Original Article

Reconstruction by bone transport after resection of benign tumors of tibia

A retrospective study of 38 patients

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Abstract

Background: The commonly used reconstructive options after post resection defects in bone tumors like megaprosthesis, autograft, allograft, bone graft substitutes and recycled bone have their own demerits on a long term. Bone transport that regenerates patient's own bone is a less explored option of reconstruction after resection of benign bone tumors and reports on this are limited. This technique is very much relevant in tibia where Ilizarov fixator is surgeon and patient friendly. We report our experience.

Materials and Methods: This is a retrospective series of resection and bone transport in 38 patients with benign tumor of tibia. There were 14 males and 24 females with mean age of 23.40 years (range 9–40 years). Lesion was located in proximal third tibia in 27, middle third in two and distal third in nine patients. The diagnosis was giant cell tumor in 32, chondroblastoma in three, chondromyxoid fibroma, enchondroma and desmoplasic fibroma in one patient each. The resection was intercalary in 28 and transarticular in 10 patients. Osteosynthesis was monofocal in three, bifocal in 31 and polyfocal in four cases.

Results: Mean followup was 7.22 years (range 1.5–15 years). Mean resection length was 10.21 cm (range 3–22 cm). The mean duration of external fixator was 308.03 days (range 89–677 days) and mean external fixator index was 36.14 days/cm (range 16.84–97.43 days/cm). Twelve patients had difficulties in the form of 11 problems and five obstacles that were successfully managed. None of the patients had local recurrence of tumor or any long term complication. Mean Musculo-skeletal Tumour Society score at final followup was 27.18 (90.60%).

Conclusions: Bone transport is an excellent option after resection of benign tumors of tibia with good local control and functional outcome, despite minor difficulties that need timely management.

Key words: Benign bone tumor, resection, bone defect, bone transport, distraction osteogenesis, ilizarov
Mesh terms: Ilizarov technique, bone lengthening, osteogenesis, distraction, bone neoplasms

Introduction

Primary bone tumors are more commonly benign than malignant[1] and tibia is the second commonest site accounting for 27.60% of all benign bone tumours.[2] Local resection/wide excision is associated with low rates of local recurrence compared to intralesional treatment,[3] but the reconstruction of the resultant bone defect remains challenging.[4] Resection of benign tumors has to be as much joint and physis-sparing and as much biological as possible to ensure good long term results.[5] Reconstruction of juxta-articular defects poses a special problem with regard to space available for implant purchase.[6] The commonly used options of endoprosthesis, autograft, vascularized autograft, allograft and bone graft substitutes have their own disadvantages and long term results are not satisfactory, especially after resection of large segments and juxta-articular tumours.[7]

Distraction osteogenesis by the external fixator of Ilizarov has shown promising results in extensive bone defects associated with trauma, nonunion and infection[8] and the fixator is more surgeon and patient friendly in the leg than at other sites. Ilizarov reconstruction gives the added advantage of concurrent filling up of soft tissue defects, biomechanical stability, early functional loading and opportunity for limb lengthening and deformity correction

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later especially in pediatric tumors. It also increases the possibility of joint sparing and physis-sparing resection as less subarticular bone is needed for purchase compared to implants used in other forms of reconstruction.

Bone transport has been sparingly used after resection of benign tumors and reports on this are less in number.[4,9] Ours is a center with vast experience in Ilizarov methodology in the management of large bone defects. We believe that it is a biological and robust reconstructive option when it comes to large postresection defects of bone tumors. With benign tumors the challenges of concurrent chemo-radiotherapy do not exist as with their malignant counterparts. Since these tumors do not affect the survival of the patient, the surgeon is bound to select the most enduring reconstruction modality out of all options available. In this background, we undertook a retrospective study of benign tumors of tibia treated by resection and reconstruction by distraction osteogenesis by Ilizarov external fixator. The objective of the study was to assess the results in terms of local recurrence, occurrence of challenges and complications and long term function.

