The Foot 25 (2015) 110–113

Contents lists available at ScienceDirect

The Foot

journal homepage: www.elsevier.com/locate/foot

Case report

Bilateral heel pain in a patient with Diamond–Blackfan anaemia

Loren T.R. Charles*, Adil M.S. Mehdi, Dennis Baker, Max R. Edwards

Trauma and Orthopaedics Department, South London Healthcare NHS Trust, Princess Royal University Hospital, Farnborough Common, Orpington, Kent BR6 8ND, United Kingdom

HIGHLIGHTS

- Two rare conditions are linked for the first time.
- The case prompts discussion of the aetiology and assessment of calcaneal stress fractures.
- A treatment algorithm for these fractures is proposed.

ARTICLE INFO

Article history:
Received 24 February 2013
Received in revised form 2 February 2015
Accepted 2 March 2015

Keywords:
Bilateral
Calcaneus
Stress
Insufficiency
Fatigue
Fracture

ABSTRACT

A rare case of bilateral calcaneal stress fractures in a patient with Diamond–Blackfan anaemia is described. This has not been previously reported in the literature. A calcaneal stress fracture is an important differential diagnosis in a patient presenting with heel pain. Bilaterality of symptoms should not exclude this diagnosis and clinicians should be especially vigilant with predisposed patients.

1. Introduction

According to Wolff's law, bone is a dynamic tissue which remodels in response to mechanical stress. Repetitive compressive and tensile strains cause adaptive changes to occur, initially in the trabeculae and then in the cortices to strengthen the bone. With sufficiently excessive, prolonged or recurrent loading however, osteoclastic resorption exceeds osteoblastic reparative activity and micro-fractures form as the rate of damage exceeds the rate of repair [1–3]. Ultimately, if there is insufficient time for healing, mechanical failure will ensue and a stress fracture occurs [3]. Stress fractures may be classified by their cause into fatigue fractures and insufficiency fractures [4].

Fatigue fractures are caused by the repetitive loading of normal bone at stresses below the ultimate tensile strength. This type of stress fracture was first recognised by Briethaupt, a German military surgeon, who noted painful feet in marching soldiers in 1855 [5]. Later in 1897, Stechow of the Prussian Guard first attributed the condition to metatarsal fractures [6] and these injuries became known as 'march fractures'. The cause was not correctly hypothesised until Hartley in 1942 proposed the theory of 'bone exhaustion', a principle similar to metal fatigue [7]. Fatigue type calcaneal stress fractures were first reported in 1937 [8] and in 1944 Hullinger reported 53 cases in military recruits [9]. Prospective studies have demonstrated a 31% incidence of fatigue fractures in soldiers, though they also commonly occur in athletes, dancers and recreational runners who average more than 25 miles per week [2,10].

Insufficiency fractures occur in abnormal bone deficient in elastic, mineral or structural resistance which is subjected to normal loading [2,3]. They affect 1–5% of the population and occur most commonly in the pelvis, proximal femur and vertebral bodies [1]. Osteoporosis is the most common cause of a multitude of conditions which weaken bone, and may be primary or secondary in origin. Causes of primary osteoporosis include hormonal, genetic, environmental or dietary causes. Secondary osteoporosis is usually the result of chronic medical conditions such as rheumatoid arthritis and drugs such as corticosteroids [1–3,10,11].

A patient with Diamond–Blackfan anaemia presenting with bilateral heel pain and subsequently found to have bilateral calcaneal insufficiency stress fractures is reported. The association between Diamond–Blackfan anaemia and stress fractures has not to our knowledge been previously described. Diamond–Blackfan

* Corresponding author. Tel.: +44 1689 865700.
E-mail address: lorencharles@doctors.org.uk (L.T.R. Charles).

http://dx.doi.org/10.1016/j.foot.2015.03.002
0958-2592/

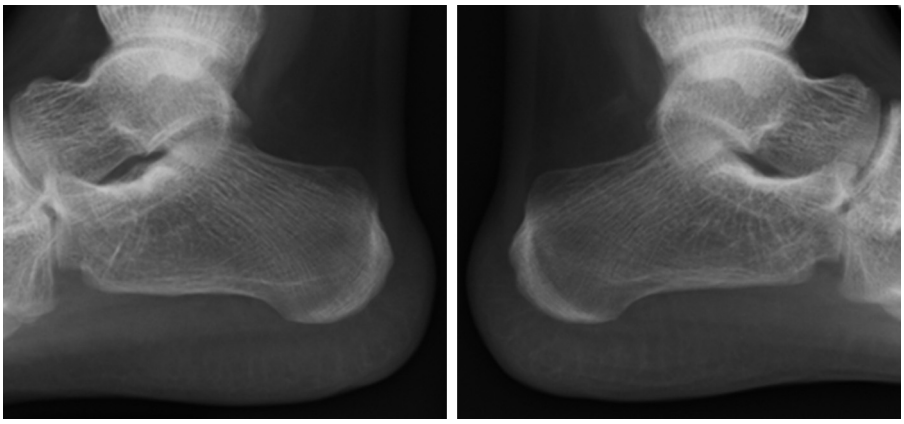

Fig. 1. Right and left lateral calcaneal radiographs.

