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Case Report

Different approach to charcot neuroarthropathy: A case report

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ABSTRACT

Introduction and importance: Charcot neuroarthropathy (CN) is a degenerative, progressive disease affecting the ankle and foot and it is usually a disabling factor in diabetic patients. Surgical management of CN aims to obtain a painless stable plantigrade foot which can be achieved through fusion. Achieving joint arthrodesis in CN usually carries a high failure rate.

Cases presentation: We presented two patients with late-stage CN foot deformity. The first case is a 52-year-old female with CN on her left ankle and presented without any infection or prior correction. The second case reported a 47-year-old man with complaints of deformity on his right ankle, he had undergone surgical treatment with an external fixator before, and now presented with infection in the surgical site.

Clinical discussion: Ankle arthrodesis has been considered by many as the treatment of choice for severe and late-stage CN foot. This treatment aims to give a rigid enough fixation which will maintain the stability of the ankle joint and prevents further destruction of surrounding tissue. Multiple modalities of treatment are available and must be chosen accordingly to each clinical case. Minimal implants and the use of multiple bone grafts could be considered as a plan of treatment. Both patients have promising and positive results from the two procedures.

Conclusion: Treatment of CN Foot with internal plate fixation combined with fibular strut graft seemed to give promising results, both radiographically and functionally. Furthermore, a slight modification of treatment with a minimal implant or iliac graft may be considered.

1. Introduction

Charcot neuroarthropathy (CN) is a degenerative, progressive disease affecting the foot and ankle and it is usually a disabling factor in diabetic patients. Charcot neuroarthropathy was first described by neurologist Jean-Martin Charcot in 1868 [1]. This disorder is often initiated by trauma to a neuropathic extremity, the trauma then progresses to the bones and joints of the extremity leading to a limb-threatening condition seen in the late complication of diabetes [2]. Two main theories are proposed to explain the development of Charcot neuroarthropathy. The neurotraumatic theory states that joint destruction is the result of cumulative trauma unrecognized by an insensate foot. In contrast, the neurovascular destruction theory states that bone resorption and ligament laxity are secondary to a neural-controlled vascular reflex. Most experts believe that a combination of these pathways is responsible for the destruction seen in Charcot foot [3]. Although the incidence rate of CN is less than 1% in the general population, it rises to as high as 13% in High-risk patients with diabetes, with equal risk between men and women [1].

The aim of treating this deformity is to achieve a plantigrade, stable foot that can fit into shoes and to also prevent recurrent ulceration [3]. The treatment method for these deformities depends on many factors, such as the location, phase of the disease, deformity, the presence of an infection, and other comorbidities. Modalities of treatment vary from basic shoe modification to more radical limb amputation [3]. In most cases, ankle arthrodesis has been considered to be the treatment of choice for severe and late-stage CN foot. To achieve ankle arthrodesis, fixation is needed and thus implants are used. However, the infection rate of CN is quite high, therefore minimizing the use of implants is mandatory. Another material that is needed to support the fixation and helps with osseointegration is the fibular strut graft which has been generally used to fill defects and further strengthen the construction as it is used as a cortical allograft with full-thickness [4].

Through this case reports, we intend to give more perspective of the treatment for both cases, in which both are treated using internal fixation with fibular strut graft and also iliac bone graft but with a slightly

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Fig. 1. The clinical picture of the patient's left ankle.

Fig. 2. X-ray imaging of the patient's left ankle.

Fig. 3. X-ray imaging of the patient's left foot.

different approach. These two cases provide us with the comparison of the treatment modalities for CN foot.

2. Methods

This paper has been reported in line with the SCARE Criteria [5]. This paper reported two cases retrospectively performed in a single center by a foot and ankle orthopaedic surgeon. Our center is a learning-based hospital located in Bali state at the tertiary level – Sanglah Public Hospital. Both cases are diagnosed with Charcot neuropathy and non-consecutive case. The management of the patient was fibular strut graft and arthrodesis performed by single orthopaedic surgeon.