Materials And Methods

38 patients with a mean age of 23.40 years (range 9–40 years) with a benign tumor of the tibia treated with resection and reconstruction of the resultant bone defect with bone transport using Ilizarov apparatus between 1985 and 2010 were included in this retrospective study. [10] There were 14 males and 24 females. The institutional approval was taken. Only patients having minimum followup of 18 months were enrolled. We reviewed all clinical notes, radiological investigations and pathological reports and data were available for all cases. The tumor location was proximal third of tibia in 27 cases, middle third in two cases and distal third in nine cases. All patients underwent thorough clinicoradiological assessment. The diagnosis was giant cell tumor in 32, chondroblastoma in three, chondromyxoid fibroma in one, enchondroma in one and desmoplasic fibroma in one patient [Table 1]. Out of 32 giant cell tumors, 25 were of Campanacci[11] Grade II and seven Grade III. One of the cases of giant cell tumor was recurrent following curettage and bone grafting at another hospital 3 months back.

Whenever it was possible to achieve good clearance with enough subchondral bone to pass wires for a single ring in the juxta-articular fragment, we did intercalary joint sparing resection. Accordingly the resection was intercalary in 28 and transarticular in 10 tumors of which seven belonged to proximal and three distal tibia. Eight out of 10 lesions that needed transarticular resection were giant cell tumors extending close to articular cartilage [Figure 1]. The other cases were aggressive epiphyseal chondroblastoma [Figure 2] and enchondroma with suspicion of malignancy. 15 of these 38 patients (giant cell tumour - 12, chondromyxoid fibroma - 1, enchondroma - 1, Desmoplastic fibroma - 1) were <18 years of age with open physis. Thirteen of them were intercalary resections and all of them were physis-sparing. The tumors were resected with a margin of 2 cm, wherever possible. In juxta-articular intercalary resections just enough bone to permit, reconstruction was left in the joint side. After closure of surgical wound, Ilizarov external fixator was applied to leg segment. Excluding the seven patients who had knee arthrodesis, out of 31 patients, knee had to be immobilized in 10 cases for an average of 4.5 months. We planned monofocal osteosynthesis (no corticotomy) for small defects, where acute docking is attained after tumor resection and kept compressed for 3 weeks, following which the same site is distracted gradually to achieve limb length. The majority of defects needed bifocal osteosynthesis (docking - one level, corticotomy and distraction - one level) with corticotomy of uninvolved metaphysis. Polyfocal transport (docking - one level, corticotomy and distraction - two levels) was planned in cases of extensive defects and two cases of knee arthrodesis involved transport of a femoral segment down [Figure 3]. After a latent period of 7 days (3 weeks in case of monofocal osteosynthesis), distraction was commenced at the corticotomy site(s) at a rate of 0.5–1 mm/day and progressively adjusted according to clinicoradiological

Table 1: Distribution of disease and treatment related variables and results among histopathological diagnosis groups

Diagnosis	Number of patients	Mean age (years)	Sex	Location	Type of resection	Mean RL (cm)	Type of osteosynthesis	Mean EFI (days/cm)	Mean MSTS score
Giant cell tumor	32	24.13	Male: 12	P/3: 23	IC: 24	10.06	Mono: 3	28.04	26.97
			Female: 20	M/3: 1	TA: 8		Bi: 25		
				D/3: 8			Poly: 4		
Chondroblastoma	3	24.33	Male: 1	P/3: 2	IC: 2	13.33	Bi: 3	36.25	29.33
			Female: 2	M/3: 1	TA: 1				
Chondromyxoid fibroma	1	10	Male: 1	P/3: 1	IC: 1	5	Bi: 1	33.60	28
Enchondroma	1	21	Female: 1	P/3: 1	TA: 1	12	Bi: 1	32.67	26
Desmoplastic fibroma	1	13	Female: 1	D/3: 1	IC: 1	9	Bi: 1	43.56	28

RL=Resection length, EFI=External fixator index, MSTS=Musculo-skeletal Tumor Society, P/3=Proximal third of tibia, M/3=Middle third of tibia, D/3=Distal third of tibia, IC=Intercalary, TA=Transarticular, Mono=Monofocal, Bi=Bifocal, Poly=Polyfocal

