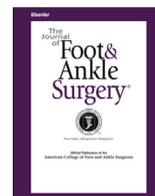




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Osteomyelitis of the Calcaneus With Pathologic Fracture

Maryellen P. Brucato, DPM, AACFAS¹, Matthew F. Wachtler, DPM, FACFAS¹,
Ellianne M. Nasser, DPM, FACFAS²¹ Attending Physician, Atlantic Health System, Morristown, NJ² Attending Physician, Geisinger Health System, Danville, PA

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ABSTRACT

Pathologic fractures of the calcaneus secondary to osteomyelitis (OM) have rarely been reported in the literature. This case series describes 5 patients who were treated in our institution for chronic OM of the calcaneus and subsequently suffered a fracture of the involved calcaneus in the absence of trauma. All 5 patients had a history of insulin-dependent diabetes mellitus and were treated with a range of surgical treatments including open reduction and internal fixation, external fixation, and excision of the fracture fragment. Three (60%) of the patients required a below-the-knee amputation of the ipsilateral limb, 1 (20%) expired, and 1 (20%) experienced healing of the fracture and the associated heel wound. Pathologic fracture of the calcaneus secondary to OM is a recognized entity, although case descriptions have rarely been reported for this challenging condition.

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The calcaneus plays a crucial role in normal ambulation and lower extremity function, making 2 types of possible ailments particularly devastating: fracture and osteomyelitis (OM). Calcaneal fractures are considered a severe foot and ankle injury, often requiring extensive surgical intervention and postoperative care with an elevated risk of complications. OM of the calcaneus is another devastating pedal disorder that is often associated with multiple complications. Independently, OM of the calcaneus can result in the need for long-term intravenous antibiotic therapy, surgical debridement and/or reconstruction, difficulty with ambulation, loss of function, amputation, and even death (1). When a fracture of the calcaneus occurs in the setting of OM, the foot and ankle surgeon is presented with a condition that is exceptionally difficult to treat successfully (salvage of a weightbearing foot), and major amputation is often the resultant outcome. Importantly, diabetes mellitus is a common comorbidity of this patient population, and it is associated with various lower extremity risk factors such as neuropathy, peripheral arterial disease (PAD), a suppressed immune response, and neuroarthropathy, all of which predispose the patient to infection and decrease the likelihood of salvaging a functionally weightbearing foot.

The Hedlund classification pertains to diabetic patients with calcaneal fractures (2). The report, published in 1998, described 3 basic fracture patterns: type I, an avulsion fracture; type II, a midcalcaneal

compression fracture; and type III, a wedge-type fracture. The study identified 22 total patients, 4 of whom had a history of a heel ulcer (2).

In this report, we describe 5 diabetic patients who were treated at our institution for chronic OM of the calcaneus and who subsequently suffered a pathologic fracture of the involved calcaneus in the absence of an acute traumatic event. The purpose of this report is to highlight this condition in terms of its morbidity and mortality, with the hopes of instigating future observational studies and further discussion of its evaluation and management.

Case Series*Identification of Patients*

A chart review was conducted by 2 of the authors (M.P.B. and M.F.W.) on all patients who presented to the podiatric surgery service between January 2010 and December 2012. We inspected each record on a consecutive master registry to look for patients who fit the inclusion criteria. Inclusion criteria were defined as patients presenting with a calcaneal fracture found on plain radiographs with a prior history of OM in the same calcaneus. The presence of OM needed to be documented preceding the fracture via positive wound culture combined with either histological evidence in the bone biopsy or magnetic resonance imaging (MRI) findings suggestive of infection, namely increased signal intensity on T2 images showing bone marrow edema within the calcaneus (3,4). Additionally, to be included, the patient had to have a current ulceration of the ipsilateral heel at the time of the calcaneal fracture, with a positive probe-to-bone sign, suggestive of OM (5). The presence of diabetes mellitus was not a distinct inclusion criterion, and

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Address correspondence to: Maryellen P. Brucato, DPM, AACFAS, Morristown Memorial Hospital, Atlantic Health Podiatric Surgery, 103 Colfax Avenue, First Floor, Pompton Lakes, NJ 07442.

