

Management of thromboangiitis obliterans using distraction osteogenesis: A retrospective study

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ABSTRACT

Background: Thromboangiitis obliterans (TAO), also known as Buerger disease, is characterized by thrombosis in medium-sized arteries and veins along with a marked inflammatory response. TAO can be managed by using the principle of distraction osteogenesis to induce neoangiogenesis. We report thirty patients of TAO in the age-group of 20–50 years were treated with the Ilizarov method.

Materials and Methods: Patients with severe rest pain (n=30), absent distal pulsation (n=30) and ulcer (n=6) and/or gangrene of the toes (n=4), who had failed the conservative pharmacological modalities of treatment were included. A lateral tibial corticotomy was performed and distraction applied by use of olive wire. A two-ring frame was used for all cases.

Results: Of the 30 cases, 25 patients became pain free while 1 had partial relief of pain. The four patients worsened on treatment and developed frank infection were amputated. The average distraction was 1.8 ± 0.3 mm (range: 1.5–2.2 cm). The mean consolidation period was 98 ± 11 days, after which the fixator was removed. The total duration of treatment was 122 ± 23 days. There were two cases of osteomyelitis in our series; both healed after debridement and both patients had good result, with relief of pain. The ulcers started healing by 4–6 weeks.

Conclusion: The principle of the distraction osteogenesis including neoangiogenesis can be used for treatment of TAO and has an acceptable complication rate.

Key words: Distraction osteogenesis, Ilizarov ring fixator, thromboangiitis obliterans

INTRODUCTION

Thromboangiitis obliterans (TAO) was first described (1879) by Winiwarter who called it endarteritis.¹ In 1908, Leo Buerger published his observations on young men with severe ischemic changes in the extremities.² These patients were addicted to cigarette smoking and often had migratory superficial phlebitis. Buerger called the syndrome thromboangiitis obliterans because the acute histologic features were characterized by thrombosis in both arteries and veins and were associated with a marked inflammatory

response. The condition became more commonly known as Buerger disease.^{2,3} TAO is commonly seen in the age-group of 20–40 and although there is a male predominance several cases have been reported in women.^{3,4} The disease has also been reported infrequently in nonsmokers.^{3,5} TAO clinically presents as distal ischemia, with patients having claudication, rest pain, and toe ulcers.⁶ Discontinuation of tobacco in all forms is the first and most effective step.^{7–9} Medications such as isopraste,¹⁰ clofibrate,¹¹ cyclophosphamide,¹² and calcium channel blockers¹³ have been used with varying success. Surgical modalities include thrombolytic therapy, arterial reconstruction, bypass vein grafts, lumbar sympathectomy, microvascular omental transfer and, when required, amputation.^{3,14–17} Newer modalities like spinal cord stimulation, angiogenesis by autologous bone marrow mononuclear cell implantation, and gene therapy have shown promise.^{18–23} However, the ideal modality for treatment of TAO is still undecided.²⁴ One of the principles of treatment of chronic ischemia is to achieve neovascularization. Both periosteal elevation and distraction osteogenesis have been shown to increase neovascularisation.^{25–28} Ilizarov has studied effect of distraction osteogenesis and shown that corticotomy and distraction of bony fragments increases the blood supply to the entire limb. The intensive formation of new blood vessels under the influence of tension stress takes place not

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only in the bones but also in the soft tissues.^{26,29} He was the first to use distraction osteogenesis in the management of TAO. In the treatment of TAO, bone widening stimulates considerable vascular hypertrophy in the regenerate, without altering limb length.²⁹ A few other studies have also been reported from Russia,³⁰⁻³⁵ but only two studies are reported in the English language.^{36,37} We retrospectively analysed 30 patients of TAO treated by distraction osteogenesis.

