

Managements of Type 2 Tibial Hemimelia; Short Review on Treatment of Tibial Defects and The Results of Four Patient Treated Using a Modified Brown Procedure

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ABSTRACT:

BACKGROUND:

Tibial hemimelia is a very rare deformity and problematic syndrome usually associated with variable visceral and skeletal deformities. The treatment is always difficult and challenging. Amputation was the preferred treatment option specially in complete tibial absence, however, a conservative management sometimes used in other types of the deformity or in cases of amputation refusal.

OBJECTIVE:

To review the methods of management of chronic tibial defects and to study the results of operative treatment of 4 patients with type ii congenital tibial hemimelia.

PATIENT AND METHOD:

All along thirty-two months period and after a proper clinical and radiological assessment we treated four male children with a mean age of 8.5 years who were all right sided type 2 tibial hemimelia according to Jones and Kalamchi with absence of the distal tibia. Treatment consisted of proximal fibula transfer to the lower end of the tibial stump (brown's operation) followed by Syme's amputation and prosthetic fitting.

RESULTS:

In all cases the transferred fibular shaft to the distal end of the tibial remnant was united within two to three months. The fibulae grew in length and the size to be tibialized and the leg increased in length as we preserved the distal fibular epiphysis.

CONCLUSION:

The results were acceptable as we saved and reconstructed the limb, we gained an acceptable knee function and stable prosthetic fitting that aided proper rehabilitation.

KEY WORDS: brown's procedure , tibial hemimelia.

INTRODUCTION:

Congenital deficiency of the tibia; including tibial hemimelia; aplasia; and dysplasia; is a very rare tibial longitudinal lower limb deficiency occurring in one per million live births. Congenital deficiency of the tibia should be differentiated from the common congenital deficiency of the fibula with a short tibia. In congenital deficiency of the tibia, the fibula is usually intact but there is aplasia or marked dysplasia of the tibia. Children with this abnormality are usually born with marked shortening and bowing of the involved leg. Flexion contracture of the knee and a skin dimple overlying the proximal tibial region are commonly present. The foot is quite rigid in

varus and supination, and points inward toward the perineum. There is marked shortening of the first metatarsal.⁽¹⁾

In most cases there is no clear etiology for congenital tibial deficiency, still, some autosomal dominant forms and rarely recessive forms are seen. For the hereditary forms, disease is usually bilateral, in a retrospective review in 22 year, tibial deficiency patients showed 79% association anomalies of hip, hand or spine.⁽²⁾

For classification of tibial hemimelia Jones system is useful one it's based on radiographic features present during infancy, it classify it into four types according to the x-ray findings.⁽²⁾

Type 1, the tibia cannot be seen on radiographs at birth. In subtype 1a the tibia is completely absent and the ossific nucleus of the distal femur is small or absent. In type 1b the cartilaginous analogue of the tibia is present at birth but it is not ossified and can only detected with ultrasonography or MRI and the distal femoral epiphysis is normal.

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Type 2, the proximal part of the tibia is present at birth radiographically but the distal tibia is not seen.

Type 3 the distal part of the tibia is present and ossified but the proximal portion of the tibia is

absent. This type is the least common form.

Type 4, the tibia is short and there is distal tibiofibular diastasis. In this type the articular surface of the distal tibia is absent and there is proximal displacement of the talus (figure 1).⁽¹⁻⁴⁾

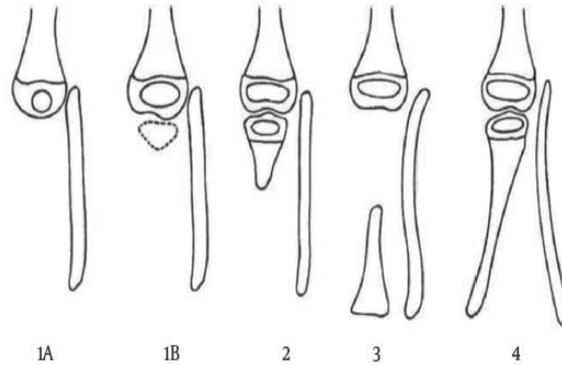


Figure 1: Jones' Classification⁽²⁾

Tibial defects or loss is always challenging for the physician and difficult for the patient. There are Four main causes of these defects: congenital, infections, high energy fractures and bone tumors.⁽²⁾

