The treatment of tibial defects following chronic pyogenic haematogenous osteomyelitis in children

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Abstract
The aim of this paper was to review the results of treatment of 31 children, aged 3 to 12 years, with tibial defects resulting from haematogenous osteomyelitis seen between 1989 and 2006. Common features were skin defects, discharging sinuses, pathological fractures, sequestra and pseudarthroses, with a duration of 4 to 12 months prior to reconstruction. The defects ranged from 2 to 20 cm, 18 were in the proximal third of the tibia, 10 in the middle third, and three in the distal third. Surgical treatment consisted of repeated debridement, sequestrectomy, gentamycin beads and soft tissue cover for skin defects, followed by reconstruction at a later stage. Bone grafting was done by using cancellous chips in cavitating defects (Papineau technique) (n = 2), onlay grafting in defects < 2 cm (n = 5), corticocancellous square segments threaded and fixed over a Kirschner wire in the defect (n = 15), and fibular transference to the proximal tibia in large defects with poor skin and fibrosis (n = 9). All grafts healed well at 1 to 17 years follow-up. Complications of shortening (1 to 20 cm), equinus (1), ankle varus (3) and recurvatum (1) were related to the infective process. All patients are ambulant.

Introduction
Acute haematogenous pyogenic osteomyelitis is still a major problem in developing countries despite the advent of antibiotics and surgical decompression. The tibia is the commonest site of involvement. Most children present late with severe disease, requiring aggressive surgical intervention and prolonged hospitalisation. The chronic form of the disease may lead to extensive necrosis of bone, pathological fractures and formation of sequestra, with resulting segmental defects, cavities, discharging sinuses and pseudarthroses. Injudiciously placed incisions over the subcutaneous surface of the tibia lead to further problems in treatment due to exposure of bone, destruction of the periosteal tube with defective formation of involucrum and superinfection with a multiplicity of organisms. In such conditions the overlying skin is unhealthy, with adherent scarring, discharging sinuses and persistent infection. Scar tissue and avascular bone act as an impenetrable barrier to antibiotics. For healing to be successful sequestra, scar tissue and sinuses must be removed, and dead space eliminated. The reconstruction of bone defects of the tibia resulting from chronic osteomyelitis is therefore challenging. It is further compounded by disease atrophy, growth plate closure, fibrous ankylosis of the joints, angular deformity, and subluxation of the tibiofibular joints.
Local cancellous bone grafting options may result in flare-up of the infection and repeated fractures and non-union. Some children have undergone amputation\textsuperscript{2,6} for chronic osteomyelitis of the tibia. Such defects are best managed by alternative grafting methods such as ipsilateral fibular transfer or bypass grafting to avoid the site of infection, pedicle grafts, free vascularised grafts or bone transport. In children there have been few reports of reconstruction of bone defects following chronic osteomyelitis of the tibia.\textsuperscript{2,3,5,6}

The aim of this paper is to present the results of the methods used to bone graft the tibia following bone defects resulting from chronic pyogenic osteomyelitis.

Patients and methods

The case records and radiographs of 31 children with defects of the tibia following primary haematogenous osteomyelitis were reviewed retrospectively. There were 21 boys and 10 girls with an average age of 7.5 years (3-12 years) seen between 1989 and 2006. The majority (22) were referred following treatment at a peripheral hospital.

The reconstruction of bone defects of the tibia resulting from chronic osteomyelitis is challenging.
Staphylococcus aureus was the causative organism cultured in all cases in the acute phase. The duration of the chronic phase was between 4 to 12 months before reconstructive surgery was undertaken. About one-third had an incision over the anteromedial surface of the tibial shaft for drainage of pus, exposing bone (Figures 1a and 2a).

Clinically, the common features were discharging sinuses with pus, exposed bone (n = 15), skin defect over subcutaneous border of tibia (n = 7), sequestra and pathological fractures (n = 24), and established pseudarthroses (n = 7).

The defects ranged in size from 2 cm to 20 cm (av. 7 cm). They were located in the proximal one-third in 18 patients, middle third (n = 10), and distal third (n = 3). The haemoglobin at the time of surgery ranged between 8.8 gm% and 11.2 gm%, white cell count of 6.8-10.9 g/L, ESR 25-78 mm/Hr, and the albumen was low in 10 children. Other sites of bone involvement occurred in three children. All children had completed an initial course of cloxacillin for 6 weeks. There was no evidence of retroviral disease in this patient group.

