

Review

Charcot Neuroarthropathy in Patients With Diabetes: An Updated Systematic Review of Surgical Management



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ABSTRACT

Charcot neuroarthropathy (CN) of the foot and ankle is a demanding clinical dilemma, and surgical management can be very complicated. Historically, the evidence guiding surgical management of CN has been small retrospective case series and expert opinions. The purpose of the present report was to provide a systematic review of studies published from 2009 to 2014 and to review the indications for surgery. A Medline search was performed, and a systematic review of studies discussing the surgical management of CN was undertaken. Thirty reports fit the inclusion criteria for our study, including 860 patients who had undergone a surgical procedure for the treatment of CN. The surgical procedures included amputation, arthrodesis, debridement of ulcers, drainage of infections, and exostectomy. The midfoot was addressed in 26.9% of cases, the hindfoot in 41.6%, and the ankle in 38.4%. Of the 30 studies, 24 were retrospective case series (level 4), 4 were controlled retrospective studies (level 3), and 2 were level II studies. The overall amputation rate was 8.9%. The quality of the published data on the surgical management of CN has improved during the past several years. Evidence concerning the timing of treatment and the use of different fixation methods remains inconclusive.

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Charcot neuroarthropathy (CN) of the foot and ankle is a destructive process that poses an immense challenge to foot and ankle specialists. The development of neuropathic fractures and/or dislocations in the foot and ankle predisposes the patient to increased morbidity, decreases patient-reported quality of life, and increases the risk of foot ulceration and potential for major amputation (1,2). Early recognition and intervention is imperative to prevent permanent deformity, ulceration, and the possibility of limb loss.

Although any disease process that results in peripheral neuropathy can lead to CN, this condition is currently most closely associated with diabetic peripheral neuropathy and was first linked to diabetes mellitus (DM) by William Riely Jordan in 1936 (2). The American Diabetes Association has estimated that 29 million people in the United States, or 9.8% of the population, have DM (3). Of these patients with DM, 14.5 million people, or 4.9% of the

population, will develop peripheral neuropathy (4). The precise incidence of CN is unclear but might affect 8.5 per 1000 people with DM annually (5).

The initial treatment of CN in the foot and ankle traditionally has been nonoperative, using offloading devices such as total contact casting, Charcot restraint orthotic walker devices, and bracing (2,6). The goal of treatment, whether operative or nonoperative, is to achieve a plantigrade foot with osseous stability (2,7). A stable plantigrade foot can reduce the likelihood of ulcer formation, which, in turn, can reduce the rate of infection and amputation. Pinzur (8) has demonstrated that 60% of patients with midfoot CN achieved a desired endpoint without the need for surgery. However, when surgical intervention is necessary, the published data to guide treatment decisions have been based on noncontrolled retrospective case series, case reports, and expert opinion (9).

A systematic review by Lowery et al (9) examined all the published data regarding the surgical management of CN from 1960 to 2009. At that time, all published data on this topic was from retrospective case studies, expert opinions, or case reports (level 4 or 5 evidence). No study directly compared different fixation methods, timing of surgical intervention, or the outcomes of patients who had undergone amputation. The purpose of the present systematic review was to provide an update on the current trends regarding the surgical

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management of CN and to determine whether the quality of the published data has improved since 2009 (9).

Materials and Methods

A Medline search was performed from July 1, 2009 to June 23, 2014 using the key terms of *Charcot*, *arthropathy*, *neuroarthropathy*, *neuro-osteoarthropathy*, and *surgery*. The abstracts of all studies cited were reviewed, and the studies that reported on the surgical management of CN were included for further review.

Inclusion Criteria

Included in the study group was any report written in English that discussed the surgical management of CN of the foot and ankle secondary to DM, including reviews and retrospective case series.

Exclusion Criteria

Excluded from the study group was any report that was not written in English and any that did not discuss the surgical management of CN. Studies that discussed patients with nondiabetic causes of CN (ie, leprosy, syringomyelia, syphilis, and alcohol) were excluded as were those describing CN in areas of the body other than the foot and ankle.