Variable options are always available for the treatment of tibial defects, however, the choice and prognosis mainly depends on the cause, type and severity of the defect. Treatment of segmental loss of the tibia by tibialisation of the fibula is one of the available options for the control of tibial defects and the following review for tibial defect management is mainly dependent on that of Alireza Rahimnia et al 2011.⁽²⁾

One option is the **Papineau's** Technique which mainly used for chronic infections.^(3,5-7)

Allograft is another usual method for management of bone defects mainly following resection of tumors. Larger grafts found to be complicated by possible greater risk of infection, nonunion, fracture, rejection, and tumor spread.⁽⁸⁻¹⁰⁾

Bone Transport utilizing **Ilizarov technique** in used in osteomyelitis, fractures or agenesis. It involves bone transport with compression-distraction procedure. An advantage of this procedure is the ability to address the associated malalignment during the transfer, also it reduce the risk of bone atrophy and joint stiffness by early patient ambulation.^(3,6,11-14)

The Ilizarov fixator appears to be highly efficient in progressive correction of lower limb length discrepancies and articular or bony angular deviations, it showed good results in congenital tibial deficiencies.⁽¹⁵⁾ Altaf A. et al 2008 reported an excellent results for the treatment of tibial hemimelia using Ilizarov's technique with substantial lengthening and multi-planar deformity correction in a centralized fibula.⁽¹⁶⁾

Free Vascularized Fibular Graft became popular in filling bone defects. The fibular graft is harvested with a long pedicle of peroneal vessels. Those grafts are able to heal soundly with low incidence of infection. However contralateral vascularized grafts associated with high morbidity, its technically difficult and needs experience and equipment. Donor side ankle instability and peroneal nerve injury still a serious possible complications specially with other side pathology.⁽¹⁷⁻²⁰⁾

Medial Fibula Transfer Using Variable Techniques as Tuli's technique mostly for defects caused by fractures and infection he transfer in stages the proximal and distal ends of the fibula to the tibia in a trap door fashion with fixation and POP, (Figure 2).⁽⁷⁾

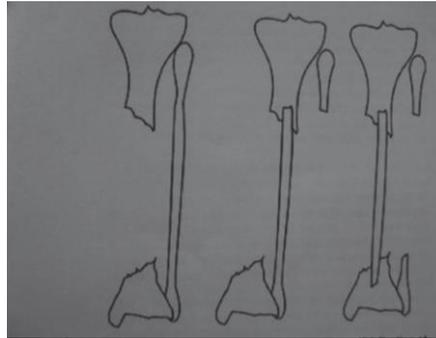


Figure 2: Tuli's technique for medial transfer of the fibula.⁽⁷⁾

Tuli, studied of 21 patients with tibial defects treated by his technique he found that hypertrophy of the fibula occurred in all patients with doubling of the size in four years after the synostosis.⁽⁷⁾ However in other study treating a congenital pseudoarthrosis of the tibia in a boy the bone resorbed rapidly.⁽¹⁰⁾

Medial transport of the fibula with the **Ilizarov frame**. Catagni used the Ilizarov frame for medial transfer of the fibula in seven patients with

massive traumatic tibial defects 7 - 28 cm, patient age ranges between 23- 63 years (Figure 3). Bone graft needed in four sites for two patients for nonunion or to augment new bone formation. All patients were ambulant during the whole procedure. POP cast used after frame removal.⁽¹²⁾ Simillar method is highly effective in managing congenital tibial defect together or deformity correction.^(15,16)

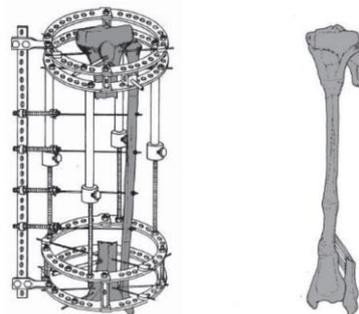


Figure 3: Medial Transfer of the Fibula via the Ilizarov Frame.⁽¹²⁾

Huntington procedure, Cassab used the Huntington technique in doing 11 cases of ipsilateral side to side fibular transposition to the tibia in patients aged between 16 -61 years for defects of 4 -22 cm caused by trauma, osteomyelitis or tumor excision. Osteotomy of the fibula was done and after it was fixed to the

tibia bone grafts was used at the junctions of the two bones (Figure 4). The limb was put in cast and weight bearing postponed until union of the tibiofibular junctions, healing occurred in 8 patients after a mean of 10.5 months and it was strong enough for weight bearing.^(5,13)