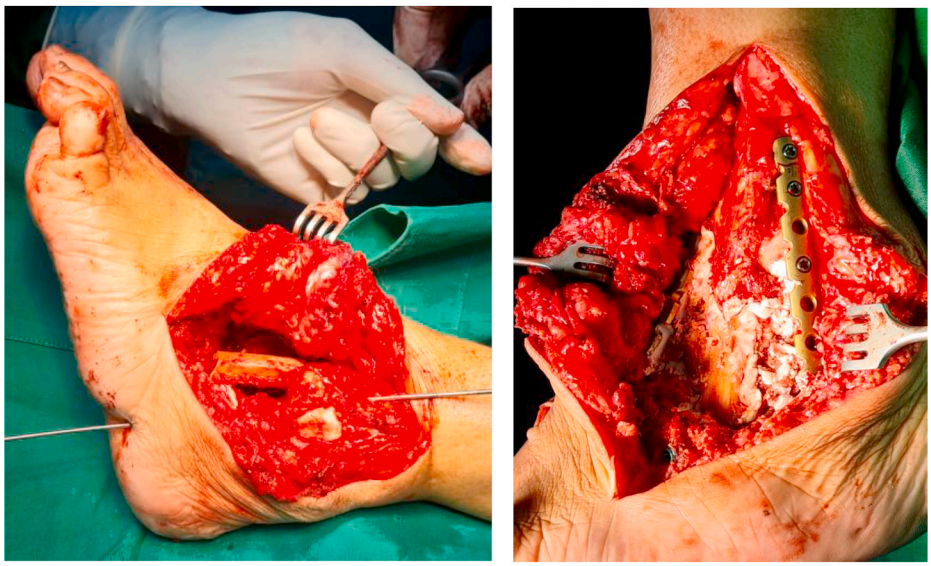
3. Case 1

A 52 years old female came to the clinic with a crooked left ankle that are progressive since 2 years before. She also said that the deformity was initially started with pain and swelling which increasing progressively. The patient also complained that she could not walk properly and need assistance to mobilize. She had a history of diabetes mellitus type 2 for the last 20 years and routinely treated by insulin. Patient also reported that there are no family history of charcot arthropathy and diabetes mellitus. The patient confirmed that she followed the treatment by the previous doctor in the past adherently. Patient has no history of drug abuse, alcohol abuse, and patient socioeconomies status is fairly well. From the physical examination, we found obvious deformity on the left ankle including the foot that was characterized by bony prominence on the medial-lateral side with flattened on the plantar side (rocker bottom type) without any sign of inflammation and infection process (Fig. 1).

From radiographic imaging, we could see the destruction of the left ankle joint with the absence of the talus which is associated with the absence of tibiotalar and subtalar joints (Fig. 2). From the foot X-ray, we could see the bony resorption of the midfoot region that hardly recognized which bones are involved and which are preserved of forefoot region without any bony deformation (Fig. 3).

The patient was diagnosed with Left Charcot Joint Eichenholtz Classification Grade 2. From laboratory results, we found that random blood glucose is 148 mg/dL and HbA1C is 8.4%.

During surgery, we did a lateral incision approach. Firstly we found the distal part of the tibia-fibula and tarsal bone such as talus, cuboid, navicular are already absorbed and changed into fibrous tissue. We did the debridement of all of the devitalized soft tissue on the bone followed by arthrodesis of the ankle and the midfoot. We harvested fibular strut graft about 10 cm in height and amount of cancellous graft from iliac



A B

Fig. 4. (A) Clinical picture of fibular strut graft implanted to the bony defect area on the midfoot. (B) Clinical picture after internal fixation applied (with locking plate and screw).

