Freiberg’s Disease of Bilateral and Adjacent Metatarsals

Callum John Donaldson\textsuperscript{a} Ziad Harb\textsuperscript{b} Laila Hussain\textsuperscript{c} Roland Walker\textsuperscript{c} Ali Abbasian\textsuperscript{c}

\textsuperscript{a}Department of Surgery, University of Cambridge, Cambridge, UK; \textsuperscript{b}Department of Orthopaedic Surgery, Ashford and St Peter’s Hospitals NHS Foundation Trust, Chertsey, UK; \textsuperscript{c}Department of Orthopaedic Surgery, Guy’s and St Thomas’ NHS Foundation Trust, London, UK

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Abstract
Freiberg’s disease is a rare condition characterized by avascular necrosis of the lesser metatarsal heads. The incidence is highest in females, and the disease is usually unilateral, affecting just a single metatarsal. In this report, we describe the first male case of Freiberg’s disease involving the second and third metatarsals in both feet, with the aim of highlighting the condition as a potential cause of such forefoot pain. One year of follow-up is provided.
Introduction

Freiberg's disease, first described in 1914, is characterized by avascular necrosis of the lesser metatarsal heads [1]. Many cases of Freiberg's disease are asymptomatic or resolve spontaneously before treatment is sought, and this makes estimating the prevalence of the disease challenging. As such, no quantitative estimate of the prevalence of Freiberg's disease currently exists in the literature. However, Freiberg's disease is widely regarded as a rare phenomenon [2]. The incidence is highest amongst adolescent females, although the disease can develop at any age and in either sex [1, 2]. The second metatarsal is the most commonly affected, followed by the third [3]. The disease is usually unilateral and involves just a single metatarsal [4, 5]. To our knowledge, only 1 report describing bilateral involvement of both the second and third metatarsals exists in 2 female patients aged 42 and 72 years [6]. This is, therefore, the first reported male case of Freiberg's disease in bilateral and adjacent metatarsals.

Case Report

The patient, treated between November 2015 and November 2016, provided his consent to be included in this report.

This 63-year-old male presented to clinic with a 1-year history of bilateral forefoot pain, localized to the second and third metatarsophalangeal joints (MTPJs). He reported that the pain was worse on the left foot and rated the severity as 9 out of 10 on the visual analogue scale. He was able to walk pain free for only 5 min. The patient had previously been treated at another hospital, with a 5-week course of oral antibiotics, with no effect on his symptoms. He did not report pain in any other joints.

The patient did not report any history of trauma or injury. His past medical history was, however, significant for type II diabetes, which was diagnosed at a similar time to the commencement of the forefoot pain. This was under control with metformin. The patient smoked 6 cigarettes and drank approximately 10 units of alcohol per day.

On examination, there was erythema and dorsal swelling bilaterally. He had intact medial longitudinal arches, early hallux valgus with pronation of his first toe, tight gastrocnemii and positive Silfverskiold tests bilaterally. He had a painful range of motion of his left second MTPJ, palpable dorsal osteophytes and plantar tenderness of the second and third MTPJs bilaterally.

Plain radiographs showed sclerosis of the metatarsal shafts and flattening of the heads in his second and third metatarsals bilaterally (Fig. 1). Magnetic resonance imaging (MRI) showed bilateral second and third avascular necrosis of the metatarsal heads with high signal of the metatarsal shaft more pronounced in the second metatarsals (Fig. 2).

Orthotics, with a forefoot offloading insole and incorporated metatarsal bar to reduce the point pressures on the affected metatarsal heads, were provided but did not improve the bilateral forefoot pain. As conservative measures failed to improve symptoms, the patient underwent a left foot second and third MTPJ debridement and second and third Weil's osteotomy. Weil's osteotomy was performed using standard technique via a dorsal incision. Weil's osteotomy was preferred because the whole head was involved and collapsed, meaning that
simple debridement would not have sufficed. This is because simple debridement does not
address the root cause, which is the relatively long second and third metatarsals within the
anatomical cascade. Similarly, closing dorsal wedge or rotational osteotomies were also not
possible because very little intact cartilage was seen on the plantar articular surface. The de-
gree of shortening was determined by intra-operative image intensifier guidance to restore
the natural metatarsal cascade (Lelièvre’s parabola). A single cut was performed, taking into
account the thickness of the saw blade, meaning that there was natural elevation of the head
and avoidance of plantarisation. Fixation of the osteotomies was completed with Ortho Solu-
tions™ (Maldon, Essex, United Kingdom) twist-off screws. Intra-operatively, the affected
metatarsals showed typical features of degenerative articular surfaces with a necrotic metat-
sarsal head, secondary osteophytosis and synovitis. Post-operative instructions permitted full
weight bearing in a DARCO MedSurg™ Shoe (DARCO International, Huntington, WV, USA), to
offload the forefoot, for 6 weeks. At 2 weeks post-operatively, the wound had healed without
any complications. At 6 weeks, the patient reported that he was doing very well and was pain
free, whilst radiographs revealed good bone healing. He subsequently underwent the same
procedure on the right foot 6 months after the initial operation and, following the same post-
operative procedure as the contralateral extremity, made a similarly favourable post-opera-
tive recovery, with no complications, good wound healing, full return to normal activities and
the patient happy with his outcome (Fig. 3). Given the success of bilateral debridement and
Weil’s osteotomies in managing the patient’s symptoms, his gastrocnemius tightness was man-
aged with physiotherapy, typically the first-line treatment for this condition, instead of at-
tempting a recession procedure with its inherent risks. He continued to do well and was dis-
charged from our care 1 year after the initial operation.