Figure 4: Huntington Technique of Medial Transfer of Fibula.⁽¹³⁾

Surgical options for congenital tibial hemimelia:

Most patients with complete tibial hemimelia (Jones type 1a) require knee disarticulation, which usually provides good functional results.⁽²¹⁾ Centralization of the fibula combined with a Syme's amputation (i.e., the Brown procedure) has frequently been used to treat this deformity however, this approach is prone to failure, and the patient often requires a subsequent knee disarticulation. Failures are due to marked knee instability and the progressive development of knee flexion contracture because of unopposed hamstring pull. Thus, the Brown procedure is rarely indicated for patients with complete tibial hemimelia.^(22,23)

For partial tibial hemimelia treatment of Jones types 1b and type 2 deformities, in which the proximal part of the tibia is present, excellent functional results can be obtained by the use of the Brown procedure by fusing the proximal fibula to the upper part of the tibia. Fusion of the fibula to the tibia may be done in an end-to-end position with intramedullary pin fixation or in a side-to-side position using a screw for fixation, all above done with Syme's amputation and subsequent prosthetic fitting, it seems to be the best management for the distal part of the limb because of the severe foot and ankle deformity and instability. After the synostosis has healed, these children are able to function as well as other Syme's-level amputees and can participate in normal sports activities.^(24,25)

For a Jones type 4 deformity, the best treatment is a modified Syme's ankle disarticulation performed when the child reaches walking age. Functional results are usually excellent.⁽²⁵⁾ Other

techniques, such as tibial lengthening and foot repositioning, may make it possible to retain a plantigrade foot, but functional reconstruction is difficult because of talus and calcaneus deformities and the absence of a distal tibial articular surface.⁽²¹⁾

For the rare Jones type 3 deformity, the limited data available show that these patients function relatively well as below-knee amputees following a Syme's or Chopart's amputation. Some of these patients may be candidates for tibial lengthening, depending on the anatomy of the ankle joint.

Authors have attempted to treat tibial hemimelia with surgical equalization of leg length, production of a plantigrade foot, and creation of a stable knee. Traditional leg-lengthening procedures, soft-tissue reconstruction, and casting have not reliably achieved these goals in patients with tibial hemimelia.⁽²⁴⁾

PATIENTS AND METHOD:

In this study 4 case of type 2 congenital tibial hemimelia in children who were 3-13 years old at time of presentation were treated using the Brown procedure by centralized medial fibular transport (without proximal fibular excision) and a second stage Syme's amputation.

All patients (n=4) in this study had were evaluated, assessed, treated & followed up during the period from Feb 2010 to Nov 2012.

All patients were male (male to female ratio is 4:0), their age ranges form 3-13 years (mean of 8.5 years), all had unilateral right sided involvement (Right: Left is 4:0) , only one patient has bilateral tibial hemimelia having the contralateral side with type 1 involvement (Table 1) and 3 of our patients were 2nd degree relatives.

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There were an associated deformities in three of our patients (Table 1); one patient with deformity of missing first and second rays with extra digit projected from midfoot, and missing of middle

finger in right hand, the other patient had left foot deformity in form of equinovarus, and the last patient had contralateral type 1 tibialhemimelia as mentioned. All patients had normal hips and lumbosacral spine.

Table 1: Information of patients included in this study with age, residence, side involvement, gender, & associated anomalies.

Case No.	Age/Year	Residence	Gender	Side involved	Type of hemimelia	Associated anomalies
1	3	Za'faranya Baghdad	Male	right	Type 2	non
2	7	Al-Ameen Baghdad	Male	right	Type 2	deformity of missing first and second rays with extra digit projected from midfoot,& missing of middle finger in right hand
3	13	Za'faranya Baghdad	Male	right	Type 2	left foot deformity in form of equinovarus
4	11	Kut	Male	right	Type 2	Type 1 Left tibialhemimelia with 1 st ray absence

Preoperatively and before starting treatment it was important that patient's parents understand the real situation as well as the possible future results and possible complications that may occur.