Surgical technique
The reconstruction was done in stages. All patients had two to three repeated debridements in theatre under general anaesthesia. Sequestrectomy, curettage of bone ends, cavities, and infected granulation tissue was performed till bleeding bone was encountered. Fibrous tissue at the site of the defect was excised. Gentamycin beads were inserted in all infected defects for 2 weeks. Four children had gastrocnemius or soleus flaps to cover skin defects by a plastic surgeon. Three children had split skin grafts. The decision to bone graft was made intra-operatively at the time of removal of the gentamycin beads.

Bone reconstruction was of four types. The Papineau technique, placing layers of cancellous chips in exposed cavities followed by skin grafting, was performed in two children. Onlay grafting using corticocancellous chips was used in five children with defects < 2 cm. Segmented iliac crest bicortical grafts (± 1.5 × 1.5 cm ) threaded over a Kirschner wire (1.8-2 mm) were fixed in defects between 2-8 cm augmented with cancellous chips. In large defects > 8 cm or in a bed with scarring and poor adherent overlying skin, or established pseudarthrosis with tapered ends, proximal fibula transference to the lateral aspect of the tibia was performed in nine children. The distal fibula was synostosed to the tibial remnant 3-4 months later.

Technique of fibula transfer
An anterolateral incision was made to expose the head and neck of the fibula, tibialis anterior and proximal tibia. The peroneal nerve was identified and isolated, and the fibula was divided below its neck. After decorticating the lateral surface of the upper tibia, the fibula was transferred deep to the tibialis anterior, and fixed to the lateral surface of the tibia with screws or K-wires. Cancellous chips were placed around the transfer. The limb was immobilised in a plaster cast.

Postoperative treatment
Antibiotics were used for 2 to 3 weeks in all patients. The plaster casts were changed at 4 to 6 weekly intervals until consolidation was seen. All children with defects > 2 cm were immobilised in above-knee callipers until solid union occurred.

In six children with proximal fibula transfer, a distal tibiofibular synostosis with iliac crest bone was done through an anterolateral incision after 3 to 4 months. In three remaining children, the parents refused further surgery and chose to use BK callipers.

Further procedures
Three children with segmented bicortical grafts required repeat grafting with cancellous chips due to resorption of the graft.

Results
Follow-up ranged from 1 year to 17 years (av. 6 years). Healing occurred in all cases by 3.5 to 6 months after the last procedure. Three patients had flare-up of the osteomyelitis, but settled with intravenous antibiotics for two weeks. Shortening ranged from 1 to 20 cm (av. 4 cm). A varus deformity of the tibia occurred in six children (20º-30º). One patient had a severe equinus deformity (30º). Three children had ankle varus (10º-15º). One child had a recurvatum deformity of the proximal tibia. All patients with shortening and foot deformities had evidence of growth plate damage of the tibia on radiographs taken prior to surgery. The tibia showed widening of the shaft in all patients, remodelling of the varus deformity occurred in three children, and hypertrophy of the fibula transfer occurred in all nine patients. Three children had distal fibular epiphysodeis for ankle varus and three children had upper tibial osteotomy to correct progressive varus of the tibia.

Epiphysodeesis of the contralateral limb was done in two children to control limb length deficiency. All children are ambulant, three with callipers. All shortening was treated with a shoe raise. The patient with 20 cm shortening developed a severe equinus deformity, which compensated for the leg length deficiency, and wears a moulded surgical boot. The deformities that occurred were the result of damage to the growth plates of the tibia. The methods of treatment described cannot correct all deformities but resulted in healing of the osteomyelitic defects.

Discussion
In developing countries there is a high incidence of osseous defects and pseudarthrosis following primary haematogenous osteomyelitis in children.
The management of such defects of the tibial diaphysis is challenging. The basic principles in the treatment of any long bone infection is based on radical debridement of all compromised tissue, early provision of vascularised soft tissue cover with elimination of dead space followed by delayed autogenous grafting.\(^2\)\(^,\)\(^5\) Gristina \textit{et al}\(^8\) have shown that non-viable bone stimulates adherence and colonisation by pathogenic bacteria. This leads to further ischaemia and multiplication of bacteria and chronicity.

The success of surgical treatment depends on the removal of ischaemic tissue and breaking the cycle of bone death, sequestrum formation and spread of infection. When the infection is severe and of long duration, the longitudinal continuation of the periosteal tube may be interrupted, leading to defective formation of the involucrum.\(^5\)

\begin{figure}
\centering
\includegraphics{figure2a.png}
\caption{Image shows osteomyelitis of the shaft of the entire tibia complicated by a surgical incision over the medial subcutaneous surface of the tibia.}
\end{figure}

\begin{figure}
\centering
\includegraphics{figure2b.png}
\caption{Radiograph shows large sequestrum involving the tibial shaft.}
\end{figure}

\begin{figure}
\centering
\includegraphics{figure2c.png}
\caption{Radiograph shows large defect of the tibia following sequestrectomy.}
\end{figure}

\begin{figure}
\centering
\includegraphics{figure2d.png}
\caption{Radiograph shows fibular transference to the proximal tibia fixed with a Kirschner wire, healing of the tibial shaft; distal tibiofibular synostosis was not performed.}
\end{figure}

