

# Bilateral Charcot neuroarthropathy following successive surgical arterial revascularisation to both legs in a patient with diabetes — an interesting case history

Neil Baker and Isam Osman

Citation: Baker N, Osman I (2023) Bilateral Charcot neuroarthropathy following successive surgical arterial revascularisation to both legs in a patient with diabetes — an interesting case history. *The Diabetic Foot Journal* 26(1): 36–9

## Key words

- Charcot
- Neuropathy
- Peripheral arterial disease
- Vascular intervention

## Article points

1. A very good blood flow is a prerequisite for active Charcot development
2. Although this is a rare case, it is advised to be vigilant in monitoring a hot, swollen, inflamed foot and leg after vascular intervention in a patient with existing neuropathy
3. Early detection and immobilisation (cast) of active Charcot with regular follow up is imperative to facilitate optimal resolution and reduction in deformity
4. Differential diagnosis is essential in the presentation of a neuropathic red/ discoloured (dark), hot, swollen foot and leg, particularly if ulceration is present.

## Authors

Author details can be found on p37

**An unusually good blood supply and peripheral neuropathy are reportedly accepted prerequisites for the pathogenesis of Charcot neuroarthropathy (CN) in people with diabetes. There is anecdotal evidence that CN does not occur in the presence of peripheral vascular disease. We report the first case of a person with diabetes who had revascularisation surgery performed to both legs independently, which was subsequently followed by CN in the corresponding feet. This case illustrates the importance of a good blood supply in the development of CN. Furthermore, it supports the concept that reduced arterial supply is protective against the development of CN. This case also highlights the need for regular follow-up reviews where postoperative hyperaemia is evident.**

Charcot neuro-osteoarthropathy (CN) appears to be an uncommon diabetic foot complication which may in part be due to delay/failure to diagnose or misdiagnosis and going unrecognised, resulting in gross foot deformity. Frequently, the deformity occurs in the midfoot, producing the well described “rocker bottom foot”, however, deformity is not limited to the midfoot.

Characteristically, CN presents in those with neuropathy as a pink, warm/hot and swollen foot, and lower leg. Paradoxically, pain may be described but is not a common feature; however, when present, it is frequently described as a dull ache in the lower leg. This latter symptom in conjunction with a swollen warm limb sometimes leads to the misdiagnosis of deep vein thrombosis (DVT). Additionally, deformity may sometimes be present, generally in the mid-foot which may be subtle. An increased skin temperature of 2°C at local site(s) in the affected foot compared with identical contralateral locations has been used as a diagnostic

threshold for active CN (Jones et al 2023). In our experience, however, it is more likely to observe temperature differences of 4–6°C.

X-ray examination in the early stages will show generalised demineralisation and typically fractures of the metatarsals or tarsal bones. One early radiological sign that can be seen before fractures or bony destruction is a gap at the base of the first and second metatarsals, signifying rupture of the Lisfranc ligament.

The true incidence and prevalence of CN in diabetes mellitus is unknown. There are UK reported prevalence rates of 0.04% pooled data from seven foot-care specialist centres in England (Metcalf et al, 2018), 0.3% at a regional referral centre in Ireland (O’Loughlin et al, 2017) and 0.53% from a national registry study in Denmark (Svendsen et al, 2021). Thus, it appears that although diabetic peripheral polyneuropathy is common, CN appears to be comparatively rare. The exact pathogenesis of CN is still not fully

understood; however, the presence of an unusually good blood supply and neuropathy are accepted to be prerequisites (Rajbhandari et al, 2002).

A novel theory (Jeffcoate et al, 2005) has clearly directed that CN is to be viewed as an inflammatory process affecting bone and soft tissues in persons with peripheral polyneuropathy. Subsequent studies have supported this theory of CN being an inflammatory condition (Mabilleau et al, 2008; Uccioli et al, 2010; Petrova et al, 2014; 2015; Jeffcoate and Game, 2022). This again supports the necessity of good arterial supply to mount this extraordinary and prolonged inflammatory response to unperceived injury.

Sadly, due to the abnormal inflammatory nature of this condition, delayed diagnosis and suboptimal management, the acute phase of CN can be protracted. In these situations, this may result in gross foot deformity, which in some instances can warrant lower-limb amputation. That aside, the resultant deformed and biomechanically dysfunctional foot is prone to the abnormal plantar forces, resulting in difficult to heal foot ulcers and footwear fitting problems. There has been a reported 6 to 12 times increased risk of major amputation in those ulcerated CN compared to those without ulceration (Wukich et al, 2017; Sohn et al, 2010). Furthermore, pooled data from studies estimate a mean 5-year mortality of 29% in those suffering with CN (Armstrong et al, 2020).