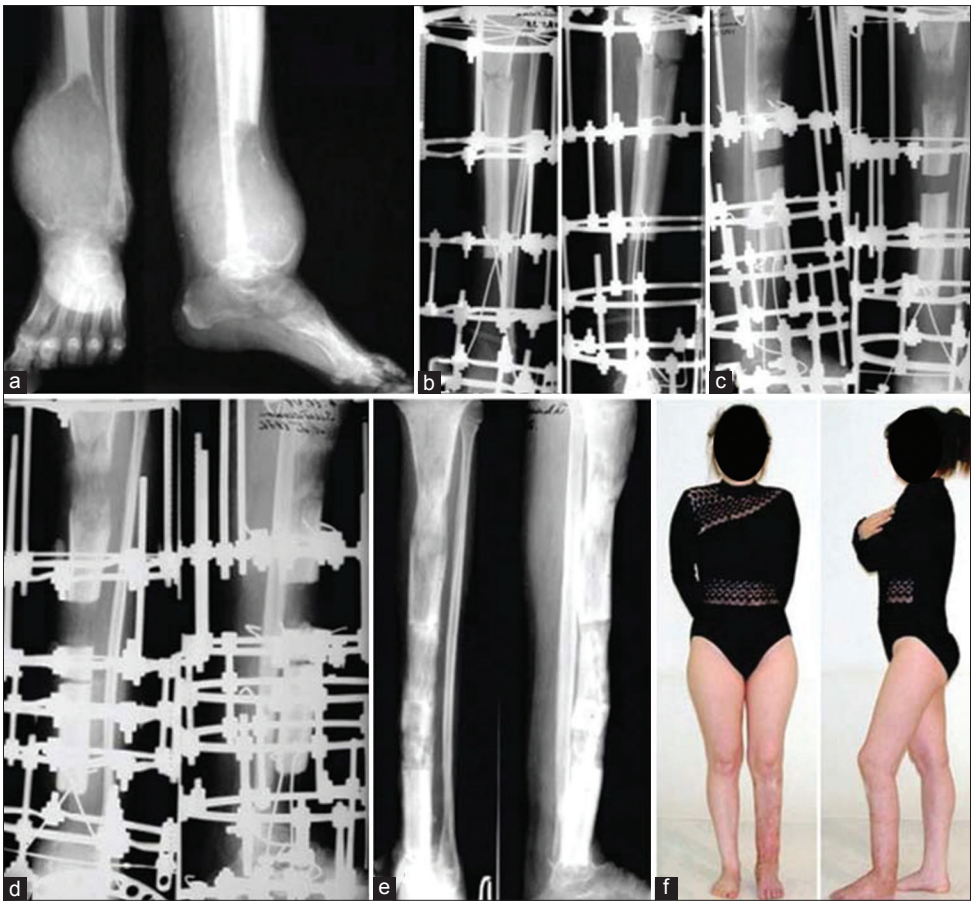

Figure 1: (a) X-rays anteroposterior and lateral views of leg bones with ankle of a 33 year old female with giant cell tumor of left distal tibia. (b-d) X-rays leg bones anteroposterior and lateral views during the period of distraction osteogenesis to fill up 8 cm defect after transarticular resection. (e) X-rays leg bones anteroposterior and lateral views showing consolidation (f) clinical photograph at 1 year followup

Figure 2A: (a) X-rays leg bones with knee joint anteroposterior and later views of 18 year old female showing aggressive chondroblastoma of proximal third of right tibia (b) X-rays during bone transport to fill up 17 cm after joint sparing resection

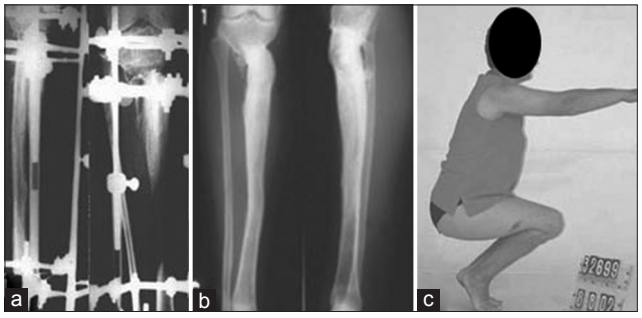

Figure 2B: (a) X-rays of same patient showing bone transport in progress (b) X-rays anteroposterior and lateral views at 15 years followup showing very good consolidation (c) Clinical photograph at 15 years folowup showing range of motion

assessment. Bone was gradually transported into the defect following osteogenesis at the site (s) of distraction and upon achieving cortical contact compression was applied at the site of the defect.