anaemia is a rare congenital red cell aplasia that classically presents with anaemia in the first four months of life. 90% of cases are sporadic; in the 10% of familial cases most show autosomal dominant inheritance [18,19]. Two-thirds of cases have a hypoplastic macrocytic anaemia when diagnosed and although severity is variable it can become life threatening [18,19]. Approximately 30% of patients have congenital abnormalities including skeletal anomalies affecting the lateral aspects of limbs and short stature. There is also an increased risk of osteogenic sarcoma [12,18,19]. About 30% of patients respond to corticosteroid treatment but in those who do not, regular blood transfusions are often required [18,19]. Recurrent transfusions lead to chronic iron overload unless a chelating agent is administered to remove excess iron from the body. However, these agents chelate calcium as well as iron and this can lead to secondary osteoporosis.

2. Case report

A 26-year-old lady presented to the fracture clinic with a month long history of spontaneous onset bilateral heel pain, causing her to limp and limiting her activity. A past history of a congenital anaemia was noted at the initial review although this was not fully defined. Initial examination revealed medial heel tenderness at the plantar fascia insertion compatible with a diagnosis of bilateral plantar fasciitis. The patient's mother had suffered with this condition and had been successfully treated with steroid injections. Radiographs of the affected areas were unremarkable (Fig. 1).

An ultrasound scan demonstrated bilateral thickening of the peroneal longus tendons but no evidence of plantar fasciitis or further abnormality. Initial management consisted of ultrasound guided steroid injections into the peroneal longus tendon sheaths, orthotics and physiotherapy.

Six weeks after injection and following a course of physiotherapy, the patient reported no improvement. For further evaluation magnetic resonance imaging (MRI) of both feet was performed.

The MRI sequences demonstrated bilateral irregular lines of altered signal intensity consistent with stress fractures bilaterally in the posterior parts of the calcanei. Apart from small bilateral retrocalcaneal bursae, no other abnormality was seen.

On subsequent review her background of Diamond–Blackfan anaemia was explored thoroughly. She reported receiving long-term monthly blood transfusions as well as recurrent desferrioxamine and deferasirox to chelate iron and reduce chronic overload. Additionally corticosteroids had been administered during her transfusions as she had suffered a previous transfusion reaction. In keeping with Diamond–Blackfan anaemia it was noted that the patient was of short stature. She had bilateral valgus leg alignment despite previous limb re-alignment surgery as a child and had flexible pes planovalgus. Furthermore, she stated the onset of her symptoms coincided with her starting a new job, which required an increased amount of walking. It is likely that the increased and repetitive mechanical stress, combined with osteoporosis and probable subtly abnormal biomechanical loading through her calcanei, caused the stress fractures.

Conservative measures were instigated, including the fitting of bilateral Aircast Boots (DJO Incorporated, San Diego, USA), with single wedges, weight bearing as pain allowed and activity modification. A dual X-ray absorptiometry scan demonstrated a *T*-score of −3.3 consistent with severe osteoporosis (*T*-score is within the normal range if it is −1.0 or above, −2.5 or below indicates osteoporosis and −2.5 or below in the presence of one or more fragility fractures describes severe osteoporosis, as defined by the World Health Organisation and the International Osteoporosis Foundation) and she was referred to rheumatology for osteoporosis treatment. At a further review six weeks later she reported significant symptomatic improvement and subsequently returned to full activity without recurrence.

3. Discussion

This case links two rare conditions for the first time. It also demonstrates how calcaneal stress fractures may be misdiagnosed as other conditions, such as plantar fasciitis which is the commonest cause of heel pain [13] and shows the importance of gaining a detailed history to establish any relevant past medical history and predisposing factors.

Heel pain is common and the cause can be difficult to diagnose. It can be broadly divided into being of either osseous, soft tissue or systemic origin [13]. Calcaneal stress fractures commonly cause plantar or diffuse heel pain with weight bearing that is typically relieved by rest [14]. In addition to plantar fasciitis, a differential diagnosis includes insertional Achilles tendinopathy, retrocalcaneal bursitis, tarsal tunnel syndrome, heel pad atrophy and neoplastic or infective processes [11,13]. Bilateral heel pain should prompt assessment for systemic illness, including rheumatoid arthritis and the seronegative spondyloarthropathies [13].

A systematic clinical examination should assess for deformity, help localise the pain, assess foot and ankle movements and elucidate mechanical dysfunction. In the presence of a calcaneal stress fracture, inspection may reveal abnormal foot alignment and there may be heel swelling with tenderness over the os calcis or on medial to lateral calcaneal compression [13]. As demonstrated by this case, bilaterality of symptoms does not exclude a stress fracture. This is supported by a case series in which of the 134 United States military recruits who sustained calcaneal fatigue stress fractures, 73% were bilateral [7].