The distinction of whether a proximal portion of the tibia remains, and whether this portion is meaningfully powered by a quadriceps mechanism in our patients was made by direct palpation of the fragment and active extension of that fragment due to the older age of our patients, and this was further proved by imaging.

All the patients was clinically and radiologically evaluated when first seen; and were examined for the presence of other congenital abnormalities and Jones classification applied. The four legs were all type 2, the proximal tibia was present in all four limbs and formed normal articulation with the femur and active quadriceps contraction elicited.

Radiographs showed that the proximal third of the tibia was present in normal relation with the femur, but it was tapering out distally and the distal two thirds of the tibia were absent. The fibula was shorter than the normal fibula of the other leg, but it was well developed and proximally migrated in relation to the tibia.

METHOD:

Our treatment plan was transection of the proximal fibula at the level of the distal tip of the proximal tibial remnant and transfer of the distal fibula to fuse with the tibial remnant as described by Brown (Figure 5,A). Although it would seem preferable to wait until the proximal tibial anlage ossifies, Jones, Barnes, and Lloyd-Roberts reported that stability can be achieved even when the proximal tibia is purely cartilaginous.⁽²³⁾ We did not resect the proximal fibula in all of our patients.

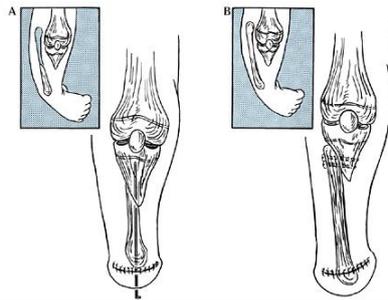


Figure 7: On the left (A): Transfer of the fibular into the tibial anlage in type 2 deformities of the tibia. on the right (B): Side-to-side fixation of the fibula into the tibia after resection of the upper portion of the fibula⁽²³⁾

At a second stage, after healing of the osteotomy site, the foot is amputated to make prosthetic rehabilitation easier. Retention of the foot during the proximal tibial reconstruction is helpful because it serves as a fixation point for the long leg POP cast that is applied after the first operation.

Operative techniques

Proximal Tibiofibular Synostosis

- Under general anesthesia, supine position, using above knee joint tourniquet
- Make an anterolateral incision beginning at the proximal tibia and extending distally and anteriorly to the middle third of the tibia.
- Dissect a sufficient portion of the anterior compartment musculature from the proximal medial tibia to expose the proximal tibial cartilaginous or the bony proximal tibia.
- Leave the proximal attachments of the fibula intact, but perform a subperiosteal dissection of the fibula.
- At an appropriate point opposite the distal end of the proximal tibial anlage, perform an osteotomy of the fibula. Then it was transferred in front of the interosseous membrane, behind the anterolateral group of muscles, to be in contact with the cut end of the tibia; leaving the fibular collateral ligament; the fibular head; the proximal physis and the periosteal sleeve intact.
- Drill a Steinmann pin of appropriate size distally through the medullary canal of the fibula out the plantar aspect of the foot about 1-2 cm proximal to tibial end.
- Reduce the fibula on the proximal tibia, and drive the medullary pin retrograde into the proximal tibial remnant.
- If necessary, pass the pin into the distal femur for stability.

- Distally, bend the pin 90 degrees, and cut it off below the level of the skin to be removed 6 to 8 weeks later. Immobilize the leg in a long cast.

Syme's amputation

As a definition the standard Syme's amputation consists of a bone section at the distal tibia and fibula 0.6 cm proximal to the periphery of the ankle joint and passing through the dome of the ankle centrally. The tough, durable skin of the heel flap provides normal weight bearing skin.

In our work, the Syme's amputation was a second stage surgery that was done after 3 months period in all our patients, after tibialization surgery, we preserve distal end of transferred fibula in order to maintain distal growth of distal fibula and prevent the complication of bone overgrowth from fibula.

After treatment

The POP cast was usually used postoperatively for several weeks until sound union occurs.

Each patient looked at two-weeks intervals for change of dressing and care for surgical incision.

All patients were frequently examined for the stump care after Syme's amputation and schedule for application of splint.

Afterward an orthosis worn to allow weight bearing, the latter is commenced after average of three months period.