E-mail address: marebeary1@gmail.com (M.P. Brucato).

any patient treated for presumed OM without documentation (culture plus biopsy or MRI findings) was excluded. Based on these criteria, a total of 5 diabetic patients who were treated at the primary (M.P.B.) and secondary (M.F.W.) authors' institution, namely the Atlantic Health System, in Morristown, New Jersey, and who were initially diagnosed with calcaneal OM and subsequently sustained a fracture of the involved calcaneus were identified and included in this review. All 5 of these patients presented with no obvious history of acute trauma localized to the involved heel. The first treatment on a patient started in November 2010 and the last treatment date was September 2012, so the duration of the observation period for inclusion in this review was ~23 months. Although not a requirement for inclusion in our review, all 5 of the patients were treated with 6 to 8 weeks of intravenous antibiotics and partial calcanectomy at the time that the OM was diagnosed. At our hospital, this investigation (N < 25) qualified for expedited review, and our sole requirement was that protected health information had to remain confidential.

Demographic exposures that we recorded included patient age, sex, body mass index, side involved, duration of OM at the time of fracture (days), fracture pattern type based on the diabetic calcaneal fracture classification (2), presence of insulin-dependent diabetes, presence of anemia (hemoglobin < 10 g/dL), smoking history, history of end-stage renal disease, and history of PAD. We also recorded wound culture, bone biopsy, and MRI results. Wound cultures from the heel ulcers were recorded from the time that the patients were diagnosed with, and subsequently treated for, OM. The results of the probe-to-bone test were also logged for each patient (5). The end point (outcome) for each patient was determined to be healing of the fracture and ulcer, amputation, or death. Follow-up examined in this study stopped when any of the 3 end points was reached, which ranged from 10 months to 2 years.

Results

A statistical description of the 5 patients is shown in the Table. The mean patient age was 55 ± 6.9 years, and the mean body mass index was 29.39 ± 6.16 kg/m². Only 1 (20%) of the patients was male, and the right side was involved in 2 (40%) of the cases. All of the patients had insulin-dependent diabetes and a history of cigarette smoking, and in each case the probe-to-bone clinical test suggested the presence of OM of the calcaneus (5). In this series, moreover, 4 (80%) were anemic, 1 (20%) was on dialysis for end-stage renal disease, and 2 (40%) had PAD. Four (80%) of the fractures were type III as seen in Fig. 1, and 1 (20%) was type I as seen in Fig. 2. The mean duration of OM prior to fracture was 100 ± 51.1 days (range 30 to 165), which is summarized in Fig. 3. In addition to clinical history and appearance, including the presence of the probe-to-bone sign (5), the diagnosis of OM was based on MRI findings in 3 (60%) and biopsy in 2 (40%) of the cases. Bone cultures revealed 13 different types of organisms including *Enterococcus faecalis* (40%), β -hemolytic group B *Streptococcus* (40%), and *Diphtheroid bacillus* (40%), with the following organisms cultured only once: *Escherichia coli*, *Enterobacter cloacae*, *Enterococcus faecium*, *Candida parapsilosis*, *Clostridium cadaveris*, *Klebsiella pneumoniae*, methicillin-resistant *Staphylococcus aureus*, methicillin-sensitive *Staphylococcus aureus*, *Proteus mirabilis*, and *Pseudomonas aeruginosa*. During the follow-up period, 3 (60%) patients went on to a below-the-knee amputation (BKA), 1 (20%) healed, and 1 (20%) died (the cause of death was determined to be cardiac arrest).

Initial fracture management varied on a case-by-case basis. Three patients were initially treated with excision of the fracture fragment as seen in Fig. 4. In 1 (20%) patient, in addition to excision of the fragment, the Achilles tendon was able to be salvaged and was transferred to the remaining posterior calcaneus with anchor fixation (Fig. 5). One (20%) patient was treated with a percutaneous 8-mm cannulated screw and Achilles tendon lengthening (Fig. 6). Last, 1 (20%) patient was treated

with external fixation, which failed 2 days postoperatively, and subsequently underwent a resection of the fracture fragment (Fig. 7). All patients continued to receive local wound care after the procedures, which included negative-pressure wound therapy and biosynthetic grafting. Bone cultures that were recorded revealed 13 different types of organisms, with a mean of 3.2 organisms per patient. The most prevalent organisms were β -hemolytic group B *Streptococcus*, *Diphtheroid bacillus*, and *Enterococcus faecalis*.