MATERIALS AND METHODS

Between 1990 and 2008, 30 consecutive patients of TAO with stage III or stage IV ischemia, in the age-group of 25–50 years (mean age: 35 years), were treated by distraction osteogenesis at our hospital. Patients with age more than 50 years, diabetes, severe uncontrolled infection, or absent femoral pulse were not considered for the procedure. Nine patients had undergone lumbar sympathectomy without beneficial effect before admission to our hospital. All 30 patients had history of heavy smoking for many years. We initially checked the pulsation of the lower limb arteries, i.e., the dorsal pedis, posterior tibial, popliteal, and femoral. Patients with nonpalpable posterior tibial or dorsalis pedis arteries were considered for this procedure. Color Doppler study was done to ascertain the diagnosis and assess the status of the distal vasculature. It showed conversion from triphasic to biphasic flow in all cases, indicating severe stenosis. All patients had severe rest pain and claudication distance was less than 30 meters. Six patients had ulcers over the toes and four had frank gangrene of the toes. The popliteal artery was palpable in 14 patients. Venous engorgement and ankle and foot edema was found in three patients. Arteriography was done in two patients only; it showed block of the medium-sized arteries of the affected limb, with normal large vessels.

Technique

Corticotomy was performed on the lateral surface of the tibia.³⁸ The upper end of the corticotomy was kept around 3 cm away from the tibial tuberosity. Careful markings with methylene blue were first made on the patient's leg as part of preoperative planning [Figure 1a and b], with the entire longitudinal length being 12 cm. The end transverse incisions were 3 cm long and made such that they crossed the tibial crest with 2 cm on the medial side and 1 cm on the lateral side. The other transverse incisions were only 1 cm long and were taken on lines 3, 6, 7, 8, 9, and 10. A drill bit was passed from the medial to the lateral cortex, starting 1 cm away from the tibial crest on the medial cortex and angling the bit to exit the lateral cortex around 2 cm away from the tibial crest [Figure 1c-e]. A K-wire was then inserted through this hole to act as a guide for the second drill hole in the medial cortex. Similar drill holes were created at points 6, 7, 8, 9, 10, and 11. Three more drill holes were made in the proximal and distal 3-cm long incisions. These holes were

then connected with a 5-mm osteotome. The osteotome's edge was inserted in the first drill hole on the medial side and it was angled towards the second hole. The cortex below the first drill hole was cut. The osteotome was now inserted in the second hole and angled towards the first hole, and the cortical cut was then completed between the first and second drill holes [Figure 1f and g]. Similarly, osteotomy was completed between the holes 6-7, 7-8, 8-9, 9-10, and 10-11. Three sides of the rectangle were osteotomized. Two 5-mm osteotomes were then inserted in the longitudinal cut and turned 90°, cracking the fourth side of the rectangle [Figure 1h]. Thus, a rectangular corticotomy of a total size of 3 × 12 cm was performed. Three olive wires equidistant to each other were then passed through the longitudinal cut into the lateral cortex of the rectangular piece. The distal end of the olive wire was connected to the slotted threaded rod, which was connected the longitudinal plate of the Ilizarov frame. The frame consisted of two rings [Figure 1i-k].

Postoperative care

The distraction was started after a latent period of 9 days. The latent period was extended if the osteotomy was displaced by 5 mm to start with, and such latency is advisable to prevent poor regenerate. We aimed at a distraction of 1 mm per day till a distraction of 20 mm was achieved;³⁰ however, the distraction rate was customized according to the quality of the regenerate. The patient came for followup at 3 weeks post surgery and regenerate quality was assessed on the radiograph. When the regenerate was poor, the distraction was stopped for a week and then started again. The decision to remove the apparatus was based on radiographic appearance of good callus at the distraction site, as judged by the presence of homogeneous radiopacity. Olive wires were removed by cutting off the ends and by gentle hammering. During the entire period the limb was not immobilized in any cast or splint and patients were encouraged to bear full weight from the first postoperative day itself. Final assessment was done as per the suggestions of Patwa and Krishnan³⁶ and the results were classified as 'excellent' (no rest pain, healed ulcers or amputation stump, no claudication, resumption of previous occupation, and unaffected domestic ambulation); 'good' (absence of rest pain, healed ulcers or amputation stump, claudication distance of 1–2 km, able to continue previous occupation, and domestic ambulation affected sometimes); 'fair' (absence of rest pain, persisting/recurrent ulcers, claudication distance of <1 km, change to alternative occupation, and routine domestic ambulation affected); and 'poor' (major amputation needed).