Results

The search was performed in June 2014. For the purposes of the present study, the terms *Charcot arthropathy*, *neuroarthropathy*, and *neuropathic arthropathy* were used interchangeably. The search results are summarized in Figs. 1 to 3. A total of 209 reports were cited during a 5-year period (2009 to 2014). Of these reports, 30 met the criteria for inclusion in the present review (10–39). Of the 30 studies, 2 (6.6%) were level 2 prospective comparative studies, and 4 (13.3%) were level 3 retrospective case-controlled studies. The remaining 24 studies (80%) were level 4 case series. Level 5 studies were not included for statistical interpretation in the present study; however, some expert opinions were used for discussion purposes.

The surgical procedures performed included amputation, arthrodesis, debridement of ulcers, drainage of infections, and exostectomy. The use of internal or external fixation and the need for posterior muscle group lengthening were recorded.

Overall, 860 patients were reported to have undergone a surgical procedure for the management of CN during the study dates. The number of patients reported per study ranged from 1 to 195, with a mean and median number of patients per study of 28.6 and 11, respectively.

Of the 860 procedures, 330 involved the ankle (38.4%), 358 involved the hindfoot (41.6%), 231 involved the midfoot (26.9%), and 2 specifically involved the forefoot (0.2%). The percentages do not equal 100%, because some patients required surgery at >1 anatomic region. In 3 studies, the exact location of the surgery was not identified. These

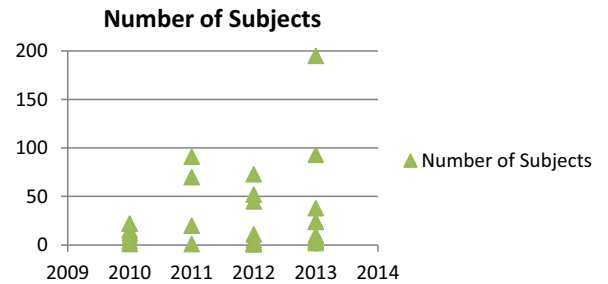


Fig. 2. Number of subjects per study.

3 studies accounted for a total of 278 surgical procedures. Of these surgeries, 307 used internal fixation (35.7%) and 427 involved the application of an external fixation device (49.7%). In addition, 196 of 860 (22.8%) also included tendo-Achilles lengthening. Of the 30 studies, 22 included joint arthrodesis as a treatment option, either alone or in addition to another procedure. A total of 77 amputations were performed, for an overall amputation rate of 8.9%. Not all the studies were consistent in defining fusion using radiographic or clinical parameters; thus, a determination of nonunion rate would be difficult.

Two studies compared the use of internal versus external fixation (15,17). One additional study discussed surgical versus nonsurgical treatment options (11). Also, 2 studies compared surgical correction versus amputation for patients with CN (11,29). One report discussed CN of the first metatarsophalangeal joint, which was treated with arthrodesis through a dorsal locking plate (38). Also, 1 study directly compared the cost of limb salvage versus amputation in patients with CN (21).

Discussion

Patients with CN encounter increased morbidity and a decreased quality of life (1). The Medical Outcomes Study 36-item Short Form survey physical component scores in patients with CN are 1 standard deviation lower than those of patients with end-stage renal disease requiring hemodialysis, cardiovascular disease, and Parkinson's disease. Another recent study reported that patients with CN had Foot and Ankle Ability Measurement scores that were 2 standard deviations lower than a control group of diabetic patients without CN (40). Patients with CN have an increased risk of major amputation, with rates as high as 28% if ulceration is present on the initial evaluation (41). The amputation rate in the present systematic review was 8.9%, similar to the 7% rate reported in an additional study (41). Sohn et al (42) found that patients with CN patients and foot ulcers were 12

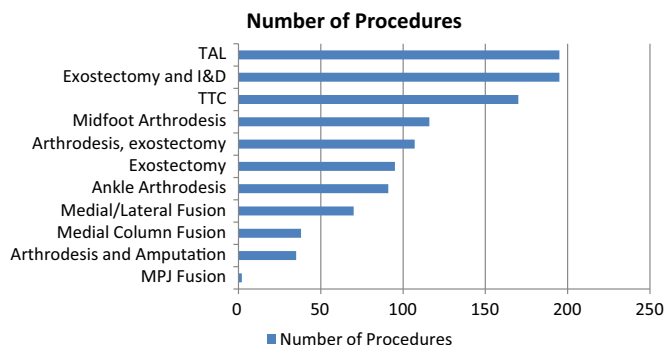


Fig. 1. Number of described surgical procedures. I&D, incision and drainage; MPJ, metatarsophalangeal joint; TAL, tendo-Achilles lengthening; TTC, tibiotalarcalcaneal.

