ that has progressed to the point of stress fracture.

One mechanism that has been proposed for inguinal disruption⁹ is repetitive stressing of the adductor and abdominal muscles during the kicking motion potentially leading to altered symphysis force distribution⁴. A magnetic resonance study examining the pelvis of professional and amateur soccer players with chronic groin pain (greater than three months) demonstrated inflammatory changes at the symphysis pubis in 97/100 subjects. In comparison, none of the 100 control subjects, soccer players and rowers, demonstrated any inflammatory changes at the symphysis ($\chi^2=188.34$, $p<0.001$). Inflammatory changes found in the chronic groin pain group include isolated

adductor micro tears in 48.4% of subjects, isolated osteitis pubis in 9.3% of subjects, and combined adductor micro tears and osteitis pubis in 42.3% of subjects. The increased prevalence of isolated adductor micro tears as well as combined micro tears with osteitis pubis in comparison to findings of isolated osteitis pubis, led the authors to hypothesize that adductor micro tears may be the precipitating event in this syndrome. Theoretically, dysfunction of the adductor musculature leads to an altered distribution of forces across the symphysis pubis causing increased stress and subsequent bone edema through this joint.¹⁴ To our knowledge there have not been any prospective studies done to examine this theory, but in the case of an athlete with a history of repetitive groin strains, this represents a logical progression of injury (Figure 3).

A number of similar MRI studies have demonstrated the high incidence of pathological changes surrounding the symphysis pubis in athletes with chronic groin pain. Not represented by a high incidence in these studies however, are associated fractures of the pubic bones.

Review papers examining the findings of inguinal disruption⁹ through advanced imaging make note of the potential for stress fractures to occur in conjunction with these injuries, however no indication of prevalence or incidence is reported^{5,17}. Stress fractures are believed to be a result of chronic stress that limits the bone's ability to remodel itself. Associated risk factors include being female, having nutritional deficits, and changes in training regiments that place greater stress on the bone.^{17,23-24}

One MRI study examined 52 athletes with groin pain of greater than 3 months compared with six asymptomatic control athletes. Findings in the groin pain group consisted of osteitis pubis, adductor dysfunction, and the pre-hernia complex, which consisted of sports hernia, conjoint tendon tear, external oblique tear, and rectus abdominis sheath tears. There were no fractures reported in this study.¹⁸ Another study of 141 patients diagnosed with either inguinal disruption or sports hernia demonstrated numerous findings of bone edema, secondary cleft signs (osteitis pubis), rectus abdominis, and adductor tendon injuries.⁹ Unlike the previous study, this particular study had one incident of pubic ramus fracture; however no demographic information was given for this patient.¹³ Further examination of the literature revealed only three case reports and one case-series of non-traumatic pubic fractures in an athletic population indicating an apparent rarity of

these injuries. In total, these four papers discussed six pubic fractures; four were avulsion injuries of gracilis¹⁹⁻²⁰, one was an avulsion injury of adductor longus²¹, and one was a combined pubic bone stress fracture with an associated adductor longus avulsion.²² Furthermore, a prospective longitudinal cohort study of 54 elite male soccer teams (2379 players) followed over eight consecutive seasons (1,180,000 hours of exposure) found that stress fractures accounted for a mere 0.5% of all injuries, with only 6% of these injuries located in the pelvis, thus indicating an extreme rarity of pelvic stress fractures in elite soccer players.²³

While the patient in this case was male with no recent change in his training, he was diagnosed with Crohn's disease six months prior to presentation which may have led to undiagnosed nutritional deficiencies. Crohn's disease is a relapsing inflammatory bowel disease (IBD) that can affect anywhere along the digestive tract, although most commonly affecting the terminal ileum (45%) and colon (32%). This disease is typically thought to occur in adulthood however over the past few decades there has been a general increase in pediatric onset inflammatory bowel disease (IBD) with approximately 20-30% being diagnosed before 18 years of age.²⁵ While there is no specific known cause of Crohn's disease it has been attributed to a combination of genetic, immunobiologic, and environmental factors.²⁶⁻²⁷ The presentation of Crohn's typically consists of gastrointestinal (GI) symptoms such as abdominal pain, constipation or diarrhea, with the passage of blood, mucus, or both. Depending upon location of the disease process, Crohn's disease can also cause malabsorption of various nutrients related to bone and general health leading to numerous extra intestinal disorders such as anemia, arthropathy, and osteoporosis.²⁶

Numerous population based studies have shown that patients diagnosed with IBD have a 21% to 40% increased risk of fracture.²⁸⁻³³ A number of factors have been suggested to increase the fracture risk in Crohn's patients including nutritional deficiencies, malabsorption, systemic inflammation, increased disease severity, and corticosteroid use; however, the evidence is lacking on a single causative factor.²⁸⁻³³ The patient in the current case was diagnosed with Crohn's disease a mere six months prior to presentation and had never been prescribed corticosteroids, however, there is evidence to suggest decreased bone density in newly diagnosed patients. A recent study

of 60 adolescents (mean age 12.2 ± 2.1 years) with newly diagnosed IBD examined bone mineral density as well as serum 25-hydroxy vitamin D at the time of diagnosis and compared them to fifty-six age and sex-matched children without IBD. Serum level of 25-hydroxy vitamin D was significantly lower in the IBD group compared to controls ($p=0.04$) and age-matched bone density Z-scores had a mean of -1.4 ± 0.9 in the IBD group indicating osteopenia in these newly diagnosed adolescents.¹⁶

Childhood and adolescence are critical periods for bone mineralization and growth. Malabsorption due to IBD may hinder proper bone formation thereby increasing the risk of fracture in this population.³¹ While it cannot be definitively known for certain whether or not IBD has contributed to the fracture in this case, it is likely to have played a role. Another relevant differential diagnosis in this case is pubic apophysitis. A retrospective case series compared an age matched symptomatic adductor related groin pain group to asymptomatic controls and found that all symptomatic subjects demonstrated stress-related physical changes of the pubic apophysis versus their counterparts.³⁴ The changes to the symptomatic apophysis included widening, asymmetry and small rounded cyst-like expansions according to these authors. Although a stress fracture in this case is likely due to the added bony burden of Crohn's disease, pubic apophysitis should be considered especially since the conservative management would be very similar.³⁴

Summary

Although quite rare, stress fractures of the pelvis are important considerations for the diagnosis of chronic and recurrent groin pain. Although these injuries can be quite painful and may require a lengthy recovery, they are not usually associated with significant complications. These injuries rarely require surgery and generally respond quite well to symptomatic treatment, temporary immobilization and rehabilitation.¹² The injury sustained by the athlete in the present case was likely a result of previous groin injuries leading to an altered force distribution, inguinal disruption⁹, and subsequent stress fracture. This may have also been the product of overtraining combined with an underlying nutritional deficiency caused by Crohn's disease. Stress fractures of the pelvis should be considered as a potential differential diagnosis of chronic or recurring groin pain particularly with female athletes. Especially

when seen following recent and sudden increases in training volume, or with athletes presenting with underlying nutritional deficiencies.

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