Case Report
Knee arthrodesis with autologous patella graft for treatment of Charcot knee with severe bone loss: a case report

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Abstract: Background: Charcot arthropathy refers to a progressive destructive joint disease associated with nerve damage and periarthritis insensitivity. Diabetes, tabes dorsalis, syringomyelia are the main causes. Case report: Herein we present the case of a 60 year old male with Charcot knee complicated by tabes dorsalis, in which posterior tibial bone loss, joint instability, bone fragility and susceptibility to infection were intractable problems. Knee arthrodesis with double locking compression plates were performed using autologous patella graft to fill in the bone defect and structurally reconstruct the tibial plateau, providing increased surface area for bony union. Findings obtained 10 months postoperatively showed maintenance of solid fusion with no complications. Conclusion: The technique described here appears to be safe and reproducible based on the current results, providing significant functional improvements in the patient with Charcot arthropathy. This case indicates that the technique could be an option in the operative management of Charcot knee with severe bone loss.

Keywords: Charcot arthropathy, tabes dorsalis, knee arthrodesis, bone loss, autologous patella graft

Introduction
Charcot arthropathy was first described 140 years ago by J.M. Charcot but it was not until 1936 that W.R. Jordan established the association between Charcot arthropathy and diabetes mellitus, which is the leading cause of the condition worldwide today [1]. Although most commonly observed in the foot and ankle, there is a predilection for other sites including the knee, hip, shoulder and spine, depending on the underlying etiology [2]. There is a lack of specificity in the manifestations of Charcot arthropathy that distinguish it from other similar diseases. Besides joint swelling and deformity, lack of pain and sensory loss can be prominent symptoms. The absence of appropriate nociception leads to excessive overload bearing or repetitive articular cartilage damage and bone defects, eventually leading to fracture or joint dislocation. Therefore, Charcot arthropathy should be taken into consideration as soon as possible when eliminating the possibility of other diseases.

Tabes dorsalis is a form of tertiary parenchymal neurosyphilis that arises in 2% to 9% of individuals with untreated syphilis and has an incubation period of 3 to 50 years post-infection [3]. The lesion principally affects the posterior funiculus of the spinal cord, with cranial nerves also vulnerable. Clinical manifestations of tabes dorsalis may take decades to appear after the initial infection. Following more organized public health measures and the introduction of penicillin in the 1950s, the incidence of neurosyphilis has declined significantly and as a result, tabes dorsalis has become a rare disease [4]. The incidence of Charcot arthropathy in patients suffering from tabes dorsalis ranges from 6% to 10%, which is an unneglectable number [5].

Generally, total knee arthroplasty (TKA) is not recommended as a therapy for Charcot arthropathy due to the high risk of internal fixation failure. Over the last few years, these traditional views have been questioned, with an increasing number of reports of Charcot knee arthropathy.
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Case presentation

A 60-year old male was presented to our hospital with a swollen left knee and restricted movement since an initial sprain he had experienced while walking down the street 7 days prior. He was unable to stand or walk, but did not feel any pain. His condition gradually deteriorated until he was finally admitted to our hospital. In the last 4 years, he had consulted numerous clinics with a long history of backaches and flash pain in his lower limbs. However, the treatments appeared ineffective and there had been no clear diagnosis. The patient had received a bipolar femoral head replacement (BFHR) in another hospital 2 years previously due to a right femoral neck fracture. The affected side dislocated on 3 occasions (Figure 1A-C) following the operation due to an unknown cause, with correction achieved after one manual traction reduction and one open reduction procedure successively. By the time the third dislocation happened, total hip replacement (THR) was ultimately performed 6 months after BFHR in another medical institution (Figure 1D). Unexpectedly, right hip luxation occurred again only 2 months after THR and was cured by manual traction reduction. The prosthesis remained in position until this present injury. The patient confirmed that he did not have a history of diabetes, blood transfusions, or genetic diseases.

In terms of physical findings, the passive and active range of motion of the left knee was -20°/130° in flexion without tenderness at the joint spaces. The manual varus and valgus stress test demonstrated apparent instability while the anterior and posterior drawer test showed positive results. On neurological findings, a clear loss in superficial sensation and bathyesthesia below the level of Th10 were observed in addition to the loss of bilateral tendon reflexes of the lower limbs. Muscular strength of the bilateral limbs was grade V with normal muscular tension and no induction of pathological signs.

