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Complex treatment of congenital pseudarthrosis of the tibia with periosteal grafting and intramedullary fixation

Kompleksowe leczenie wrodzonego stawu rzekomego goleni z zastosowaniem przeszczepu okostnej i stabilizacji śródszpikowej

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S u m m a r y

Introduction. In the last years to improve the outcomes of congenital pseudarthrosis of the tibia (CPT) treatment, new procedures were added to the standard: periosteal grafting, bone morphogenic protein (BMP) and bisphosphonate. In this study we analyze results of CPT treatment with periosteal graft combined with intramedullary nailing.

Materials and methods. Retrospective study of medical records and the radiographs of 7 patients treated because of CPT with resection of the hamartomatous periosteum, splitting of proximal tibial end or pseudarthrosis site resection, IM fixation, periosteal and cancellous bone grafting from the iliac bone was made. Mean age at the surgery was 5.6 years. Six patients had NF-1 and one patient – idiopathic CPT. LLD ranged 1.0-10.0 cm (average 4.7). The anteroposterior preoperative scan of all patients were studied for assessments of ankle joint mortise, where proximal migration of the fibula was evaluated according to Malhotra scale for fibular shortening. The patients were classified according to El-Rosasy-Paley classification: 2 patients as type I, and 5 patients as type II. All of them were type IIC according to Crawford classification. The average follow-up after the index operation was 3.1 years (range 2.1-4.6).

Results. Primary pseudarthrosis union of the tibia was achieved in 6 patients, at a mean time of four and half months (range 4-6 months). Failure to obtain bone union after 13 months in youngest patient needs reoperation with the same technique.

Conclusions. Primary union of CPT in most of the cases can be obtained and maintained with limited pseudarthrosis resection, periosteal and cancellous bone grafting and intramedullary rod fixation.

Key words: congenital pseudarthrosis of the tibia, neurofibromatosis, operative treatment, intramedullary fixation, periosteal grafting

S t r e s z c z e n i e

Wstęp. W ostatnich latach w leczeniu wrodzonego stawu rzekomego piszczeli (WSRP) pojawiły się nowe metody, takie jak zastosowanie przeszczepu okostnej, białek morfogenicznych (BMP) czy bifosfonianów. W naszym ośrodku od 2006 r. wprowadziliśmy technikę operacyjną złożoną z: ograniczonej resekcji stawu rzekomego z szerokim usunięciem zmienionej okostnej, stabilizacji śródszpikowej oraz obłożeniem przeszczepami z kości gąbczastej i okostnej pobranymi z talerza kości biodrowej.

Materiał i metody. Materiał obejmuje 7 pacjentów ze średnią wieku 5,6 lat. 6 chorych prezentowało objawy neurofibromatozy typu I. Według klasyfikacji El-Rosasy-Paley typ I WSRP występował u 2 chorych, typ II u 5. Według klasyfikacji Crawford wszyscy należeli do typu IIC. Konfiguracja stawu skokowo-goleniowego była oceniona na radiogramach w projekcji a-p, a do oceny proksymalnej migracji strzałki zastosowano skalę Malhotra skrócenia strzałki. Skrócenie kończyny wahało się od 1 do 10 cm (średnia 4,7). Średnia okresu obserwacji wynosiła 3 lata i 1 miesiąc (od 2,1 do 4,6).

Wyniki. Pierwotny zrost WSRP uzyskano u 6 chorych w okresie od 4 do 6 miesięcy (średnia 4,5). U najmłodszego chorego brak zrostu po 13 miesiącach wymagał reoperacji z zastosowaniem tej samej techniki.

Wnioski. Wygojenie wrodzonego stawu rzekomego piszczeli może być uzyskane i utrzymane po zastosowaniu kompleksowego leczenia złożonego z ograniczonej resekcji kostnej, szerokiej resekcji patologicznej okostnej, stabilizacji śródszpikowej piszczeli i strzałki połączonej z przeszczepami z kości gąbczastej i okostnej.

Słowa kluczowe: wrodzony staw rzekomy piszczeli, neurofibromatoza, leczenie operacyjne, stabilizacja śródszpikowa, przeszczep okostnej

INTRODUCTION

Congenital pseudarthrosis of the tibia (CPT) remains one of the most challenging pediatric problems in management and prognosis. It is uncommon disorder where the incidence is 1 per 140 000 to 190 000 patients. The diagnosis usually is made early under two years of age, while very rarely occur in older than eleven years of age. CPT has heterogeneous clinical picture. It may present as established pseudarthrosis in early age or only anterolateral bowing of the tibia that can fracture spontaneously and presents picture of real pseudarthrosis. The same situation may be initiated by osteotomy at the bowing site (1).

The precise etiology is still unclear, although 50% to 55% of patients with CPT have neurofibromatosis-1 (NF-1), but CPT may develop even without symptoms of NF-1. The cause may be neurofibroma grows within bone cortex leads to fracture and interference of bone union although this theory is very rarely confirmed histologically (1) except cases of Green and Rudo (2). They concluded that in true NF-1 cases, the Schwann and fibroblast cells should be seen in soft tissue but in electronic microscope, usually only fibroblast was seen in the pseudarthrosis site.

Although the main pathology in CPT is still unknown, the recent studies showed that the basic pathology is hyperplasia of fibroblast and formation of dense fibrous tissue. This invasive fibromatosis is located in the periosteum and between the bony ends surrounding the tibia which causes compression, osteolysis and persistence of pseudarthrosis (3-7).

According to Hermanns-Sachweh et al (8), the most striking finding was thickened periosteum with accumulation of nerve cells surrounding small arteries, causing subtotal or complete obliteration and local hypoxia which interfere the osteogenesis effect of the periosteum. It results in impairments of vascularization, which leads to decreasing of osteogenic capabilities. As the basic pathology in CPT is located in surrounding soft tissue while the bone is involved secondarily, Aegerter speculated that if all tumour tissues were removed, the normal callus would form (4).