Success depends on the removal of ischaemic tissue and breaking the cycle of bone death, sequestrum formation and spread of infection.
Daoud and Nade concluded that the status of the periosteum was of primary importance in predicting the subsequent evolution of the disease. Incisions over the medial tibial surface further compromise the periosteum and result in a scarred and avascular graft bed which is not receptive to conventional grafting techniques. The timing of sequestrectomy has been debated. Bosworth suggested radical diaphysectomy. Daoud suggested aggressive osseous debridement. Fowles allowed maximum development of involucrum and revascularisation of the sequestrum by delaying sequestrectomy.

A variety of techniques has been developed to reconstruct the tibia, including onlay, and inlay cancellous grafts, strut bone grafting, tibiofibular synostosis, fibular bypass grafts, segmental bone transfer and vascularised fibular transfer. The size and extent of the defect and the experience of the surgical team dictates the method used. Whatever the method used, it is preceded by staged debridement, curettage and sequestrectomy, obtaining a granulating bed without any signs of infection, followed by the second stage 4-6 weeks later, of reconstruction of the tibial defect.

Cancellous grafts were recommended by Papineau et al for defects of bone with difficult skin closure mainly in exposed bone and cavities. Skin coverage was obtained by split skin grafts or epithelialisation. The technique is time-consuming, requires prolonged hospitalisation and has a considerable associated morbidity. A 5% amputation rate has been reported. Recurrence and scarring has been reported. In this series, this method was used in two patients and skin graft was performed to obtain cover of the bone surface.

De Oliveira treated bone defects of the diaphysis with autogenous corticocancellous grafts. Healing was difficult, sclerosis was usually present and the problems were recurrence and infection.

Daoud, Agiza and Griffiths showed that cortical grafts were slow to incorporate. Daoud found poor incorporation with free fibula strut grafts. Problems were recurrent infection, displacement, non-union, graft fracture, peroneal nerve palsy. Boyd and Dawson found a high rate of graft fracture and emphasised the morbidity of surgery on the normal contralateral leg and suggested that this method not be used for large defects. In this series, onlay corticocancellous grafts were successful in small defects less than 2 cm. Good incorporation and remodelling of the tibia occurred.

In defects 2-8 cm, inlay grafting method using corticocancellous segments of iliac crest threaded over a Kirschner wire (1.8 mm-2 mm) gave good results. The success of the method depends on a good vascular bed with some periosteal continuity, usually posterolateral. The Kirschner wires provide alignment whilst the graft is incorporating. The graft showed some resorption, while the bicortical segments threaded over the wire provided some stability and increased the surface area to allow revascularisation to occur. This method has not been described in the literature, but used by Versveld et al.

The literature abounds with reports of cases where the transference of the fibula was done with successful results. This method was employed when the tibial defects were large (> 8 cm), the soft tissue and skin around the defect showed scarring, and there was lack of continuity of the periosteal tube, or an extensive pseudarthrosis. These methods were used in adults mainly following septic non-union for tibial fractures. The method depends on an intact blood supply via the soft tissue attachments to the fibula. Several authors proposed the creation of a tibiofibular synostosis either by implanting the fibula into the proximal tibial remnant, or into both the proximal and distal remnants.
or by the creation of a tibiofibular synostosis between
the fibula and the remaining tibial segments, with or
without cancellous grafting. Various modifications of
these basic three techniques have emerged.

Transference or transplantation of the fibula in one- or
two-stage procedure or tibiofibular grafting leaving the
fibula intact as a strut has been used widely in tibial
defects following trauma. Hahn18 transferred only the
upper end into the proximal tibia. Huntington19 advised
transfer of the distal fibula also, using a two-stage proce-
dure. The transferred fibula ensures continued vasculari-
sation of the graft in a poorly vascularised fibrotic bed.
Several authors have used this method of reconstruc-
tion.1,2,3,5,20

Tibiofibular synostosis, fixing the intact fibula to the tib-
ial remnants with interposed bone graft was performed in
various methods by several authors, also in adults.21-28
McMaster29 devised a salvage procedure for the tibiofibu-
lar synostosis in adults with multiple cross peg grafts
above and below the defect using cortical struts.

The experience with fibular transference in children was
reported by a few authors only.1,2,3,5,6 Agiza3 and Zahiri6 had
good results with the Huntington transfer. Daoud5 experi-
enced complications including non-union with the proxi-
mal fibula transfer, fractures of the fibula graft and prox-
imal migration of the lateral malleolus.

