Charcot neuroarthropathy following dorsiflexory wedge osteotomy of the first metatarsocuneiform joint

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Abstract:
Operating on diabetic patients comes with risk. Possible complications include poor or delayed soft tissue and bone healing, infection, disruption of glycemic control, renal issues, postoperative myocardial ischemia, stroke, and death. Pre-operatively the surgeon can attempt to mitigate the potential for a post-operative problem. One complication the surgeon often cannot mitigate is the development of Charcot Neuroarthropathy (CN). Despite a surgeon’s attempt to help the patient through treatments, there is always a risk of doing harm. An example is a surgically-induced CN. We present a case report of two diabetic patients who each underwent a first tarsometatarsal joint (TMTJ) fusion with a dorsiflexory wedge osteotomy (DFWO) to cure a sub-metatarsal head ulceration that had failed conservative treatment. This procedure can help cure chronic wounds by reducing and redistributing pressure, and preventing ray (or even greater) amputation. Although both patients’ ulcers healed roughly two months post-operation, both developed CN changes in the tarsometatarsal region approximately 2-3 months post-operation. We believe that the local traumatic event of the surgery, combined with the initiation of weight-bearing post-operation led to development of CN. This unexpected, and unforeseen complication, is not well documented in the current literature for the procedure performed. Although we could not have predicted this outcome, reflecting upon the two cases, a better pre-operative assessment of the patient, a more delicate surgical technique, and more robust fixation of the medial column could possibly have prevented these results.

Key words: Charcot Neuroarthropathy, Diabetes, Dorsiflexory Wedge Osteotomy, First Tarsometatarsal Joint Fusion, Joint Ulcer, Meary’s Angle, Medial Column Instability

INTRODUCTION
A common site for diabetic foot ulceration is the plantar first metatarsal head. Almost one-quarter of diabetic ulcerations occur at this site. If infection develops, partial or complete first ray amputation may be necessary. This amputation has a 20-60% re-amputation rate, including additional digital, transmetatarsal, or below-knee amputations. Possible curative surgery is a first TMTJ with a DFWO resection to elevate and slightly shorten the plantarflexed or long first ray. The purpose of this is to re-establish the lateral Meary’s angle and decrease the pressure to the soft tissues superficial to the bone in an attempt to heal the ulcer and prevent recurrence. However, one potential complication with this surgery is disruption of LisFranc's ligament and other ligamentous connections between the medial and middle columns. This stabilizing unit in the medial column centers around the recessed second metatarsal, often described as the keystone of the joint. Disruption of this region, either traumatic or iatrogenic in origin, can result in hypermobility and instability of the joint with potential for progressive foot deformity.

Two diabetic patients with chronic wounds that failed to heal after three months of local wound care and offloading were presented. Surgery was performed to structurally offload the ulcer. The patients’ ulcers healed completely within weeks after surgery. Other wound care
therapies consisting of biologics (ie, becaplermin gel and porcine small intestine submucosa graft) were used, and patients were given instructions to remain non-weight-bearing (NWB) through the healing process. Ulcer healing occurred in both patients, and no recurrence of ulceration, pre-ulcerative lesion (callus), or new transfer lesion was noted at 10 and 18 months follow-up. However, after a period of protected weight-bearing and approximately 2-3 months post-surgery, both patients developed erythema, edema, warmth, and pain to their surgical foot. They denied any traumatic events to their feet subsequent to their surgeries. Radiographs were taken, and acute CN changes were seen about the LisFranc joint.

The authors feel that the dissection involved in the procedures created trauma and instability to the LisFranc joint complex. This local trauma, and instability in conjunction with diabetic neuropathy, acted as a perfect storm to incite Charcot changes to the foot. Considering how quickly the CN developed from the time of surgery, along with the entire clinical picture, we conclude that the traumatic event inciting the acute CN episode was the surgery.