In total, 3 (60%) of the cases resulted in BKA of the affected limb secondary to recalcitrant OM, nonhealing ulceration, and inability to salvage the limb. One (20%) case that was initially treated with the excision of the fracture fragment remained unhealed, with an open ulceration of the ipsilateral heel until the patient expired secondary to cardiac arrest. Finally, the patient who underwent fixation with a percutaneous screw experienced healing of the ulceration and the fracture within 52 weeks. Additional treatment for the wound included negative-pressure wound therapy and biosynthetic grafting. Also of note, the fracture pattern was type III. Follow-up with the patient continued for 11 months after the ulcer healed without incidence of reulceration or recalcitrant OM. The patient was partially ambulatory owing to ulceration on her other foot. After 11 months, the patient was lost to follow-up.

Discussion

Pathologic fracture of the calcaneus secondary to OM is a rare, limb-threatening condition. These 2 distinct ailments alone typically present a challenge to the foot and ankle surgeon; when combined, they create a unique condition that is exceptionally difficult to treat.

Literature regarding diabetic calcaneal fractures is insufficient. The majority of what is written is extrapolated from the diabetic ankle fracture literature and focuses on hemoglobin A1C as well as prolonged immobilization.

In 1998, Hedlund et al (2) identified 22 diabetic patients who presented with calcaneal fractures in a 25-year period without any significant contributing trauma. The study described 3 different basic fracture patterns: types I, II, and III. Type I is an avulsion fracture pattern, type II was described as a midcalcaneal compression fracture, and a wedge-type fracture was branded as a type III fracture pattern. Only 1 patient in their review had a diagnosis of OM prior to fracture. Ten patients in total (45.5%) had a history of preceding chronic heel ulcer on the ipsilateral foot. The study cited multiple contributing factors to the etiology of the calcaneal fractures, including decreased bone mineralization (secondary to diabetes, long-term steroid use, neuropathic hyperemia, decreased weightbearing) and alteration of gait (secondary to neuropathy, retinopathy, injury, amputation). Other factors that have been previously identified as risk factors for diabetic pathologic calcaneal fractures include tight Achilles tendon and renal disease (2). The patients in our study suffered from a variety of these different stressors, with the addition of OM of the calcaneus, a previously unidentified risk factor.

The type I fracture pattern was initially described in 1991 by Kathol et al (6) as a calcaneal insufficiency avulsion fracture. The authors noted that these fractures tend to be extra-articular and limited to the posterior one third of the calcaneus. They hypothesized that these types of fractures occurred owing to the Achilles tendon overpowering a weaker area of the calcaneus in the absence of trauma. In the 1991 article, 14 diabetic patients who presented with calcaneal insufficiency avulsion fractures were identified. Of these patients, all were insulin dependent, all had neuropathy, and 4 were noted to have associated heel ulceration at the ipsilateral foot (6). Kathol et al (6) described a sequence of events through which the calcaneal insufficiency fracture occurs. The process begins with microfracture to the calcaneus from daily activity. This progresses to a nondisplaced insufficiency fracture, which may show radiographic changes. Fragmentation occurs to the insufficiency fracture,

Table
A statistical description of the case series

Case	1	2	3	4	5	Summary Statistics, Mean ± SD (median [range]) or count (%)
Age, y	49	61	48	63	58	55.8 ± 6.9 (58 [48, 63])
Sex	Female	Female	Male	Female	Female	4 (80%) female; 1 (20%) male
Body mass index, kg/m ²	33.1	24.77	25.06	38.5	25.5	29.39 ± 6.16 (25.5 [24.77, 38.5])
Side involved	Left	Left	Left	Right	Right	3 (60%) left; 2 (40%) right
Duration of OM, d	30	165	95	130	80	100 ± 51.1 (95 [30, 165])
Fracture category*	III	III	III	III	I	4 (80%) III (wedge); 1 (20%) I (avulsion)
Diabetes mellitus	Yes	Yes	Yes	Yes	Yes	5 (100%) yes
Anemia [†]	Yes	Yes	Yes	No	Yes	1 (20%) no; 4 (80%) yes
Smoking history	Yes	Yes	Yes	Yes	Yes	5 (100%) yes
End-stage renal disease	No	No	No	No	Yes	4 (80%) no; 1 (20%) yes
Peripheral arterial disease	No	No	No	Yes	Yes	3 (60%) no; 2 (40%) yes
Probe to bone	Yes	Yes	Yes	Yes	Yes	5 (100%) yes
Calcaneal culture	Yes	Yes	Yes	Yes	Yes	5 (100%) yes
Biopsy indicates OM	No	No	No	Yes	Yes	3 (60%) no; 2 (40%) yes
MRI indicates OM	Yes	Yes	Yes	No	No	2 (40%) no; 3 (60%) yes
Treatment of fracture	Excision of fragment	Excision of fragment	External fixation	Percutaneous screw + TAL	Excision of fragment	3 (60%) excision fragment; 1 (20%) external fixation; 1 (20%) percutaneous screw + TAL
Outcome	BKA	BKA	BKA	Healed	Died	3 (60%) BKA; 1 (20%) healed; 1 (20%) died

Abbreviations: BKA, below-the-knee amputation; MRI, magnetic resonance imaging; OM, osteomyelitis; SD, standard deviation; TAL, tendo-Achilles lengthening.