The patients started crutch-assisted partial weight bearing walking from 2nd day of surgery and gradually progressed

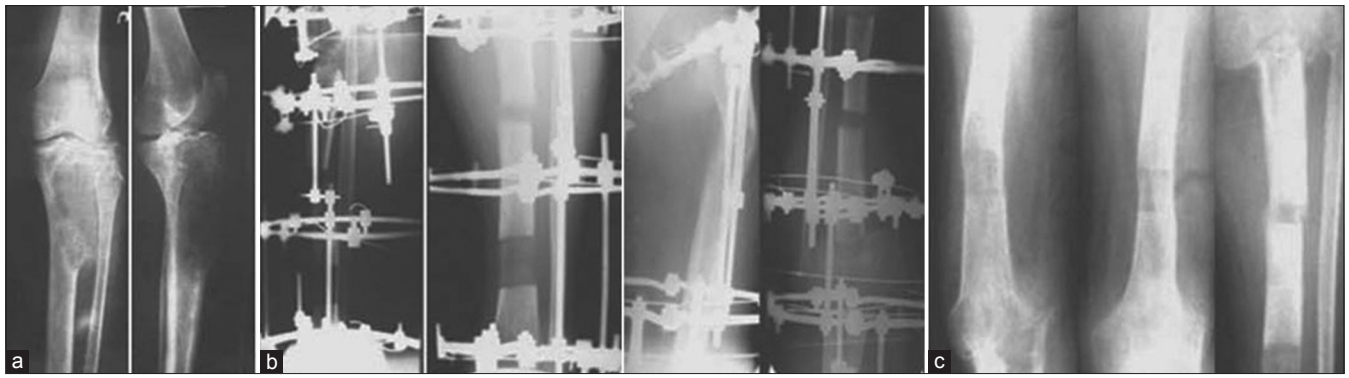

Figure 3A: (a) X-rays anteroposterior and lateral views of a 32 year old male showing recurrent giant cell tumour of proximal tibia (b) Reconstruction of 19 cm defect by lengthening of tibial and femoral segments using bone transport with Ilizarov (c) X-rays after removal of fixator showing femoral and tibial regenerate and arthrodesis

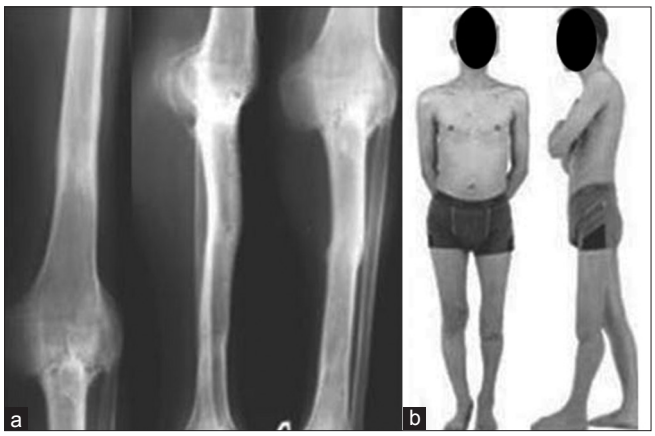

Figure 3B: (a) X-rays at 5 years followup showing consolidation (b) Clinical photograph at 5 year followup showing patient full weight bearing

to full weight bearing by 4 weeks. Radiographs were taken every 2 weeks to assess the quality of regenerate, occurrence of any deformity and monitoring the direction of bone transport. After sufficient length and compression at the site of the defect were achieved, the fixator was left in place to allow bony union at the site of the defect and good consolidation of regenerate. Decision to remove the fixator was taken after satisfactory stress testing (after removing the connecting rods) at the site of the defect and regenerate and signs of satisfactory radiological union and consolidation as evidenced by bony bridging in orthogonal views. Once the decision of removing the frame is taken, a few wires and rods were gradually removed over days to physiologically stress the sites of defect and consolidated regenerate and then fixator removed totally. The patients were then given cast extending from thigh to supramalleolar region for a variable period of 2–4 weeks depending on the findings of the stress testing during removal of fixator.