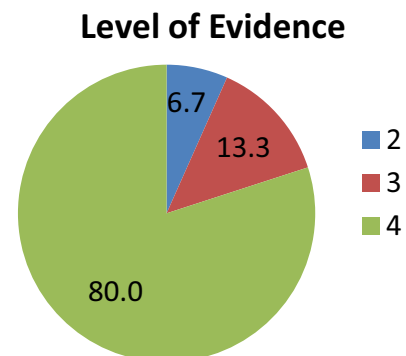


Fig. 3. Evidence levels of studies discussing surgical management of Charcot neuroarthropathy.

times more likely to require a major amputation than were patients with CN but without foot ulcers.

The anatomic location of CN has been classified by several investigators, most notably Brodsky (43) and Sanders and Frykberg (44). In both of these classification schemes, the most common site of occurrence for CN is the midfoot, and involvement of the ankle is less common. The present systematic review of 860 cases has demonstrated that during the past 5 years, the most commonly reported location for surgical management of CN was the hindfoot, with a total of 358 surgical procedures (41.6%), followed by the ankle with 330 surgical procedures (38.4%), and the midfoot with 231 procedures (26.9%). This highlights that, although midfoot and hindfoot CN is far more common than ankle CN according to the anatomic classification, the midfoot might be more amenable to bracing and other nonoperative treatment methods than cases in which the ankle is affected (8).

Of the 860 cases, 170 involved tibiototalcalcaneal (TTC) arthrodesis, accounting for 19.8% of the cases. This might indicate that TTC arthrodesis is becoming a more common procedure, with more surgeons using this technique for CN involving the ankle and hindfoot. The next 2 most common procedures in our study were exostectomy with incision and drainage and midfoot arthrodesis, with 195 (22.7%) and 116 (13.5%) cases, respectively.

In our previous study, we found no direct comparisons regarding the performance of surgery in the acute phase versus performing surgery when the foot or ankle has consolidated in the chronic phase. The present search also found no such comparisons. Currently, 2 studies (24,45) discussed surgical correction of acute CN, with a total of 18 patients examined. The ideal timing for surgical intervention remains unclear.

The present review of the 30 studies published in the previous 5 years identified 24 level 4 studies (80%), 4 level 3 studies (13.3%), and 2 level 2 studies (6.7%). In our previous review, which covered 49 years of research, 42 of the 96 articles were level 5 (44%) and 54 of 96 were level 4 (56%). No level I, II, or III studies were included. It is encouraging to discover that the quality of evidence that is guiding surgical treatment could be improving.

Our search identified 1 study that compared the cost of limb salvage versus amputation in patients with CN. Gil et al (21) studied 93 patients, 76 of whom had undergone CN foot reconstruction with application of circular external fixation and 17 who required transtibial amputation. The cost of care was collected for a 12-month period and included inpatient hospitalization, placement in a rehabilitation or skilled nursing facility, home health care, physical therapy, and the purchase of a prosthetic device or footwear. The average cost of care in the limb salvage group and amputee group was \$56,712 and \$49,251, respectively. These results were not significant statistically, and patient quality of life and mortality were not discussed in their study.

Wukich and Pearson (46) examined amputation as a primary option in patients with nonreconstructable CN to determine whether self-reported outcomes improved after transtibial amputation. Preoperative and postoperative assessments were accumulated using the Medical Outcome Study Form 36-item health survey and the Foot and Ankle Ability Measure. The study included a total of 13 patients. The mean follow-up period was 79 (range 53 to 122) weeks. The results of the study demonstrated significant improvement in the physical component self-reported outcomes after transtibial amputation. The self-reported mental quality of life also improved; however, the difference did not reach statistical significance with the numbers available. The conclusion of the study indicated that amputation has a role for selected high-risk patients with nonreconstructable CN deformities.