Table 1. Significant laboratory reports

<table>
<thead>
<tr>
<th>Test</th>
<th>Result</th>
<th>Reference value</th>
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<tbody>
<tr>
<td>WBC</td>
<td>8390</td>
<td>3500-9500/μl</td>
</tr>
<tr>
<td>Hb</td>
<td>111</td>
<td>130-175 g/L</td>
</tr>
<tr>
<td>ESR</td>
<td>62</td>
<td>0-15 mm/h</td>
</tr>
<tr>
<td>CRP</td>
<td>46.1</td>
<td>0-8 mg/L</td>
</tr>
<tr>
<td>TPPA</td>
<td>++</td>
<td></td>
</tr>
<tr>
<td>RPR</td>
<td>++</td>
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</tr>
</tbody>
</table>

Note: WBC, White blood cell; Hb, Hemoglobin; ESR, Erythrocyte sedimentation rate; CRP, C-reactive protein; TPPA, Treponema pallidum antibodies; RPR, Rapid plasma reagin. Serological tests showed significant positive results for RPR and TPPA besides mild anemia and inflammation.
Laboratory data indicated an appreciable rise in erythrocyte sedimentation rate (ESR) and level of C-reactive protein (CRP) besides mild anemia (Table 1). Both treponema pallidum antibody (TPPA) and rapid plasma regain (RPR) results were positive (+++) on a serological test, providing significant evidence of syphilis infection.

Plain radiography revealed anterior dislocation of the left knee in addition to fracture of the left tibial plateau and capitula fibulae (Figure 2). Computed tomography (CT) demonstrated a large bone defect of the posterior tibial plateau and massive bone fragments around the knee joint were also visible (Figure 3). It is difficult to precisely classify the fracture based on current theories. Magnetic resonance imaging (MRI) showed the presence of a bucket-handle meniscus tear, posterior cruciate ligament (PCL) injury with severe synovitis and arthroedema (Figure 4). An MRI of the whole spine displayed mild intervertebral disc bulge at C5-C7 and L1-L3, but no evidence of syringomyelia, meningomyelocele or related diseases (Figure 5). An emission computed tomography (ECT) scan performed at another hospital 2.5 years previously showed a concentration of radionuclide and abnormal bone metabolism in the right hip and left knee, but the results did not receive much attention at the time (Figure 6).

According to these findings, his condition was diagnosed as Charcot arthropathy caused by tabes dorsalis, with the right hip and left knee being affected. The patient received
anti-syphilis therapy with 4.8 million units of penicillin intravenously every 12 hours for 14 days on the basis of established etiology prior to being accepted for surgery. Sensory recovery of the bilateral lower limbs occurred two weeks after treatment. A retest of the patient’s RPR was weakly positive (+), which indirectly proved that this therapy was effective.

The procedure was performed with the patient supine with a tourniquet around his thigh inflated to 240 mmHg, skin prepared with entoiodine and antibiotic prophylaxis was used intravenously. A mid-longitudinal incision bypassing the patella was created extending distally to the tibial tubercle (Figure 7A). Dissection of the knee joint cavity revealed features consistent with pathological fracture, including mass blood clots around the joint and multiple brittle bony fragments without vascularity. Large bone defects on the posterior aspect of the tibial plateau were observed (Figure 7B). To prepare the femur and tibia for arthrodesis, osteotomy was performed so that the knee was aligned with 10° of flexion, 5° of valgus and 5° of extension angulation, in which position intraoperative X-ray imaging proved that lower limb alignment was satisfactory. An osteotomy mold such as is employed for TKA, was used to ensure the accuracy of osteotomy. The resected patella was selected as an autologous bone graft to rebuild the structure of the tibial plateau. After removing the cartilaginous surface of the patella, it was split into two equal parts along the mid-longitudinal line with an oscillating saw followed by meticulous clipping in order to fit into the bone defect (Figure 7C). After adjusting to an appropriate shape and size, they were reassembled and immobilized on the dorsal surface of the proximal tibia with two 4.5 mm cannulated screws (Figure 7D).