Case Reports

Two male Type-2 diabetic patients, aged 27 and 53-years-old, presented at our clinic. Both had sub-first metatarsal head ulcerations of greater than two months duration that failed to heal at another clinic. Upon presentation, the ulcers each had a fibrogranular base, measuring 1cm$^3$ and 1.45cm$^3$ respectively, with hyperkeratotic rims. They appeared non-infected with a negative probe to bone test. At our institution, the patients failed local wound care therapies (ie, sharp debridement, offloading, and total contact casting (TCC)) over the course of weekly visits for one month (Figures 1A and 2A). Curative surgery (first TMTJ fusion with DFWO) was recommended because of the structural bony abnormality of the foot (Figure 3).

Pre-operatively the patients had a lateral Meary’s angle of 21° and 11° respectively with post-operative correction to 14° and 3° respectively (Figures 4 and 5). The surgery consisted of first TMTJ fusion with a dorsally based closing wedge resection using an interfragmentary screw and 4-screw plate fixation (Figures 4 and 5). One patient received a porcine small intestine submucosa graft while the other used becaplermin gel during the post-operative healing phase.
After 4 weeks of NWB in a controlled ankle motion (CAM) boot both ulcers measured less than 0.05 cm³. The patients were kept NWB and slowly transitioned to protected weight-bearing at the 6-10 week mark. At the 8 week mark, both patients’ ulcers showed clinical healing. No recurrence of ulceration, pre-ulcerative lesion, or new transfer lesion has been noted after 10 and 18 months of follow-up respectively (Figures 1B-D and 2B-D).

At approximately 9 and 12 weeks respectively post-operation, each patient complained of sudden erythema, edema, and calor to the surgical foot. They both denied any trauma or subjective constitutional symptoms of infection. Both plantar ulcerations and surgical wounds were healed. Each patient was immediately immobilized with either a CAM boot or TCC and instructed to remain NWB.

*Angles drawn and measured by lead author using Bone Ninja application for iPad. (LifeBridge Health, Sinai Hospital of Baltimore, Baltimore, MD)
One patient was given an antibiotic after several days of no changes in foot appearance. Antibiotic choice was escalated again after no changes in appearance in the following days. Suspecting possible CN, both patients were sent for radiographic imaging which revealed acute Charcot changes. Serial radiographs were taken for both patients showing bone demineralization and failure of hardware over the subsequent weeks (Figures 6 and 7).

One patient, who is still seen in the clinic, has yet to re-ulcerate or develop even a callus at the initial site or other pedal locations at 18 months post-surgery. The patient ambulates in a Total Contact Orthotic Restraining Custom Hybrid (TORCH) Walker. With continued observation and interval radiographs, there has been progressing dorso-lateral subluxation of the lesser metatarsals at the TMTJ and new breakdown at the first metatarsophalangeal joint noted at 12 months post-operation (Figure 8).

Figure 6A. 27-year-old male. Pre-op.
Figure 6B. 4 weeks post-op (stable, post-surgical changes to 1M-MCJ with hardware intact).
Figure 6C. 11 weeks post-op (patient complains of erythema, edema, calor to foot; prescribed radiograph seen here and NWB in CAM boot. Radiographs reveal hardware failure with fracture of metatarsal bases 2 and 3 with callus formation across the midfoot). Figure 6D. 16 weeks post-op (1 month of protected NWB in TCC with decreasing signs clinically. Radiographically, continued subluxation of metatarsals and heterotrophic ossification formation).

Figure 7A. 53-year-old male. Pre-op.
Figure 7B. Immediate post-op.
Figure 7C. 9 weeks post-op (patient complains of erythema, edema, calor to foot).
Figure 7D. 14 weeks post-op (1 month of protected NWB in CAM Boot).

Figure 8Aa. 27-year-old male. AP radiographs at 12 months post-op revealing failed, broken hardware, dorso-lateral subluxation of the lesser metatarsals, increased sclerosis at the base of the first metatarsal, and destruction of the first metatarsophalangeal joint. Figure 8B. Lateral radiographs at 12 months post-op revealing failed, broken hardware, dorso-lateral subluxation of the lesser metatarsals, increased sclerosis at the base of the first metatarsal, and destruction of the first metatarsophalangeal joint.
The second patient was lost to follow-up after 10 months. At the last visit, the patient had already transitioned from CAM-NWB to a regular walking sneaker with a custom insole and also had no re-ulceration with no callus formation on the plantar aspect of the foot.