RESULTS

After a latency period of 10 days, the distraction was started. The average distraction was 1.8±0.3 cm (range: 1.5–2.2 cm). The mean consolidation period was 98±11 days (range: 84–112 days), after which the fixator was

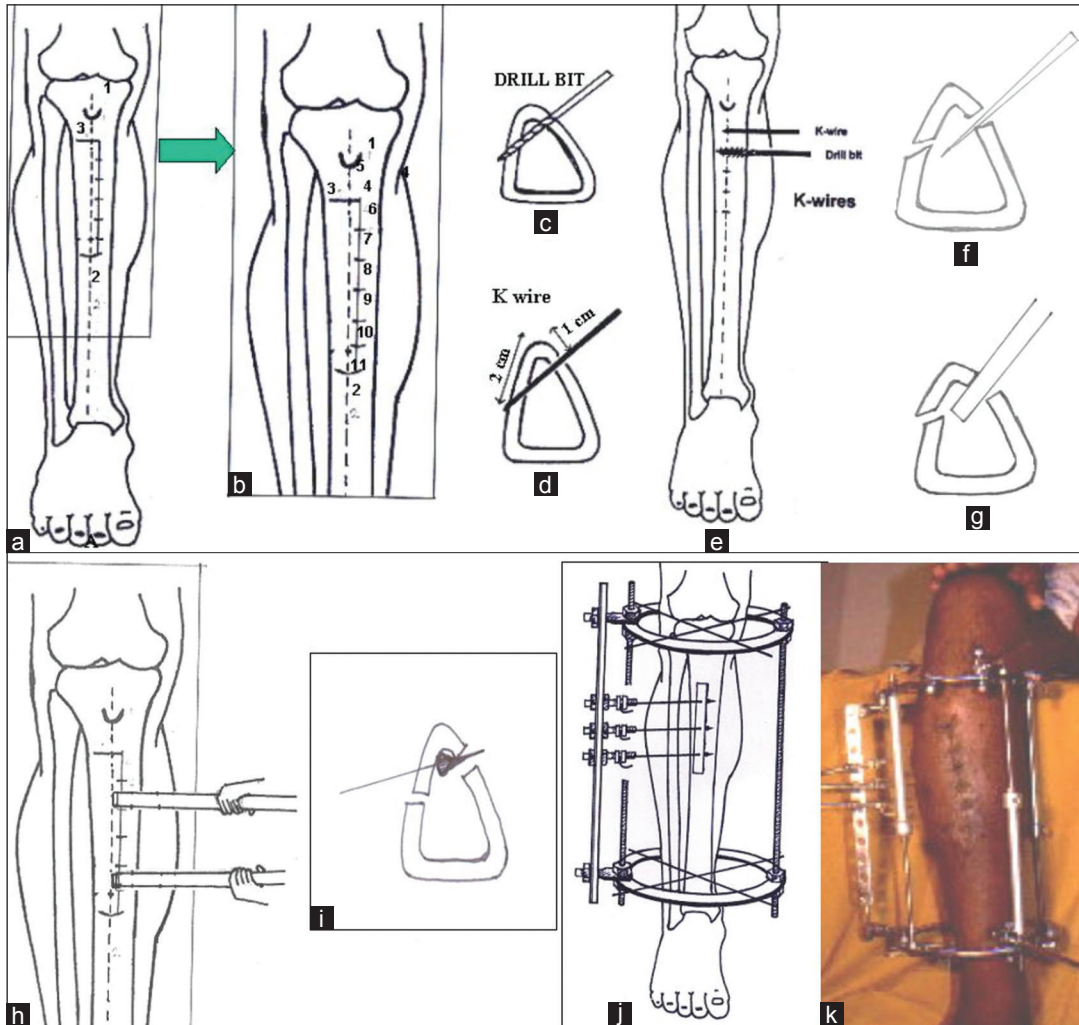


Figure 1: (a,b) Methylene blue markings on the leg; (c,d,e) drilling angle as per preoperative planning to define a rectangular corticotomy; (f,g,h) osteotome used to complete the corticotomy; (i) olive wires are passed through the longitudinal cut into the lateral cortex of the rectangular piece; (j) line diagram showing the wires are attached to threaded rods and a two-ring frame is attached to the limb (k) clinical photograph of same patient