Fig. 5. Left ankle X-ray postoperatively.

bone to fill the defect caused by bone removal. We put the strut graft for bridging the defect between the distal part of the tibia to the tarsals bone. For the final stage of the procedure, we apply pantalar arthrodesis with rigid internal fixation (Fig. 4). We used 2 pieces of reconstruction locking plate on the anterior and lateral side of the ankle joint. For adding the rigidity of fixation, 2 pieces of locking screw were applied on the lateral and medial sides (inserting from 1st and 5th metatarsal). After the procedure, we put the half-circular cast on plantigrade position. (Fig. 5). The patient had no complications after the surgery.

For the post-operative rehabilitation, we have informed the patient to do a non-weight bearing procedure with two crutches and maintain the cast for at least 2–3 weeks. The weight-bearing program will adjust with clinical and radiographic evaluation each month after the procedure.

We did a regular observation each month to the patient's condition clinically and radiographically. We found that 3 months after procedure, from radiograph imaging showed favourable results, the implant still in good alignment and no sign of failure and we suggested the patient to do a partial weight bearing when walking. (Fig. 6). The patient also stated that she felt better after the surgical procedure, that the foot is looking better and she can do daily activity with minimal assistance compared before surgery, and the pain also subsided gradually

4. Case 2

A 47-year-old man visited the clinic complaining of pain in his right

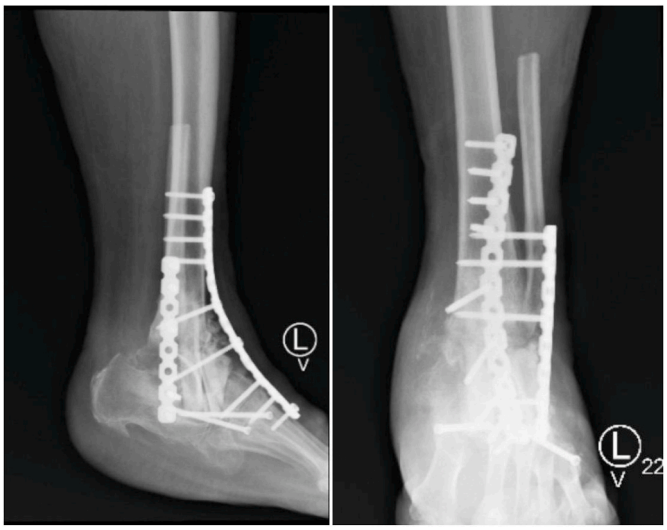


Fig. 6. Left ankle X-ray after 3 months follow up.

ankle since April 2019. The right ankle got swollen and had a wound that was infected later on. The patient also has diabetes mellitus type II which was controlled with insulin injection. The patient had neither history of smoking neither other comorbidities. No family history confirmed by the patient. Patient had no history of alcohol and drug abuse. He also had no history of allergies, and adverse reactions. A radiograph was taken, and the result was that there was some destruction of the tibia, fibula, calcaneus, talus, navicular, cuboid, cuneiform medial and lateral, also narrowing of joint and dislocation of ankle joint. All of these support the diagnosis of right charcot joint (Fig. 7).

The patient had an external fixation before to stabilize his ankle joint while waiting for the inflammation to subside. This external fixation construct got infected and had two distal screws loosened and was revised with another external fixation along with debridement and implantation of bone cement spacer 3 months later.

After a routine check-up to the orthopedic clinic and good compliance, the patient's infection was controlled and it was decided to perform arthrodesis of the right ankle. From the preoperative physical examination, it was seen that the external fixation had some distal screws loosened. There was discharge coming out from the pin site and an open wound with exposed bone at the lateral side of the ankle (Fig. 8 and 9). The patient could not actively move the ankle due to the external fixation construct. Also, there was contracture of the metatarsophalangeal and interphalangeal joint at all toes. The radiograph showed the external fixation in a good position with the ankle joint space narrowed and destruction of the tibia bone, fibula, calcaneus, talus, navicular, cuboid, cuneiform medial, and lateral. However, there were no further signs of infection (Fig. 10).