We used the fibular transfer in nine patients (Figures 2a,
b, c, d, e and Figures 3a, b). The method allowed bypass-
ing of the scarred bed and skin. The fibula was synostosed
to the proximal and distal tibia in separate stages in six
patients. (Figures 3a, b). In three remaining patients the
fibula was synostosed to the proximal tibial remnant only
(Figures 2a, b, c, d, e). The parents refused further sur-
gery and preferred to use a calliper. All patients showed
hypertrophy of the fibular transfer (Figure 3b). The tibial
remnant showed increase in size. Similar increase in tib-
ial size and healing of the pseudarthrosis was recorded by
Zahiri6 and Wilson.20

Following the studies of the blood supply of the fibu-
la, Chacha et al30 transferred a large graft of fibula
raised on a pedicle of peroneal and anterior tibial mus-
cles and peroneal vessels, and fixed the graft to the tibia
along its posterior long axis proximally and distally,
producing a sound mechanical and biological basis for
union. Similar procedures were done by Shapiro et al,31
Hertel et al,32 Khan et al33 and Chung34 in adults.
Coleman et al35 used the technique in children with
pseudarthrosis of the tibia. The method avoids neu-
rovascular anastomosis, donor site morbidity associated
with contralateral fibular transfer and does not require
the presence of an intact fibula. These methods require
extensive dissection of the graft and blood supply.
Previous local inflammation increases the risk of vas-
cular damage. Complications include pain, chronic
oedema, valgus ankle and refracture.32,34 Rotation of
island flaps can compromise the circulation.32,34
Free vascularised fibular grafts used in developed countries, to hasten healing and reduce the risk of graft fracture in defects of the tibia, have been used mainly in adults. The contralateral or rarely ipsilateral fibula is harvested with the nutrient vessels and transferred to the defect, and its vascularity established by microvascular anastomosis. The technique was first described by Taylor et al. The method is time-consuming, requires a highly skilled team and there is a risk of failure and morbidity at the donor site. Minami reported graft fracture, non-union, peroneal nerve palsy, equinus valgus and flexor hallucis tightness. Thrombosis of the repair occurred in 14 cases (13.5%). Hsu reported amputation, infection and non-union. Welland used the method to obtain union in osteomyelitis of the tibia in adults mainly. However, in haematogenous osteomyelitis the disease may run a relentless course and patients have to be carefully selected. The resources and skills are not readily available in developing countries and technical problems include purchase of graft fixation in diseased bone.

Ilizarov distraction osteogenesis is well described and has good reported outcomes in treating osseous defects, deformity and limb length in adults. A paucity of literature exists supporting the treatment of children with this method for bone defects secondary to osteomyelitis. Segmental bone transport using circular external fixators has been described to treat tibial defects. Atkins translated a fibular segment medially with olive wires into the defect in the tibia and then compressed the proximal and distal ends of the tibia onto the transported segments.

Corticotomy with bone transport and lengthening has been successful in smaller defects. These methods also require expertise and resources and have their limitations in extensive pyogenic osteomyelitis of the tibia where purchase of the wires may be a problem. The complications of the method include pin tract infection, persistent oedema, malunion, stress fractures, foot deformities, compartment syndrome, peroneal nerve injury, pain and psychological problems.

Conclusion
Cavity defects were treated successfully with the Papineau method of cancellous grafting, defects < 2 cm by cortico-cancellous onlay grafts, and in 2-8 cm defects, cortico-cancellous iliac segments threaded over a Kirschner wire as an inlay graft. Large defects or a poor soft tissue bed with fibrosis and scarring was bypassed by doing a fibular transfer. No serious morbidity occurred from the transfer. Complications including shortening, angular deformities and foot deformities were the consequences of the diffuse disease process and growth plate damage. Early sequestrectomy, only once the sequestrum was demarcated on radiographs, together with debridement, curettage and antibiotic beads in the first stage, followed at 4-6 weeks by second stage bone grafting, gave good results. Radical diaphyseotomy was not performed. In small defects of the tibial shaft of about 8-10 cm, segmented iliac crest biconcortical grafts usually result in restoration of the continuity of the tibia if there is a good vascular bed and some continuity of the periosteal tube. The surface area for incorporation and revascularisation is increased by segmentation as compared to long cortical slabs. The majority of patients can be treated this way. Fibula transfer should be reserved as a salvage procedure for long bone defects with poor overlying skin or a fibrotic avascular bed. It is an alternative to amputation. Hypertrophy of the shaft occurs with weight bearing.

No benefits of any form have been received from a commercial party related directly or indirectly to the subject of this article.

References