Discussion

Trauma and vascular compromise are the 2 most commonly cited aetiologies [7]. The sec-
ond metatarsal’s relative immobility and greater length, in comparison to the other metatars-
sals, make it susceptible to stress during normal activity and may explain the disease’s predi-
lection for this site [8]. In a similar vein, repetitive forced dorsiflexion injuries of the metatar-
sal heads, inflicted by high-heeled shoes for example, may explain the higher preponderance
of the disease amongst females [8]. A predisposition to vascular compromise is likely to exist
in those with an anatomical variant, in which the second metatarsal artery is absent and blood
supply to the metatarsal comes instead from the first and third metatarsal arteries [7]. Asso-
ciations with systemic disorders, including systemic lupus erythematosus, hypercoagulability
and diabetes mellitus, have also been noted [8, 9]. Muscular atrophy of the foot, secondary to
neuropathy, is thought to be the pathological factor in diabetes [9].

Patients commonly present with forefoot pain, localized to the metatarsal heads and ex-
acerbated by weight bearing [10]. The affected joint, most commonly the second MTPJ, may
be swollen with limited range of movement and, in advanced cases, show features of malalign-
ment and deformity [10]. The diagnosis can be confirmed by radiography which, as the dis-
ease progresses, will show increased joint space, flattening of the metatarsal head in the an-
teroposterior diameter, bony fragmentation and, finally, complete degeneration of the joint
In early stages of the disease, plain films are occasionally normal, but MRI can be used to demonstrate hypo-intensity of the metatarsal head, bone oedema as well as fragmentation and collapse, whilst bone scans may reveal a photopenic core with a hyperactive cortex [11].

In 1967, Smillie [12] described a classification of Freiberg’s disease based on the intra-operative appearance of affected metatarsals; however, all but the first stage are also appreciable radiographically (Table 1).

Conservative management should be employed first line in all cases of Freiberg’s disease and includes oral analgesia, rest, orthotics and immobilization [8]. Conservative measures aim to control pain as well as to offload the affected metatarsals and prevent progression to later stages of the disease in which anatomical restoration is no longer achievable. These have proven particularly effective in Smillie disease stages 1–3 [13]. In cases where these measures are ineffective, operative treatment may be employed. A wide variety of surgical techniques have been utilized, including joint debridement, micro-fracture, dorsal wedge osteotomy, metatarsal shortening osteotomy, metatarsal head resection and arthroplasty [14]. Whilst successes have been reported for all of the above, the evidence is currently insufficient to recommend any one procedure as the optimal technique [13, 14]. However, generally speaking, joint-preserving procedures have been shown to produce superior outcomes to joint-destructive procedures, and the latter should be reserved for Smillie disease stages 4 and 5 [13, 15].

Following promising demonstration of the technique in recent case series, osteochondral autograft transplantation may also develop a role in the operative management of Freiberg’s disease [16, 17].

We report this case report for its uniqueness and for its educational value in understanding the plethora of causes of forefoot pain. Only 1 report of Freiberg’s disease affecting bilateral and adjacent metatarsals has been published previously and concerned 2 female patients. This is the first time that the same extensive pattern of disease has been described in the less commonly affected male sex.

**Statement of Ethics**

The subject provided their informed consent for their case and associated images to be published.

**Disclosure Statement**

The authors have no conflicts of interest to declare.

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Author Contributions

Callum J. Donaldson, Ziad Harb, Laila Hussain, Roland Walker and Ali Abbasian made substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; and drafting the work or revising it critically for important intellectual content; and final approval of the version to be published; and agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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Fig. 1. Left weight-bearing antero-posterior (a), right weight-bearing antero-posterior (b), left lateral (c) and right lateral (d) pre-operative radiographs showing bilateral sclerosis of the second and third metatarsals and bilateral flattening of the second and third metatarsal heads.
Fig. 2. Left postero-anterior (a), right postero-anterior (b), left coronal (c) and right coronal (d) MRI images showing bilateral avascular necrosis of the second and third metatarsal heads, with high signal of the metatarsal shaft more pronounced in the second metatarsals bilaterally (appreciable in c, d).
Fig. 3. Left weight-bearing antero-posterior (a), right weight-bearing antero-posterior (b), left lateral (c) and right lateral (d) 6-week post-operative radiographs showing good bone healing.
### Table 1. The Smillie staging system for Freiberg’s disease

| Smillie stage | Description |
|---------------|-------------|
| 1             | A fissure fracture develops in the ischaemic metatarsal epiphysis |
| 2             | Absorption of bone has taken place, and the central portion of the metatarsal head begins to sink into itself, altering the contour of the articular surface |
| 3             | Further absorption has taken place, and the central portion of the metatarsal has sunk into itself leaving projections on either side; the plantar articular cartilage remains intact |
| 4             | The plantar hinge of articular cartilage has given way and peripheral projections have fractured to form loose bodies; anatomical restoration is no longer possible |
| 5             | The final stage shows arthrosis with marked flattening and deformity of the metatarsal head |

Information from Smillie [12].