During surgery, the patient was positioned in a supine position. The first incision was performed on the lateral calcaneus, followed by the release of fibrotic tissue. The next incision was through the anterior approach to perform distal tibia and superior talus osteotomy. We also performed a lateral surgical approach to the fibula middle third and osteotomized the distal fibula for 10 cm. Fibular strut graft fixation was

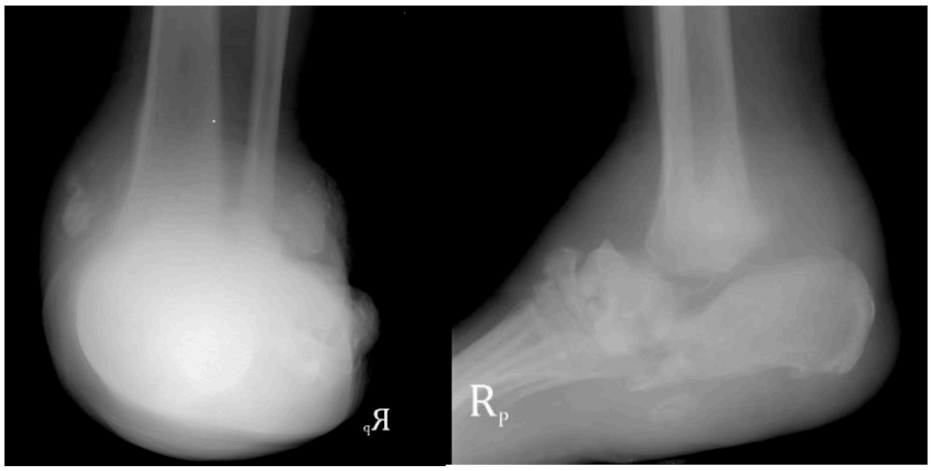


Fig. 7. Right ankle radiograph of anteroposterior and lateral view showing the destruction of the tibia, fibula, calcaneus, talus, navicular, cuboid, lateral, and medial cuneiform. There was also narrowing of joint and dislocation of ankle joint.

Fig. 8. The clinical picture of the right ankle with external fixation, pin site infection and two distal screws removed.

Fig. 9. The revised external fixation had the distal screws loosened with an open wound on the lateral side of the foot exposing the bone. There was contracture of the metatarsophalangeal and interphalangeal joint at all toes.

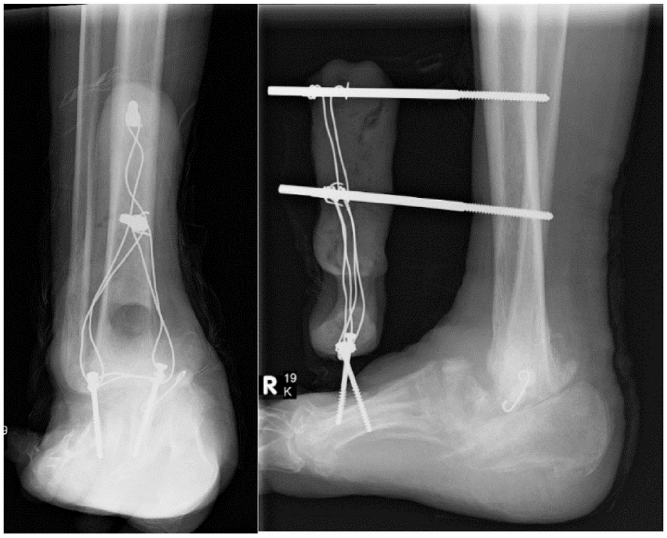


Fig. 10. Right ankle radiograph of anteroposterior and lateral view showing revised external fixation all in a good position. The joint space narrowed compared to 2 months prior, however, there were no further signs of infection surrounding the bone.

performed on the ankle, followed by tibiotalar and talonavicular arthrodesis with a distal tibia locking plate (Fig. 11). The postoperative ankle showed acceptable alignment of the tibia, the fibular strut graft, and the implant (Fig.12). There was enough soft tissue to cover the surgical wound (Fig. 13). The patient had no complications after the surgery.