Difficulties related to Ilizarov technique encountered during and after treatment were classified[12] as problems, obstacles and complications. Problems were the ones that settled without any intervention, obstacles needed intervention, but could be solved prior to end of treatment and complications were the ones that could not be resolved prior to completion of treatment. Other complications and reoperations were also systematically recorded. We calculated the lengthening index, maturation index and external fixator index[13] as the number of days spent in distraction phase, the maintenance phase and total duration of external fixation respectively for reconstructing 1 cm defect. The patients were followed up once in 3 months for 2 years, once in 6 months for the next 3 years and yearly thereafter. We monitored for signs of local recurrence of the tumor, knee and ankle function, limb length discrepancy or deformity at the site of regenerate. The functional status at followup was recorded using the Musculo-skeletal Tumor Society (MSTS) scoring system[14] where each of the six parameters – pain, functional activities, emotional acceptance, use of supports for ambulation, walking ability and gait was given a score out of five and added together to obtain a score out of 30.

Results

None of the patients was lost to followup and mean followup was 7.22 years (range 1.5–15 years). Mean resection length was 10.21 cm (range 3–22 cm) and according to the size of defect bone osteosynthesis was monofocal in three, bifocal in 31 and polyfocal in four cases. Out of four cases that needed polyfocal osteosynthesis, the second corticotomy was performed in the femur in two cases that went for knee arthrodesis and two on the tibia that finally went for ankle arthrodesis. The distraction started on 7th day of surgery at 0.50–1 mm/day at each corticotomy site and modified according to progress of bone formation. The cases of transarticular resection of the knee (seven) and ankle (three) resulted in arthrodesis [Figure 4]. Mean duration of a distraction phase was 127.11 (49–238) days, maintenance phase 175.92 days (range 28–504 days) and mean duration of wearing external fixator was 308.03 (range 89–677 days). Mean lengthening index was 15.49 days/cm

Figure 4: (a) X-rays ankle joint with distal half of leg of a patient with Giant cell tumour of distal tibia (b) X-rays during bone transport to compensate 11 cm defect. (c) clinical photographs during bone transport showing ilizarov fixator *in situ* (d) X-rays anteroposterior and lateral views at 5 years followup showing consolidation

(range 5.89–56 days/cm), maturation index 20 days/cm (range 5.60–50.75 days/cm) and external fixator index was 36.14 days/cm (range 16.84–97.43 days/cm).

One patient had intraoperative complication in the form of iatrogenic fracture while performing the corticotomy, which was immediately identified and managed by adding additional wires and fixing the fractured fragment. Difficulties during the course of treatment were observed in 12 patients of whom two had more than one difficulty. Nine patients encountered a total of 11 problems in the form of pin tract infection in eight and common peroneal neuropathy in two and delayed regeneration in one patient. The infection settled with regular dressings and antibiotics. The nerve function recovered in both patients by reducing the dose of distraction, splinting, passive range of motion exercises and neurovitamin supplementation. Delayed regeneration settled with watchful expectancy by prolonging the duration of the fixator. Five patients had obstacles of which four were breakage of wires that needed exchange of wires under anesthesia. The other patient had secondary displacement of the fragments at the site of the defect during the maturation phase and needed reduction and realignment under anesthesia. One patient who had extensive defect of 19 cm following resection of recurrent giant cell tumor was lengthened in two stages with fixator-free interval of 4 months in between. This patient had lengthening of the femoral segment also to compensate for the huge defect. We did not encounter any problem with union at the docking site. None of the patients had significant complications or long term sequelae.

All patients started partial weight bearing ambulation in the immediate postoperative period and all progressed to full weight bearing by 4 weeks. In the cases that underwent arthrodesis of the knee or ankle, the affected limb was intentionally kept short by 1.50 cm to facilitate ground clearance. All others did not have any limb length discrepancy and all patients were independent ambulators at final followup. Excluding the cases of arthrodesis of the knee, out 31 patients, 21 had full knee range of motion at final followup, the rest four had 90% and six 75% of range of motion respectively, giving an average of 91.87% of knee range of motion. Excluding the cases of ankle arthrodesis, all patients who had intercalary resection of distal third tumor had full range of motion of ankle at final followup, except one who had 90%. The mean MSTS score at final followup ranged from 22 to 30 with a mean of 27.18 (90.6%). The patients with joint sparing resection had better scores (range 28–30, mean 28 [93.33%]) compared to those with arthrodesis (range 22-28, mean 24.90 [83%]). None of the patients developed local recurrence of the tumor. None of patient who underwent joint sparing resection had any evidence of subchondral collapse or arthritic changes and knee extensor mechanism was good in all cases at final followup.