The reconstructed tibial plateau demonstrated sufficient surface contact with the femoral side. Finally, a bend-forming 13-hole limited contact-locking compression plate (LC-LCP) was applied medially and secured with locking screws in addition to conventional bicortical screws. An additional 14-hole locking compression plate (LCP) was fastened anteriorly and...
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Figure 7E. Figure 6. Emission computed tomography performed 2.5 years previously showed nuclide concentration of right hip and left knee.

secured with bicortical screws (Figure 7E). It was necessary to curve the plates, so that they would conform to the shape of the tibia and femur. Postoperative limb length measurement showed that the left leg was 1.5 cm shorter than the contralateral side, which was within an acceptable range. An above-the-knee plaster support was also fitted for 2 months postoperatively before weight-bearing training.

Two months after surgery there was radiological evidence of callus formation so the patient commenced partial weight-bearing with crutch ambulation (Figure 8A). Five months postoperatively radiographs confirmed gross knee fusion and he was allowed to fully weight bear. The final outcome obtained 10 months after surgery showed excellent consolidation without deformity and with well-maintained lower limb alignment (Figure 8B). His gait was basically steady without crutches with no complaints of pain and with no collection of joint fluid (Figure 8C).

Informed consent was obtained from the patient for publication of this manuscript using his information and images.

Discussion

The surgical therapeutic management for Charcot knee arthropathy remains controversial. Traditionally, TKA was discouraged as a management strategy of Charcot arthropathy due to the risk of secondary osteoporosis and a high incidence rate of prosthetic loosening. Recently there have been several reports of TKA being used in Charcot knee arthropathy, yet the curative effect is still ambiguous, especially the long-term prognosis. A review of the available literature revealed that the total complication rate is approximately 50% (35 complications on 72 knees), including dislocation (7%), periprosthetic fractures (7%), deep venous thrombosis (DVT, 8%), disruption of the quadriceps (4%), deep infection (3%) and other complications [8]. Kim et al. reported that 19 knees diagnosed with Charcot arthropathy accepted TKA with 47% of the total cases resulting in various complications, and only 53% of the patients with severe Charcot arthropathy showing satisfactory results at a 5-year follow-up [9]. Arthrodesis is commonly the recommended choice of surgical treatment for Charcot arthropathy, especially in cases with severe instability, soft-tissue laxity and bony destruction [10].

It is recognized that knee fusion is not easily achieved in Charcot arthropathy because of poor bone quality and underlying fundamental diseases. Furthermore, for cases with large bone defects, the situation appears more intractable. To our knowledge, although there were reports of using fibulae or orthobiologics for bone grafts, no previous reports of the surgical treatment of Charcot knee arthropathy with patella grafts have been published. Achieving long-term stability in a patient with Charcot arthropathy was considered extremely important, especially for cases with severe bone loss. It was necessary that a difficult secondary revision procedure was avoided, considering the habitual dislocation of the right Charcot hip and the underlying possibility of developing Charcot arthropathy in the contralateral hip or knee, highlighting the importance of maintaining limb length and bone stock. This case was characterized by a large bone deficiency at the posterior of the tibial plateau, which decreased the contact area of the proximal tibia and distal femur, further reducing the probability of fusion. Given the difficulty of this
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Case, it was important to secure a bone graft of sufficient volume to provide good mechanical stability. The technique described above overcame the challenges by creating a larger surface area for bony union and rigid stability with dual locking compression plates.

Vascularized fibular grafts have been the most commonly-used autogenous bone for reconstruction of skeletal defects in knee arthrodesis. Several reports have also suggested utilizing of other orthobiologics. Voss introduced the concept of artificial arthrodesis by use of a cement spacer as an alternative in treating cases with severe bone loss [11]. Capanna et al. used modular nails with metal, polyethylene or cement with gentamycin spacers around the central coupler with gentamycin for patients with bony defects [12]. Both methods proved feasible. In this case, the patella was used innovatively as an autologous bone graft to rebuild the tibial plateau. This technique has several theoretical advantages. Above all, this method clearly created conditions that were more physiological inside the main pathologic lesion compared with other orthobiologics. Autogenous bone is regarded as the reference standard for bone graft material, because it provides the specific elements required to generate and maintain bone, including scaffolding for osteoconduction, growth factors for osteoinduction and progenitor cells for osteogenesis [13]. Secondly, the choice of the patella as a source of the autogenous bone resulted in a better fit of the graft and host bone than occurs with a fibula or other orthobiologic because of the geometry of the bone surface. In addition, using the patella as a bone graft avoided creating an additional incision on the shank, certainly decreasing the risk of postoperative infection and shortening recovery time compared to harvesting the fibula. Although no published data