The patients come in for routine follow up every month after the procedure for wound care and clinically and radiographically evaluation. At the 3rd months after the operation, the infection has subsided, and the wound looks significantly improved (Fig. 14). The patients was given the instruction for a non-weight bearing treatment for the leg. The follow up radiograph shows promising results (Fig. 15), and the patients now progresses to do a partially weight bearing and continue to gives promising results. Patient also confirmed that the pain was significantly reduced compared to before the procedure.

5. Discussion

Charcot neuroarthropathy cases presents a challenge for limb salvage in diabetes mellitus patients. It presents with hard to diagnose signs and symptoms and could be misdiagnosed with simple sprains, deep vein thrombosis, osteomyelitis, cellulitis, and rheumatoid arthritis especially in the early stages of the disease. In the early stages of the diseases, conservative treatment such as offloading of the involved foot, treating the bone disease, and preventing further fractures and/or dislocation could be considered [3]. Laboratory examination also plays a critical role as the outcome indicator, with HbA1c as one of the values that should be optimized, Kavarthapu reported that an HbA1c level of 8 or less is needed before elective deformity corrections [6].

The surgical treatment for CN foot is not only based on the location or severity of deformity, pain or instability, and presence of infection, but also the patient's comorbid, and compliance [3]. There are various surgical techniques for the realignment and stabilization of the deformed CN foot. Achilles tendon lengthening, plantar osteotomy, osseous debridement, realignment osteotomy, selective or extended arthrodesis, and open reduction with various forms of internal fixation with or without external fixation are the well-known techniques.

In this case reports, both of our cases are treated with arthrodesis and some form of internal fixation. Both of the techniques chosen was based on the clinical finding of the patients. In the first case, multiple bone grafts were used, due to the clinical feature of the deformities. The deformity does not appear to be infected, but it has severe instability and massive bone destruction. After debridement and removal of the destroyed bone, the surgical team harvested two bone grafts to fill the defect left by the removal of the destroyed bone. For the final part of the procedure, two locking plates were used, one each for the anterior and the lateral side of the ankle joint with two locking screws added to provide further rigidity of the fixation. This technique was comparable to Kavartaphu and Vris who performed it first and gives a good result [6]. The patients had no complications after surgery according to Clavien-Dindo Grade 1 [7].

Contradictory to the first case, the second case has an infection affecting the CN deformity. The technique used was similar but different. The similarity was that in both cases the deformity was severe and arthrodesis with internal fixation was performed. However, in the second case, the surgical team only used minimal implants. Arthrodesis has been considered by many to be the choice of treatment for severe and late-stage CN foot [8]. It is still controversial whether internal or external fixation as the most suitable method. The argument is that internal fixation reduces the risk of infection, which is a major problem in CN foot caused by the diabetic condition [9,10]. For the second case the team choose a single distal tibia locking plate for the arthrodesis of the tibiocalcaneal joint planted in the anterior part of the foot. Although intramedullary nailing was the most researched method of internal fixation, the use of distal tibia locking plate has a comparable outcome to intramedullary nailing and is a workhorse for distal tibial fractures [11–16]. Similar to the first case, fibular strut grafts were also used to help with bone loss [17–19]. Previous studies have reported excellent outcomes from the use of fibular strut graft in the cases of CN foot arthrodesis. Compare to a previous study from Jeong, the purpose of internal fixation is to lessen the chance of infection [4]. The fibular graft consists of vascularized and nonvascularized ones. Even when it is nonvascularized, fibula autograft still has biological advantages and can accelerate bone union. Its use can minimize problems related to the implantation of hardware, therefore resulting in a lower risk of post-operative infection. This patient also had no complications after surgery based on Clavien-Dindo Grade 1 [7].