Discussion

In spite of the various biological and nonbiological options available, there is no gold standard technique for reconstruction of bone defects after tumor resection.[5] In the long term, megaprosthesis can cause problems like lack of the full range of motion, infection, aseptic loosening, mechanical failure and revisions, including the chance for amputation.[15,16] Autografts are limited by availability, donor site morbidity, inability to compensate for large defects, issues with union and chance of lysis, resorption and fracture. Vascularized autografts demand technical expertise and infrastructure which may be limited in many centres.[17] Allografts and recycled bone are also fraught with chances of infection, fracture, delayed incorporation, need for prolonged immobilisation and cannot be considered as definite options. Fresh frozen allografts are sparingly available and in addition carry risk of transmission of viral illnesses and immunological rejection.[18,19] Osteoarticular allografts give disappointing results in view of degeneration of articular cartilage and low graft survival.[20] The use of

combination of allograft and vascularized autograft is associated with length limitation, problems with fixation and technical complexity. [21] Synthetic bone is not useful in large defects, segmental defects and in major weight bearing bones.[22]

Distraction osteogenesis is an excellent biological option as it regenerates normal living bone of sufficient strength and durability, biological affinity and resistance to infection that remains intact once formed.[9] Another significant advantage is overcoming the challenge of holding small fragments in subarticular resections, reducing the possibility of the surgical plan going to transarticular resection for fear of difficult reconstruction. This increases the possibility of joint sparing and physis-sparing resections that have a definite impact on long term function.[23] Also once restored, there is negligible fear of infection, mechanical failure and a chance of revisions. In the resection of paediatric tumors that sacrifice the physis, future limb length discrepancy can be predicted and over-lengthening can be done at the time of primary treatment itself without risk of neurovascular deficit or if at all discrepancy occurs in future, it can be easily resolved with the same technique. The possibility to control and guide the bone-forming process[24] ensures precise reconstruction of the defect, which in other techniques may demand custom made prosthesis, special implants, wide variety of allografts and the expertise to properly match and shape them to the defect.

It should be noted that all our patients were initially evaluated at lower tier hospitals and wherever applicable counseled for curettage versus resection for tumor clearance and in cases of resection counseled for conventional versus Ilizarov reconstruction. Many of our patients were the ones who opted for resection even when the option of curettage was possible (in view of reducing the chance of local recurrence). In cases where resection was decided they opted for Ilizarov reconstruction and hence referred to our institutions. Some of them were late presenters with large volume disease. We also had an unusual presentation of tumors like metadiaphyseal and diaphyseal location of chondroblastoma[25] and giant cell tumour.[26] In many of these cases where we succeeded in a joint sparing resection and biological reconstruction, the alternate option available was transarticular reconstruction and megaprosthesis that would have sacrificed the native joint. All the difficulties that we encountered were either self-limiting or could be successfully resolved by intervention. All reinterventions were limited to the period of wearing external fixator, unlike the reports of intralesional treatment where recurettages may be needed at unpredictable intervals due to local recurrence of tumour.[3] It is important that the patients must be counseled regarding the nature and duration of treatment, and rehabilitation, chances of the period of external fixation getting prolonged and possibility of repeated interventions. The limb length could be restored in all cases and none of our patients had long term sequelae. The absence of local recurrence ensures adequate clearance of the tumor. However Tsuchiya *et al*.[9] has documented neovascularisation associated with distraction as evidenced by 1.7–2.3 times increase in blood flow to the limb by 99mTc angiography. Based on this they have cautioned that distraction osteogenesis may increase the chance of local recurrence of tumors, especially the aggressive ones like giant cell tumor and reemphasized the importance of adequate tumor clearance and satisfactorily wide margins. The functional outcome as measured by range of motion of joints and MSTS scoring was also encouraging.

We conclude that bone transport by Ilizarov external fixator is an excellent reconstruction option after resection of benign tumors of tibia with good local control and functional outcome.

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