DISCUSSION

After a 4 week period of conservative treatment, a reduction in wound size less than 53% is a cutoff that predicts a poor healing outcome at 3 months.⁵ If this occurs, a change in plan should take place as in this case report⁴ which proposed a classification system to help dictate the need for surgery in a diabetic patient.⁴ Their system ranges across four class: elective, prophylactic, curative, and emergent. Three distinguishing factors upon which classification is determined are the presence or absence of neuropathy; an open wound; and acute, limb-threatening infection.⁴ Based on the classification system, our cases represent curative surgery in an attempt to structurally offload the ulcer, correcting the etiology of the problem.

A first TMTJ fusion with a DFWO is described for correction of a plantarflexed first ray, indicated in structural, non-reducible deformities. These include footvalgus or cavovarus foot types, or the diabetic foot with ulcer or pre-ulcerative lesion.¹⁶⁷

The lateral Talo-1ˢᵗ Metatarsal Angle (lateral Meary's angle) measures the apex of deformity in pes cavus or planus foot types. A normal Meary's Angle is measured at 5 ± 4⁸ or has been described as parallel lines⁸ (Figure 3). The first TMTJ fusion with a DFWO is performed to reduce sub-metatarsal head one pressure. This cut can be made subjectively, as done here, or objectively on the field with C-arm assistance. Creating a pre-operative template to excise the correct amount of bone to achieve an angle within normal limits has been recommended. Geometry can be used to plan a wedge excision for desired sagittal plane correction.⁶ The apex of the osteotomy is plantar with cuts made: (1) proximally in a dorsal-proximal → plantar distal and (2) distally in a dorsal-distal → plantar-proximal orientation (Figure 3, blue shaded region). Variations in osteotomies include a proximal cut can being made (1) dorsal → plantar, parallel with the joint (medial cuneiform cartilage) while the (2) distal cut is made distal-dorsal → plantar-proximal. Under correction (too small of a wedge removed) can result in continued deformity and ulceration while overcorrection (too large of a wedge removed) can result in transfer lesion.

The LisFranc joint connects the bases of the metatarsals to the cuneo-cuboid block with ligamentous support dorsally, plantarly, and between interosseous surfaces.⁹ The unique configuration of this joint recesses the second cuneiform several millimeters more proximally than the adjacent cuneiforms (first/third) to accept the base of the second metatarsal, increasing the joint’s overall stability.⁹ The first or medial interosseous ligament, known as LisFranc's ligament, is the strongest of the three interosseous ligaments. It arises from the distal lateral surface of the first cuneiform, inserting onto the lower-half of the medial surface of the second metatarsal base. Occasionally, secondary fibers run from the first cuneiform base to the base of the first metatarsal.⁹ This structure is vital in maintaining stability of the midfoot across the columns (ie, medial, middle, lateral), especially since there are no interosseous connections between the first and second metatarsal bases.¹⁰ It is important to consider this ligament and how it may be negatively affected in the procedure described above.

CN is a progressive deformity of bones and joints, most often seen in the foot and ankle. Rates have been recorded from 0.08% in the general diabetic population to upwards of 13% in high-risk diabetic patients.¹¹ It typically occurs 8-12 months after the diagnosis of diabetes, usually in the fifth to sixth decade of life, and males are more commonly affected than females.¹² There are two prevailing theories of its cause (ie, neurotraumatic and neurovascular) that are often intertwined.¹³⁻¹⁵ CN can be classified many ways, whether it be by radiographic findings (Eichenholz) or location
of the deformity (Brodsky, Schon).\textsuperscript{11,15} Often the
disease is precipitated by trauma and results in
soft tissue swelling, warmth, and pain especially
upon weight-bearing (Figure 9). However, in