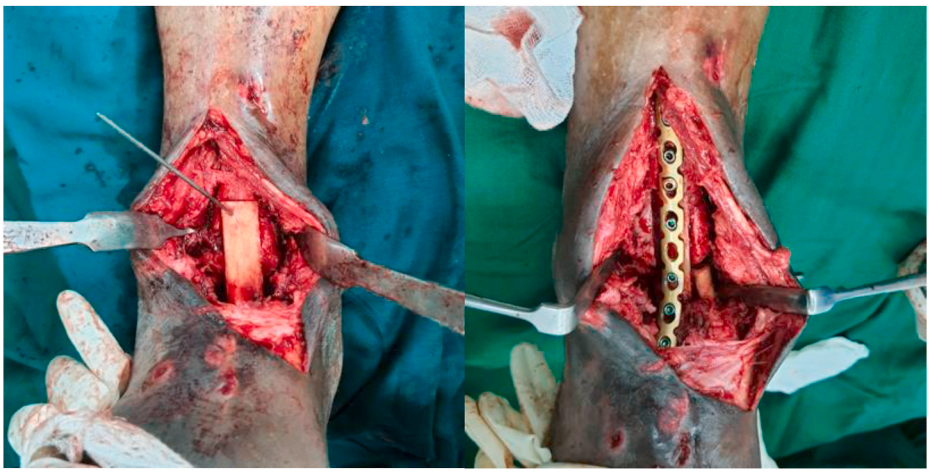


Fig. 11. The surgical approach was through the anterior part, exposing the distal part of the tibia. A fibular strut graft was placed at the anterior part and fixed with a distal tibia locking plate.

Fig. 12. Postoperative right ankle radiograph of anteroposterior and lateral view showing the bones and the plate in good alignment.

Through this paper, we intend to give a new perspective on how to treat charcot neuroarthropathy deformity. From the first case, we know that using multiple bone grafts may be another option to treat severe instability in CN foot deformity. Another positive outcome is seen in the second case, where the use of minimal implant in the form of a distal tibia locking screw proves to be successful in achieving arthrodesis. This suggest there is another method which gives promising result in achieving ankle arthrodesis other than intramedullary nailing.

6. Conclusion

In the case of severe CN foot, multiple modalities of treatment may be used considering the clinical condition of the deformities. A rigid fixation using an internal fixator may be considered to lessen the infection rate while providing a rigid enough fixation, and minimal implant using a single distal tibial locking plate can also be considered. To provide with the osseointegration, bone grafts such as the fibular strut graft and cancellous graft from the iliac should be considered. In those two cases the arthrodesis seemed to yield promising results radiographically and functionally, but long-term outcomes still need to be confirmed to thoroughly prove the efficacy of both procedures.

Ethical approval

The work has been approved by the appropriate ethical committees related institution in which this procedure was performed.

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N/A.

Fig. 13. The postoperative clinical picture of the right ankle.

Fig. 14. The clinical picture of the right ankle (3 months after the procedure).

Author contribution

1. Subawa W: Study concept and design, performed surgery.
2. Nugraha HK: Data collection, Writing the paper.
3. Afandi R: Writing the paper, final editing and publication.
4. Rusdianto IA: Data collection, data analysis and editing.

Registration of research studies

1. Name of the registry:
2. Unique Identifying number or registration ID:
3. Hyperlink to your specific registration (must be publicly accessible and will be checked):

Research registration

This is not a 'first in humans' report, so it is not in need of registration.

Guarantor

Subawa W.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Declaration of competing interest

All authors declare that no conflict of interest in formulating this article.

Fig. 15. Right ankle radiograph of anteroposterior and lateral view 3 months after procedure.

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The authors declare that they have no conflict of interest in this study.

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