* Hedlund et al (2).

† Anemia defined as hemoglobin < 10 g/dL.



Fig. 1. Radiograph of case no. 1, presentation of type III wedge fracture.



Fig. 2. Radiograph of case no. 5, presentation of type I avulsion fracture.

typically resulting in a secondary fracture line. Finally, the fracture fragments displace (6).

The type I fracture pattern has a corresponding collocation in the Brodsky classification, a classification system of Charcot deformities

based on anatomic location. Type I fractures correspond to a Brodsky type IIIb (7). Brodsky described the type IIIb Charcot deformity as a pathologic fracture of the posterior tubercle of the calcaneus leading to deformity and collapse of distal joints (7).

There have been minimal reports of pathologic calcaneal fractures in patients with a history of chronic ulcerations of the heel, and none of the reports showed documented OM before the time of fracture. In 2004, Roldan (8) reported a single case study involving a pathologic fracture of the calcaneus from presumed OM following a chronic heel ulceration of 2 months. The OM was not documented or diagnosed before the fracture. Athans and Stephens (9) reported a case series of 3 diabetic patients who sustained open calcaneal fractures referred to as pathologic fractures secondary to diabetes, not OM. All 3 had preceding heel ulcerations, and 1 had confirmed OM.

In 1979, Coventry and Rothacker (10) published a case report that documented an insulin-dependent diabetic female with bilateral stress fractures of the calcaneus. The patient did not have significant trauma before the fractures; however, there were altered biomechanical stresses combined with a history of polyneuropathy (10). Platts-Mills et al (11) later reported a diabetic patient who presented with a calcaneal avulsion fracture in the complete absence of trauma, which was then treated with a single 6.5-cm screw.

Although all 5 patients in the current study had been treated previously with a partial calcaneotomy, there are no prior reports that show this would weaken the calcaneus to the point of fracture with regular load bearing. In 2002, Bollinger and Thordarson (12) reported that the partial calcaneotomy is a viable alternative option to BKA for treatment of OM.

Any type of foot and ankle fracture in the setting of OM presents a distinct challenge regarding treatment. This is compounded by the fact that 25% of all calcaneal fractures have wound complications (13). When these 2 types of injuries occur simultaneously in the calcaneus, it creates a perfect storm of an extremely complicated surgical challenge.

Traditional open reduction and internal fixation is not a completely viable option because bone quality is most likely compromised. Screw holding power is directly proportional to bone density. Traditionally, the surgeon wants to avoid placing hardware inside of infected bone for fear of seeding the hardware with bacteria. The single patient in our series who was ultimately treated with open reduction and internal fixation of the calcaneus was first treated with long-term intravenous antibiotics for a course of 6 weeks, and the treating surgeon believed