Figure 7. A. A Mid-longitudinal incision bypassing the patella of left knee; B. Large bone defects of posterior tibial plateau were observed during the operation; C. The intact patella was sawed into two half of pieces; D. The tibial plateau was structurally reconstructed with reassembled patella graft and immobilized with two cannulated screws; E. Two bend-forming locking compression plates were placed anteriorly and medially of the knee joint.
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Table 2. Pre-operation and post-operation SF-36 score

<table>
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<tr>
<th>SF-36</th>
<th>PF</th>
<th>RP</th>
<th>BP</th>
<th>GH</th>
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<th>SF</th>
<th>RE</th>
<th>MH</th>
<th>PCS</th>
<th>MCS</th>
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<tr>
<td>Pre-op</td>
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<td>0</td>
<td>40</td>
<td>30</td>
<td>70</td>
<td>33</td>
<td>67</td>
<td>60</td>
<td>90</td>
<td>230</td>
</tr>
<tr>
<td>Post-op</td>
<td>40</td>
<td>50</td>
<td>74</td>
<td>47</td>
<td>75</td>
<td>89</td>
<td>100</td>
<td>64</td>
<td>211</td>
<td>328</td>
</tr>
</tbody>
</table>

Note: PF, Physical functioning; RP, Role-physical; BP, Bodily pain; GH, General health; VT, Vitality; SF, Social functioning; RE, Role-emotional; MH, Mental health; PCS, Physical component summary; MCS, Mental component summary. The patient’s SF-36 score increased markedly compared to preoperative status, which indicating his quality of life had improved a lot both physically and mentally after receiving the surgery.

Figure 8. A. Plain X-ray image obtained 2 months postoperatively shows initiation of union process; B. Anteroposterior and lateral radiography of left knee 10 months after knee arthrodesis exhibited plenty formation of osteotylus and favourable fusion condition; C. Steady standing posture 10 months after surgery without any upholser.

has compared the clinical results of a patella graft technique and other orthobiologics for Charcot knee fusion, our data suggest that patella autograft may accelerate bone union, minimize hardware-related problems, and result in a lower risk of infection.

The prerequisites for successful fusion include full contact of cancellous bone surfaces, satisfactory limb alignment and rigid fixation. The ideal limb alignment for knee fusion should be in the "neutral range": 5° to 7° of valgus and 10° to 20° of flexion [14]. Either excessive or insufficient angulation may increase the limb-length discrepancy, instability of gait and finally leading to the failure of fusion. The fixation methods in knee arthrodesis include external fixator, intramedullary nail and plate fixation. Although intramedullary nailing produces the highest rate of fusion of the three methods above, considering the loss of bone, inferior bone stock and mild anemia in this present case, which would contraindicate of external fixation and intramedullary nailing, the double-plating fixation method was chosen. Kuo et al. reported using a dual-LCP technique to achieve fusion in 3 patients with a 100% success rate [15]. This technique has the advantage of providing axial compression of the fusion construct and fixed angle stability with the locking screws, which cannot be achieved through intramedullary nailing. The use of an above-the-knee cast for an average of 18 weeks (ranging from 12 to 53 weeks) postoperatively prior to weight bearing is also recommended in order to restrict body activity and promote fusion [16].

At a follow-up visit 10 months postoperatively, the patient showed no complications around the wound and neither instability nor pain were presented. He could walk on his own for 500 meters without a cane or walker. His SF-36 score had greatly improved compared to before the surgery (Table 2). A recent plain radiograph showed that a large quantity of osteotylus had formed and the joint was basically fused with no sign of internal fixation loosening or breaking. In this case, the patient’s gait was steady and the quality of his daily life had significantly improved compared with preoperatively. Although there is currently no apparent complication, long-term follow-up is necessary and important.
In conclusion, a satisfactory outcome was obtained without infection or nonunion in performing a successful knee arthrodesis for a patient suffering from Charcot arthropathy, with the use of a free, reassembled patella graft. Based on our understanding of the literature and our experience with this case, application of patella graft should be considered in similar cases with severe bone defect when performing knee arthrodesis.

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Disclosure of conflict of interest

None.

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References