removed. Fixator removal was guided by patients' pain relief and onset of ulcer healing. The total duration of treatment (duration from surgery to return to work) was 122 ± 23 days (except in the case of one patient, who returned to work 5 months post surgery). The mean followup was 4.58 ± 3.1 years (range: 2–20 years) at which time the final results were evaluated. In four patients the pain did not subside and the gangrene of the toes worsened. These patients had to have below-knee amputation. One patient had no relief of pain even though his ulcers healed. The remaining 25 patients became pain free and were satisfied. According to our clinical scoring system, 25 patients had excellent results, 1 had a fair result, and 4 patients had poor results. In the patients with excellent results, the ulcers started to show signs of healing by the end of the first month [Figure 2] and most of the ulcers had healed by the time the fixator was removed. Trophic changes also showed signs of reversal, with decrease in skin discoloration. All the corticotomies had united at



Figure 2: Clinical photographs showing nonhealing wound over the great toe (a) started healing by the 5th week post surgery (b) and remained healed at 3 years' followup (c)

the end of the followup period [Figure 3]. Two patients developed osteomyelitis of the corticotomy fragment. In one case, this was due to erroneous placement of the

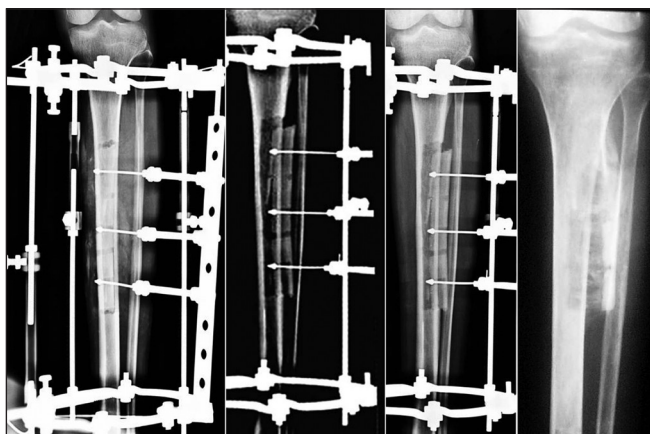


Figure 3: X-rays of leg bones (anteroposterior view) showing a corticotomy procedure at various stages of distraction followed up to 21 months post surgery, with complete union of the corticotomy

wires, with the fragment being pulled anteriorly, leading to gaping of the lower two incisions which was noticed at 4 weeks' followup. This resulted in osteomyelitis of the fragment, which was therefore removed. The second case of osteomyelitis of the fragment developed at the end of the distraction period. This was treated with removal of implant and debridement. Surprisingly, however, both patients had relief of pain.

DISCUSSION

Various surgical modalities are used for the treatment of severe TAO. Although distraction osteogenesis has been shown to stimulate neoangiogenesis, widespread use of this technique in the treatment of TAO is not reported. We have used this technique in 30 patients with acceptable results.

The exact incidence of Buerger disease in India is not known, but it is not an uncommon disease. In a retrospective review of 100 patients with ischemic toe ulcerations, TAO proved to be the final diagnosis in 9%.³⁹ In another study of 700 patients with small-artery disease, TAO was the final diagnosis in 3.7%.³ At present, patients with TAO comprise less than 1% of all patients with peripheral vascular disease in the US. In Israel and Eastern Europe, the incidence is approximately 5%, whereas in Japan it has been reported to be 16%.³