Anecdotally, it is accepted that CN does not occur in the presence of peripheral vascular disease and, in fact, a number of studies have demonstrated a greater hyperaemic microvascular blood flow in patients with CN compared with those with neuropathy alone (Shapiro et al, 1998; Veves et al, 1998; Baker et al, 2007).

We report a unique case report of CN, following arterial bypass surgery in both limbs, which supports the concept that a good blood supply is necessary in the development of CN.

## Case report

A 61-year-old female presented with a 1.5cm<sup>2</sup> neuro-ischaemic ulcer of the plantar surface of the right first metatarsal head associated with intermittent claudication in that the same leg. She had a 22-year history of type 1 diabetes complicated by peripheral polyneuropathy, retinopathy, nephropathy,

peripheral arterial disease (PAD), hypertension and ischaemic heart disease.

Neuropathy was confirmed by an inability to detect vibration and 10g pressure. Clinically PAD was evident by a weakly palpable right posterior tibial pulse, absent dorsalis pedis and intermittent calf claudication. The ankle brachial pressure index (ABPI) measured at the posterior tibial artery was 0.58 on the right and 0.9 on the left, confirming right-sided arterial disease.

A femoral angiogram demonstrated a significant and moderate stenosis in the right superficial femoral artery (SFA) and the distal popliteal artery, respectively. The SFA was successfully treated with a transluminal angioplasty with subsequent ulcer healing. Unfortunately, 6 months later, the claudication and ulcer re-occurred. Repeat femoral angiogram revealed a moderate stenosis in the right external iliac artery, re-stenosis of the SFA and a complete occlusion of the distal popliteal artery. The SFA was successfully treated again by transluminal angioplasty; however, following a multidisciplinary vascular radiology team meeting, this was considered insufficient and a right femoro-popliteal bypass was subsequently performed. The patient recovered uneventfully and, with offloading and regular wound-care, her ulcer healed within 6 weeks.

Two years later, she developed a hot, painful, right foot. There was no history of trauma but her foot was swollen and 4 °C hotter than the contralateral limb, and had a subtle deformity of medial cuneiform-first metatarsal joint. A Doppler ultrasound examination of the right leg confirmed that the graft arteries were still patent and the ABPI was 1.03. A foot X-ray showed a fracture of the medial cuneiform with widening of the bases of the first and second metatarsals characteristic of Lisfranc ligament rupture (*Figure 1*).

A diagnosis of CN was made and the limb was immobilised for approximately 8 months until the CN became quiescent; despite this, her right mid-foot collapsed causing the typical deformity of CN. She has had no further problems with this foot being managed by regular podiatry and bespoke footwear.

Later that year, she developed a new ulcer on the heel of her contralateral left foot. Her pedal pulses were palpable but her ABPI on that side had

## Authors

Neil Baker is Consultant Diabetes /Vascular Podiatrist, Sana Clinic Al Shaab Kuwait; formerly Ipswich Hospital NHS Trust; Isam Osman is Consultant Vascular Surgeon, King Saud Medical City, Riyadh, Saudi Arabia; formerly Ipswich Hospital NHS Trust;

Figure 1. Plain X-ray to show early signs of Acute Charcot — rupture of Lisfranc ligament.



Figure 2. Plain X-rays of Charcot in both feet.

deteriorated within 12 months from 0.87 to 0.5. A femoral angiogram showed multiple stenoses in the left SFA and a long occlusion of the popliteal artery. These were successfully treated with a fem-distal bypass. Despite the ulcer healing well, 8 weeks later, she developed pain and swelling in the left mid-foot which was found to be 3°C hotter than the contralateral limb with no obvious deformity.

Initially, infection was considered, but microbiological cultures of biopsied tissue were negative. The foot X-ray showed fractures at the base of the second and third metatarsals with no features consistent with infection anywhere in her foot. The affected limb was immediately placed into a below-knee cast with weekly changes for wound care to her heel ulcer that healed uneventfully. Serial X-rays showed typical changes in keeping with CN, thus confirming our diagnosis of CN. Despite immobilisation within a non-removable cast, she developed characteristic mid-foot deformity. She remained in a cast for 6 months while waiting for her skin temperature to reduce to that of her contralateral limb. She was provided with made-to-measure footwear and orthoses, and had no further foot ulcers or CN relapse.

## Conclusion

This case clearly illustrates the importance of a good blood supply in the development of CN and, to our knowledge, is the first reported case of bilateral CN following revascularisation to both legs. It also shows that regular follow-up following revascularisation is essential, not just to review flow patency, but in those with peripheral neuropathy. ■

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