![Figure 9. Clinical and bony changes seen with Charcot Neuroarthropathy. Reproduction of image thanks to Georgeanne Botek, DPM and illustrator Joseph Kanasz. (Botek, #12)](image)

the diabetic population, the inciting event can result from minimal to no trauma and display a
muted clinical appearance with little to no pain experienced.\textsuperscript{16} Diabetics with long-standing
and/or poorly controlled diabetes, neuropathy,
a history of ulceration, recent trauma, renal
disease, or post-transplant are more likely to
develop CN than other diabetics. This process
is often triggered by local inflammation such
as ulceration, infection, trauma, or recent foot
surgery. That being said, a reported 50%
of patients with CN do not remember any
precipitating event or trauma.\textsuperscript{13} In addition to
these clinical signs, radiographically the clinician
will see bone resorption and fragmentation, joint
dislocation to bony consolidation, osteosclerosis,
and fusion as one progresses through the
development, coalescence, and reconstruction
phases of Eichenholz.\textsuperscript{11}

The procedure performed here is a variation
of the one described by Dr. Paul Lapidus, a
fusion of the first metatarso-cuneiform joint
for first ray pathologies.\textsuperscript{17} By performing the
first TMTJ fusion with DFWO, and removing
a portion of the distal medial cuneiform, the
structural integrity of the joint is compromised.
To test intercuneiform instability at this level
while performing Lapidus surgery for hallux
valgus, Fleming et al. (2015) described an
“intercuneiform hook test.” After isolated first
TMTJ fusion in 38 patients, 28 (73.68\%) of
his patients still had gapping between medial
and central columns requiring a transverse
oriented screw to increase stability.\textsuperscript{10} Studies
have shown that first TMTJ fixation, combined
with intercuneiform arthrodesis, limits first ray
motion more so than isolated medial column
arthrodesis.\textsuperscript{16} In our instance, this instability could
either have been idiopathic or iatrogenic in origin
due to surgical technique. Regardless, it was not
taken into account to either test for and/or further
stabilize with additional fixation.

LisFranc injuries are a rare occurrence
with a slight increase seen among diabetics.\textsuperscript{16}
The injury can be osseous, ligamentous, or
both anywhere across the TMTJ. Charcot
syndrome incited by LisFranc injuries has been
documented in the literature, but to the authors’
knowledge never of iatrogenic cause.\textsuperscript{16,18,19}
There are also documented instances of CN
induced from surgical interventions including
an Austin bunionectomy for hallux abducto
valgus, Waterman-Green for hallux limitus, tibial
sesamoidectomy, first metatarsal DFWO for
sub-metatarsal head one ulceration, traumatic
Lisfranc’s dislocation repair, various forefoot
amputations, lateral calcaneal neurectomy, and
post lower extremity bypass surgery.\textsuperscript{19–21}

Differential diagnosis for acute CN should
include infection (ie, cellulitis and/or abscess),
osteomyelitis, trauma/fracture, inflammatory
osteoarthritis, rheumatologic conditions, acute
gout, tumor, and deep venous thrombosis.\textsuperscript{11,14,22}
Misdiagnosis rates have been reported at 25%
and 80\% in two different studies.\textsuperscript{11,12} In addition to
the clinical picture and history of present illness,
future laboratory (ie, CBC, BMP, ESR, CRP, Rh-
factor, uric acid, and bone culture) and imaging
(ie, MRI, PET-CT, and bone scan) modalities can
assist in coming to the correct diagnosis, whether
that be CN or another pathology.\textsuperscript{11–13} Of note, there are no laboratory criteria or hematologic markers for the definitive diagnosis of CN, but these tests can help narrow the differential list.\textsuperscript{11} If a patient with long standing, poorly controlled diabetes with neuropathy presents to the office with a red, hot, swollen foot without history of open wound, CN should be the number one differential.\textsuperscript{11,13}