The clinical course of Buerger disease is protracted and painful, but relatively benign. In this series, all patients reported being heavy smokers for many years, most of them since adolescence and smoked *bidis* (made by filling a leaf-tube with raw tobacco). The disease usually starts in one limb but both the lower limbs may be affected. If the patient continues to smoke, the disease may affect the upper limbs; approximately 30% of patient with Buerger

disease have involvement of their upper extremities.⁴⁰ Small- and medium-sized arteries are usually involved and the forearm, calf, or digital arteries may be occluded. The femoral and brachial arteries are usually not involved. Arterial reconstruction is usually impossible because of the distal nature of the disease, but it should be considered in segmental proximal occlusions. Microvascular transplantation of free omental grafts to areas not amenable to arterial reconstruction has been successfully employed,^{3,17} as have pedicled omental grafts.^{41,42} When gangrene occurs, amputation at the lowest possible level is indicated. In this disease, unlike in arteriosclerosis, it is often possible to do amputations of the digits with satisfactory healing.

Distraction osteogenesis can be very useful in the treatment of TAO. Corticotomy of the tibia with distraction causes neohistogenesis and increase in collateral circulation in the limb.²⁶ Ilizarov originally used the posteromedial cortex for distraction;²⁹ the ASAMI group recommended distraction of the lateral cortex, and this was the method used by us.³⁸ Meticulous care has to be taken during the planning and execution of the procedure. All incisions must be transverse ones so that, with lateral distraction of the fragment of the tibia, the wound closes. If a longitudinal incision is made, there will be a diamond-shaped gaping of the wound. This may lead to infection, and the fragment may become a sequestrum. One of our patients developed infection secondary to inappropriate wire placement, which shows that the surgeon should always be vigilant about the direction of pull by the wires when distraction begins.

Kelkar *et al.* used neovascularity created as a part of the inflammatory response to the corticotomy and periosteal elevation.²⁵ Local inflammation augments tissue oxygenation by increasing the local blood circulation through arterioles, capillaries, and venules. He included 61 patients with severe occlusive arterial disease (44 with thromboangitis obliterans, 13 with atherosclerosis and 4 with diabetes mellitus) in 50 patients in their series there was complete relief from pain at rest and amputation could be indefinitely postponed. However, these patients had to remain non-weight-bearing for 6–8 weeks. The complication rate was also high, with four patients having wound infection and three developing fracture of the tibia shaft; thus 16% (7/44) had poor results in thromboangitis group.²⁵ In our series, we had no case of tibia fracture, but there were two cases of infection. Out of the 30 patients in our series 25 had pain relief, indicating a good success rate. A comparison with other series that have used the same technique of horizontal distraction osteogenesis is presented [Table 1] and our results are similar to that reported by others. We can deduce that this technique is effective in

Table 1: Comparative analysis of results in different published series

Study	Year	No. of pts	Inclusion	Exclusion	Followup	Result	Complications
Shevstov <i>et al.</i> ³⁰	1998	265	TAO	-	3 years	236 good results	-
Shevstov <i>et al.</i> ³¹	1997	94	TAO	-		57%–70% positive results	-
Fokin <i>et al.</i> ³²	1990	28	TAO	-	2–19 months	Good results (n=18) 64%	2 fractures, 4 infections
Fokin <i>et al.</i> ³³	1993	125	TAO	-	21.6 months	94.2% good results	2 fractures, 16 local infections
Chaudhary <i>et al.</i> ³⁷	2001	30	TAO	-	21 patients with >2 years followup	>90% good results	3 below-knee amputations
Patwa <i>et al.</i> ³⁶	2011	60	TAO	Diabetes, immunocompromised patients, patients on long-term steroids	Mean 5.4 years	Excellent to good (n=48), 80%	18 pin tract infection, 2 fractures, and 2 amputations
Our series	2011	30	TAO	Age>50, diabetes, uncontrolled infection, absent femoral pulse	Mean 4.5 years	Excellent (n=25) 83.3%	2 infection, 4 amputations

TAO - Thromboangiitis obliterans

unreconstructable TAO, providing good long-term relief.

In conclusion, distraction osteogenesis can be used in patients of TAO and gives good results with acceptable complication rates. However, a larger series and, possibly, comparative studies with pharmacological agents will be required to assess the real benefits of the procedure.

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