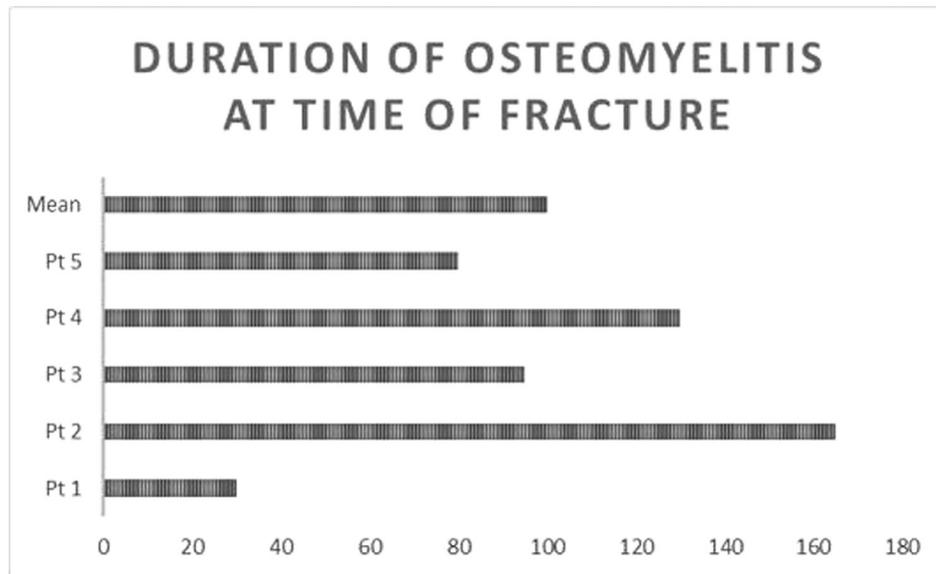


Fig. 3. Graph presentation of duration (days) of osteomyelitis at the time of the calcaneal fracture.



Fig. 4. Postoperative radiograph of case no. 2, treated with excision of fracture fragment.



Fig. 5. Postoperative radiograph of case no. 5, treated with excision of fracture fragment and reattachment of Achilles tendon with anchor fixation.

that this was sufficient. In 2017, a study by Yildirim et al (14) demonstrated successful results in pediatric fractures secondary to OM treated with wide debridement, irrigation, bone grafting, and anatomic locking plate and screws to reduce the fracture. Their conclusion was that a pathologic fracture must be stabilized in addition to conventional treatment for OM.

External fixation may be warranted to reduce and stabilize the fracture; however, that does require structural integrity of the bone,

including the infected fracture fragment. Partial calcaneotomy or excision of the fracture fragment is a choice that is complicated by the insertion of the Achilles tendon. If the Achilles tendon is not addressed, the patient may become at risk for calcaneal gait. Greenhagen et al (15) reported a case study in which a diabetic calcaneal insufficiency fracture was treated with excision of the fracture fragment with a gastrocnemius recession and reattachment of the Achilles tendon to the remaining calcaneus. It is important to note that OM was not present.

Last, BKA may present the most functional option for these patients. Although the goal is for limb salvage, the decision may be made at the time of the infected calcaneal fracture to proceed with a BKA and plan for prosthetic fitting and extensive rehabilitation once the incision is healed. It is important to note that some studies have found decreased functional ability following BKA. In 1976, Waters et al (16) found that patients had a gait velocity significantly decreased from normal ambulation. If the decision is made to go forward with BKA, the surgeon and patient should be in agreement that this would indeed provide the best result based on the patient's needs and functional ability (1,17).

The outcomes for the 5 patients included in this case series were significantly worse compared with previous reports that did not involve OM. Hedlund et al (2) reported that all 22 diabetic calcaneal fractures healed; however, 50% healed with significant calcaneal deformity. Furthermore, the patients in that study did not have documented OM.

When a diabetic patient presents with a nonhealing heel ulcer in the setting of OM, the patient should be properly offloaded to avoid a pathologic fracture. In this case series, all of the patients underwent previous partial calcaneotomy, which could have further weakened the infected bone. A recent study by Oliver et al (18) evaluated functional results in patients after undergoing partial calcaneotomy to treat OM. They hypothesized that with a plantar calcaneotomy leaving much of the Achilles tendon intact, the patient is at more of a risk to fracture the calcaneus biomechanically with the pull of the Achilles tendon overpowering the dorsal cortex. They looked at 2 cohorts: patients who had <50% of the calcaneus resected versus those who had >50% of the calcaneus resected. However, their results showed a BKA rate of 29% in all patients who underwent partial calcaneotomy regardless of the surgical technique (18).

Limitations of the current study include a small study population and the retrospective nature of our case series. Additionally, this is



Fig. 6. Postoperative radiographs of case no. 4, treated with open reduction and internal fixation.



Fig. 7. Postoperative radiographs of case no. 4, treated with open reduction and internal fixation with screw, after screw removal and healed fracture.

a purely observational study. Each patient was treated by a different primary surgeon at our institution, so it was impossible to determine the exact surgical technique of the initial partial calcaneotomy that could have contributed to the fracture of the calcaneus. Ideally, a bone biopsy at the time of fracture would have been performed as well to confirm the diagnosis of OM for a second time. Moreover, we recognize that identifying patients by the diagnosis of calcaneal fracture on a master patient list can be troublesome, because it is possible that the record keeping was not done in a rigorous or uniform fashion.

In conclusion, looking to the future, there is no definitive treatment algorithm for calcaneal fractures in the setting of OM. The outcomes in this study highlight the limb-threatening nature of this combined condition. A prospective study would be warranted to develop recommendations and guidelines for best treatment practices.

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