After considering potential differential diagnoses for the changes seen on radiographs, along with the clinical signs and symptoms and their timing, the authors felt that this was a CN event. A point that remains for debate is whether it was the trauma of surgery, or the trauma post-operatively as a result of the altered foot mechanics and pressure distribution, that incited the process.\textsuperscript{20} Performing the curative first TMTJ fusion with DFWO caused local surgical trauma and resultant inflammation along the medial column. Also, the procedure inherently reduces the strength in the LisFranc ligament, causing a LisFranc trauma-like situation, and instability on weight-bearing. The compromise of the stability between the medial and central columns (ie, first and second rays) and post-operative weight-bearing around 8 weeks, combined with neuropathy, may have resulted in altered biomechanics, pressure distribution, and stimulated Charcot changes. Other cited cases of post-operation induced CN reported similar unique features including lack of constitutional symptoms, delayed onset of symptoms following surgery, the appearance of symptoms shortly after the start of weight-bearing, and resolution of symptoms with immobilization without administration of anti-infective agents.\textsuperscript{19}

Conservative treatment of CN consists of NWB in the acute phase, typically for 8-12 weeks.\textsuperscript{11} This is followed by protected weight-bearing in devices such as the Charcot Restraint Orthotic Walker (CROW) boot/TORCH walker for a further 4-6 months. At that point, depending on the location and severity of deformity, there may be seamless transition into custom made, extra-depth shoes with regular professional foot evaluation with management of ulcerations as needed.\textsuperscript{11,15,18} Pharmacological therapy include bisphosphonates, although the mechanism is not fully understood and usefulness is debated.\textsuperscript{11,12,22} Surgical management is typically reserved for the CN foot that has infection, is unstable or un-braceable, has stability without deformity, or is equinus. Furthermore, there has been much debate whether to perform in the acute or coalescence phases.\textsuperscript{11,13,22,23} Goals of surgery include: (1) restoring alignment and stability, (2) preventing amputation, (3) preparing the foot for bracing, and (4) promoting and allowing for ambulation.\textsuperscript{23} This can be performed through a variety of procedures ranging from simple exostectomy to complicated arthrodesis reconstructions.\textsuperscript{15}

Ultimately there is still much to learn about how to safely perform elective surgery on diabetic patients. This is especially true in high-risk patients in which elective surgery is used to prevent or cure a present problem. Furthermore, the literature on treating CN is variable. While there appears to be a general consensus on conservative treatment, surgical management, especially in the face of osteomyelitis, is highly debated.

**CONCLUSION**

Many points surfaced following these surgeries. Were the patients in the best possible pre-operation health status? Did we do too aggressive of a surgery? Was there inter-cuneiform instability intraoperatively? Should there have been a more stable fixation? Was there a component of equinus that could have been addressed to redistribute bending moment forces? Would consideration of any of these questions have changed the outcomes?

Sometimes, despite our best efforts, complications arise like CN. Here, in curing one problem (ie, the initial presenting ulceration), a new one developed—CN. Catching the Charcot changes on radiograph in the early acute stage allowed us to implement immediate treatment of offloading, TCC, and transition into custom footwear. This ultimately prevented ulcer recurrence at the initial site or ulceration elsewhere in the foot. There is wide debate over
the pathophysiology of CN and the surgeon-induced iatrogenic etiology (ie, surgical trauma versus altered foot function versus neuropathy) adds to the complexity of this pathology. These cases, as well as the others in the literature, are likely a combination of the three processes that lead to CN development.

Diabetic patients are some of the most challenging to the foot and ankle surgeon. When it comes time to perform their surgery, they should be as healthy as possible. It would be helpful to predict who will develop Charcot. However, since this is nearly impossible, surgeons can do their best through obtaining tight glycemic control, optimal vascular status, improving albumin levels, renal protection, controlling or eliminating any present infection, taking the less-is-more approach, and practicing cautious transition points post-operatively on our higher-risk patients. We should also always remember that in foot surgery, changing one part of the foot alters the function and weight-bearing forces of the remaining foot.

References


References (cont.)