The following Figures (6-24) are serial photographs of one of our patients from the day of surgery until about one year after operation, they show the stages of surgical techniques, x-rays, final limb appearance and the prosthetic fitting used.



Figure 6: Preoperative photograph of one of our patients showing an abnormal anterior tissue growth with deformed varus foot

Figure 7: Preoperative radiographs.



Figure 8: Skin preparation and draping.

Figure 9: Incision markings for resection of the anterior abnormal growing tissue.



Figure 10: Excision of the abnormal growing tissue.

Figure 11: The fibula osteotomized subperiosteally above the level of the tibia1 stump.



Figure 12: The fibula was transferred in front of the interosseous membrane, behind the anterolateral group of muscles

Figure 13: Leaving the fibular collateral ligament; the fibular head; the proximal physis and the periosteal sleeve intact.



Figure 14: The fibula is now in contact with the cut end of the tibia.
Figure 15: After reduction of the osteotomised fibula, which is attached to distal end of tibial anlage, a long smooth pin fixation used to hold the reduction in place.



Figure 16: Suturing of tissue planes.
Figure 17: Complete wound closure.

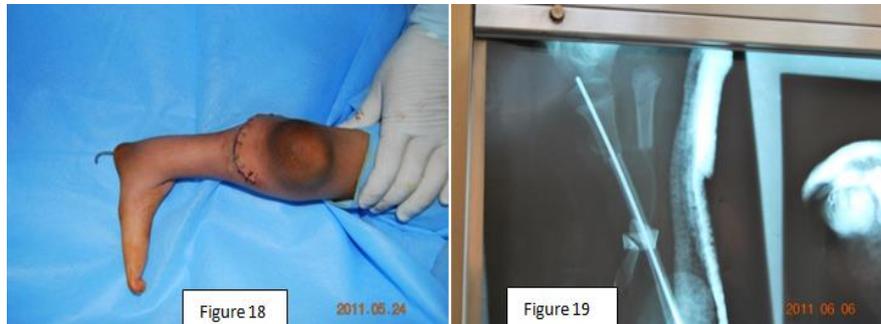


Figure 18: Postoperative photo.
Figure 19: Postoperative radiograph 14 days after operation lateral view.



Figure 20: Postoperative radiograph 14 days after operation AP view
Figure 21: 14 weeks after surgery.



Figure 22: 5 months after 1st. surgery and 2nd. Stage Syme's amputation.

Figure 23: 1 year after operation.



Figure 24: Same patient with prosthetic fitting.

RESULTS:

In all cases the transferred fibular shaft to the distal end of the tibial remnant was united within two to three months. The fibulae grew in length and the size to be tibialized and the leg increased in length as we preserved the distal fibular epiphysis.

Patient number 1: had good range of motion at ipsilateral knee joint, clean stump no local or systemic complications, prosthesis was fitted and at that time he walked independently.

Patient number 2: there was delayed weight bearing may be due to the associated skeletal deformities and the extra digit; he had operative amputation of extra digit then used the prosthesis for the type 2 tibial hemimelia side after tibialization and started independent weight bearing.

Patient number 3: had left foot deformity in form of equinovarus, on the right leg he developed 20° extension lag of the knee postoperatively and scheduled for physiotherapy and active quadriceps muscle exercise after application of prosthesis.

Patient number 4: who had type 2 tibial hemimelia of the right leg and type 1 tibial

hemimelia of the contralateral leg, regarding type 2 tibial hemimelia side he developed nonunion of the tibialization site and fibular re-union at the osteotomy site, had reoperation with fibular osteotomy and excision of 2 cm of proximal fibular segment with re-fixation of the distal segment with the tibia using rush nail with bone graft on the tibialization site.

DISCUSSION:

Owing to the rarity of this deformity (one per million live births¹), and because of the poor prognosis most of the cases of tibial hemimelia used to be neglected or treated with limb amputation. An attempt on treating one type (type 2) of this rare deformity making it even more difficult to find patients to treat and study in our country - Iraq.

This study on the surgical treatment of type 2 deformity may be the first in Iraq, we made a lot of efforts to have those four patients with this single type of a rare deformity to be included in this study during this time period, remembering that amputation used to be the usual treatment.