Palena et al (29) studied the presence of peripheral arterial disease in diabetic CN. Their aim was to evaluate whether revascularization was beneficial before surgical correction of CN deformity. Data were retrospectively collected from patients who had CN with limb ischemia and had undergone endovascular revascularization followed

by surgical treatment. The data from 10 patients were reviewed, and the limb salvage rate was 90%. One patient (10%) developed an infection that could not be controlled, and a transtibial amputation was performed. Critical limb ischemia can occur in patients with CN, and revascularization might be required to optimize the results of surgical intervention.

A rare case of metatarsophalangeal joint involvement was also discussed by Wunschel et al (38). The patient was a 43-year-old male who had ulceration to the plantar aspect of his interphalangeal joint of the hallux measuring 2 × 2 cm. He had a 5-year history of peripheral neuropathy. The ulceration had been present intermittently for 2 years and had been treated with local wound care and a reduction in weightbearing status. Radiographs showed significant progressive destruction to the first metatarsophalangeal joint. The patient underwent surgical correction, which included ulcer debridement, arthrodesis of the first metatarsophalangeal joint with a dorsal locking plate, resection of hypertrophic bone, and primary closure of the ulceration. No evidence of bone infection was found intraoperatively, and microbiologic cultures were negative for osteomyelitis. The patient healed uneventfully and had no signs of ulceration 1.5 years after surgery.

DeVries et al (17) compared the use of internal versus external fixation of CN in the ankle. They surgically stabilized 52 deformities with an intramedullary arthrodesis nail, 7 of which had the addition of a circular external fixator. The endpoint of their study was either major amputation or a braceable limb. A major amputation was performed in 10 of 45 patients (22.2%) in the intramedullary nail cohort and 2 of 7 patients (28.6%) in the combined intramedullary nail and external fixator group. The addition of the circular external fixation device did not affect the overall limb salvation rate or complication rate (17). Crim et al (15) also discussed the various fixation techniques available, focusing on locking plate technology, solid fixation bolts, and intramedullary superconstructs.

The term *superconstruct* is defined by 4 factors:

1. Fusion is extended beyond the zone of injury to include joints that are not affected to improve fixation.
2. Bone resection is performed to shorten the extremity to allow for adequate reduction of deformity without undue tension on the soft tissue envelope.
3. The strongest device is used that can be tolerated by the soft tissue envelope.
4. The devices are applied in a position that maximizes mechanical function.

Superconstructs are warranted in cases with a high likelihood of failure and are appropriate in the setting of dysvascular bone, bone loss, deformity correction, and severe osteoporosis and in patients with multiple medical comorbidities that can lead to delayed healing (47). With the use of these techniques, multiple joints can be fused to correct the deformity, with large spanning fixation. Sammarco (47) identified 3 superconstructs in the reconstruction of midfoot Charcot deformity: plantar plating, locking plate technology, and axial screw fixation.

Aragon-Sanchez et al (11) reported on 2 cases of CN triggered and complicated by osteomyelitis and how limb salvage was achieved. No offloading had been performed before presentation, and major amputation had been indicated in both cases. Four major steps were used for limb salvage:

1. Partial removal of infected bone by curettage
2. Culture-guided postdebridement antibiotic treatment
3. Bed rest before placement in a total contact cast
4. Stabilization of the unstable foot using a total contact cast with an opening for performing wound care and to check healing

Table
Indications for surgery and surgical options

Surgical Indications	
Nonhealing foot ulcer or impending ulceration	
Osteomyelitis	
Pain	
Surgical options	
Exostectomy	
Realignment arthrodesis	
Internal/external fixation	
Amputation	

Both patients also received intraosseous instillation of Dermacyn[®] (Oculus Innovative Sciences, Herten/Roermond, The Netherlands) wound care, which were received as free samples from the company; however, the number of cases was inadequate to draw any conclusions regarding the value of this adjunct therapy.