M.Nabil Khalifa and Nabil A.Ghaly (2004), treated eight children with type 2 tibial

hemimelia in the period between 1997 and 2003, they had five girls and three boys. Their average age when first seen was six months. They all had unilateral affection. Five of them were affected on the right side, three cases showed other associated abnormalities while one had a cleft palate; the other had contralateral congenital clubfoot and the last case showed polydactyl in the other foot.¹ we can notice the small number during six years work with a different age and gender distribution and they had 5 out of 8 but not all on the right side, associated anomalies are still common. Its nearly simillar study reflecting the rarity of the disease and the difficulties in its treatment.

Frantz in 1961⁷ and many others reported a higher male to female ratio in congenital tibial hemimelia those results are not suitable for comparison with our study as all our patients were males, it's clear that our sample is small and selective and it does not reflect the real prevalence of the disease in Iraq.

In our study we treated older children (mean age of 8.5 years) if compared with other studies. Brown(1972) redefined his indications to include infants younger than 1 year of age (preferably <6 months old) with the physical potential to walk, a functioning quadriceps mechanism, and full passive extension of the knee.⁽²²⁾

A multitude of skeletal and visceral abnormalities have been reported with most of patients with tibial deficiency,^(1,2) three out of our four patients had additional skeletal abnormalities, this goes with the results of most of the literatures.

Kalamchi in 1985 advised that the ideal treatment for the type 2 congenital deficiency of the tibia is by tibialization of the fibula by side to side technique to give stability to the knee joint and for the leg length discrepancy, and for the foot deformity is to do modified Boyd amputation, implanting the distal fibula within the body of the os-calcis. He also felt that below knee amputation should be avoided since it may lead to overwhelming difficulties with bony overgrowth and recurrent skin damage at the end of the stump.⁽²⁷⁾

Other study showed that the best treatment is tibio-fibular fusion in order to provide knee stability. Leg length discrepancy should be corrected after the transferred fibulas become well developed.⁽¹⁾

Although, most authors prefer a partial amputation of the foot using a modified Boyd technique implanting the distal fibula within the body of the calcaneus and avoiding a below knee

amputation because of recurrent skin damage at the end of the stump due to bony overgrowth, they used the Ilizarov frame to align the foot and get a stable corrected ankle for full weight bearing with a plantigrade foot as well as correcting the leg length discrepancy.⁽¹⁾

In this study we found that end to end fusion of the fibular transfer to tibial remnant gave good results regarding union rate, functional outcome and psychological acceptance of patient and family, we preserve the foot in the first procedure to use it as a fixation point for application of POP cast. In addition, Syme's amputation facilitates the rehabilitation program and early prosthetic fitting, the foot amputation was performed as a second stage after bone healing.

Proximal tibiofibular synostosis is not absolutely indicated for all type 2 deformities; the literature contains reports of satisfactory prosthetic rehabilitation after Syme's amputation alone; however, if the fibula is transferred under the tibial remnant, it can be expected to remodel reliably and form into a large, tibia-like bone eventually.⁽¹⁾

In type 2 deficiencies, the tibial remnant will ossify and form a satisfactory joint. In these cases, it is usually best to create a synostosis between the existing fibula and the tibial remnant to increase the length of the lever arm. A Syme's amputation is performed at the same time, and the patient is fit with below-knee prosthesis. When performing the synostosis, it is important to achieve good alignment of the fibula in relation to the knee joint.⁽¹⁾

In all of our four patients we preserved the proximal fibula in order to maintain knee stability, but we had one nonunion complication in one of our patients with re-union of the proximal fibular osteotomy that required revision operation with excision of 2 cm from the proximal fibula.

In type 2 cases, in which a tibial segment has been preserved or the fibula has been joined to the tibial remnant, below knee prosthesis is utilized. Unlike the standard transtibial design, the socket will incorporate supracondylar and suprapatellar proximal brim lines that will aid in the control and stability of the knee and prevention of a hyperextension moment, respectively. In some instances in which knee stability is less than optimal, outside joints and a thigh cuff or lacer may be required. These are used as a last resort and often contribute to increase weakening of the musculature as a trade-off for increased control and alignment.⁽¹⁾

CONCLUSION:

Our proposed surgical treatment seems to be a successful and useful choice for older children with type 2 tibial hemimelia, it provides a suitable reconstruction of the leg with acceptable knee function and stable construct for prosthetic fitting for those children who were used to be treated with amputation previously.

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