Wukich et al (48) evaluated weightbearing radiographs from patients with and without foot ulcers diagnosed with midfoot CN secondary to DM. They identified and studied 114 patients with midfoot CN (50 with foot ulcers and 64 without ulcers). Of the patients with foot ulcers, 24% had a lateral talar first metatarsal angle of less than -27° , and 80% had a negative cuboid height. Patients with CN and foot ulcers had significantly greater deformity than did patients with CN without ulcers when measuring the lateral talar-first metatarsal angle, calcaneal pitch, cuboid height, lateral calcaneal-fifth metatarsal angle, talar declination angle, and lateral tibiotalar angle. Sagittal plane deformities were more likely to be associated with foot ulceration in patients with CN than were transverse plane deformities. Lateral column involvement was associated with a worse prognosis than medial column involvement, leading us to believe that progressive deformity of the lateral column should be monitored closely to prevent foot ulceration. Lateral column involvement can be identified by a decrease in the cuboid height, decreased calcaneal pitch, and decreased lateral calcaneal fifth metatarsal angle. Their study can assist physicians in evaluating the risk of both ulceration and the need for surgery in patients with CN using reproducible radiographic measurements (48).

In a previous systematic review from 1960 to 2009, a total of 897 procedures met the inclusion criteria. Of the 897 procedures, 534 involved the midfoot, 263 involved the ankle, and 100 included the hindfoot (9). When including the findings from our previous study with those from the present study, a total of 1757 patients were included. Of the 1757 procedures, 765 involved the midfoot (43.5%), 458 involved the hindfoot (26.1%), and 593 involved the ankle (33.8%). A total of 88 major or minor amputations were present in the 2 combined studies (5%). The indications for surgery and the surgical options are summarized in Table.

- The reports of surgical intervention during the acute phase of CN are encouraging, but no direct comparisons have been performed during the past 5 years comparing early and late surgery; therefore, evidence for the performance of surgery during the acute phase is inconclusive at present.
- The most common locations requiring surgical intervention based on a 54-year review (1960 to 2014) for patients with CN is the midfoot (43.5%), and the second most common location is the ankle (33.8%). During the past 5 years, the most common locations requiring surgical intervention for patients with CN was the hindfoot (41.6%), and the second most common location is the ankle (38.4%). Although midfoot and hindfoot CN is far more common than ankle CN according to the anatomic classification, the midfoot might be more amenable to bracing and other nonoperative treatment methods than cases in which the ankle is affected.

- Arthrodesis is useful in patients with instability, pain, or recurrent ulceration that fail nonoperative treatment. An increasing trend was seen during the past 5 years for TTC arthrodesis. This might indicate that TTC arthrodesis is becoming a more common procedure, with more surgeons using this technique for CN involving the ankle and hindfoot.
- One study in the past 5 years discussed a comparison between internal and external fixation techniques for surgical reconstruction of the foot and ankle in patients who are not infected. The addition of the circular external fixation device did not affect the overall limb salvation rate or complication rate. Although it is encouraging to see these data, overall, the data continues to be too inconclusive to recommend 1 form of fixation over another.
- The present review identified 24 level 4 studies (80%), 4 level 3 studies (13.3%), and 2 level 2 studies (6.7%). In our previous review, which covered 49 years of research, 42 of the 96 studies were level 5 evidence (44%) and 54 of 96 were level 4 evidence (56%). No level 1, 2, or 3 studies were found. It is encouraging to discover that the quality of evidence guiding surgical treatment could be improving.

In conclusion, the results from our present review suggest that the published surgical data for CN are improving, evidenced by higher level studies during the past 5 years. Also encouraging is that published data now exist that compares fixation techniques, reconstruction and amputation, and cost evaluations of limb salvage. Arthrodesis, specifically TTC, seems to be gaining popularity as a surgical treatment option for CN. However, no randomized, prospective, multicenter trials have yet been published regarding this topic, and the proper timing of surgery remains undefined. The goal of treatment, whether nonoperative or operative, is to achieve a plantigrade, stable foot that remains ulcer free. If the CN deformity involves more proximal anatomic regions (ankle and hindfoot), the need for surgical intervention becomes more likely. Despite modern techniques using improved methods of fixation and improved patient selection, approximately 9% of patients with CN who undergo surgery will require a major amputation.

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