The aims of CPT treatment are to achieve and to maintain the union until the skeletal maturity and to obtain the functional lower extremity. Difficulties to achieve these goals, regardless to surgical technique were mainly biological in nature (due to poor healing ability of the dysplastic bone segment) or mechanical (technical difficulty to fix the short osteopenic bone segment without transfixing the joint).

The modern treatment option includes pseudarthrosis site resection, shortening, grafting, intramedullary fixation or rodding in addition to reconstruction procedures with Ilizarov devices or microvascular bone transfer (9).

All these operative techniques, including the Ilizarov method which is considered as a choice of treatment of CPT, don't fully guarantee the successful treatment of pseudarthrosis.

To improve the treatment outcomes, in the past years, new procedures were added as the standard: periosteal grafting, bone morphogenetic protein (BMP) and bisphosphonate (10).

Recently, the periosteal grafting as the CPT treatment was described by El-Rosasy et al (6) and Weber (7). This technique depends on to the type of the pathology and includes: resection of the hamartomatous periosteum, splitting of proximal tibial end or pseudarthrosis site resection, intramedullary fixation, periosteal grafting from the iliac bone and Ilizarov frame fixation. Since 2006, we have used the similar technique without external fixation.

The aim of this study is to evaluate our method's results and the advantages, including the primary union after the index operation without any supplementary procedures.

MATERIALS AND METHODS

Retrospective study was made on seven CPT patients treated with the method described above between 2006 and 2009 in our center. The analysis was done on medical records and radiographies. The mean age at the surgery was 5.6 years (range 2.7-9.2). Four patients were boys and three were girls. The left side was involved in four patients and the right in three patients (tab. 1). Five patients had established fibular pseudarthrosis while in two patients the fibula was intact. Six patients had NF-1 and one patient had idiopathic CPT.

Only two patients had no previous surgery before the described index operation, while the other five patients had been treated unsuccessfully with several

Table 1. Patient's history.

Case	Sex	Age (yr.)	Side	NF-1	El-Rosasy type	Crawford type	No. of previous surgeries	LLD (cm)	Site of CPT	Fibular CP
1	M	6.1	L	Yes	II	IIC	8	7	Distal	Yes
2	F	8.2	L	No	II	IIC	5	10	Distal	Yes
3	M	5.4	R	Yes	II	IIC	2	4	Distal	Yes
4	M	9.2	L	Yes	II	IIC	4	3	Distal	Yes
5	F	2.7	R	Yes	I	IIC	0	1	Distal	Yes
6	F	3.5	L	Yes	I	IIC	0	4	Middle	No
7	M	4.1	R	Yes	I	IIC	1	4	Distal	No

surgical operations (4 to 8 times). The eldest two patients (case 1 and 2) underwent different procedures elsewhere, including: plate and screws fixation, nailing with bone graft, Ilizarov device application (twice in the case 1). Intramedullary fixation with autogenous bone graft has been used four times in one patient (case 4) and two times in another (case 3), curettage and frozen bone graft had been used in the other one (case 7). In all patients, the pseudarthrosis was associated with limb length discrepancy ranges from 1.0 to 10.0 cm (average 4.7).

The anterioposterior preoperative scan of all patients were studied for assessments of ankle joint mortise, where proximal migration of the fibula was evaluated according to Malhotra scale for fibular shortening (11).

Three patients were classified as "mild fibular migration" where the distal fibular growth plate lies between the top of the talar and distal tibial growth plate, another one had "moderate" where the fibular growth plate at the level of distal tibial growth plate and the third one had "severe migration" where the fibula growth plate proximal to the distal tibial growth plate. Only two patients had normal position of the lateral malleolus before the index operation where the distal fibular growth plate is at the level of the talar plateau. Wedging of the distal tibial epiphysis and valgus deformity of hindfoot were noted in four patients (tab. 2).

Patients were classified with their radiological and clinical presentations according to El-Rosasy-Paley classification (6). Two patients were type I (fig. 1), and five other patients were type II (fig. 2) and none of them were type III. We use this classification because it is the only classification that serves as the therapeutic guideline and provides prognostic indicators. Additionally, the patients were radiographically classified with the Crawford classification (1) and all of them were classified as type IIC. The average follow-up time after the index operation was 3.1 years (range 2.1- 4.6 years). None of the patients reached the skeletal maturity at the follow-up.

The surgical procedure consisted of wide resection of the thickened periosteum, which was dissected from the surrounding soft tissue circumferentially at the level of pseudarthrosis site. Resection of bony ends at the pseudarthrosis site to maximize the cross section of the bone, allowing realignment and reopening of the medullary canal, limited fibular resection by separate incision. Tibia intramedullary fixation with two Rush pins after anatomical alignment of the pseudarthrosis site were used in six patients, the first pin inserted from the knee and the other from the heel with the foot in neutral position (fig. 3a-3f).

Fig. 1. Case 3 – El-Rosasy-Paley Type I CPT.

The operative technique was the same on all patients, except the El-Rossasy-Paley type II cases. In these patients, internal fixations were firstly removed (locked intramedullary nail in 2 patients and Rush pins in other 2).

In one case, the tibial fixation of single Rush rod was inserted from the heel because of knee joint adhesion and the range of motion limitation. The knee joint fixed flexion contracture needs to be released additionally with the Gravitational Platelet Separation System (Biomet).

Intramedullary fixation of the fibula with Kirschner wire was done only in three patients. The fibular ends of the other four patients were tapered and very narrow,

Table 2. Pathology of the fibula and ankle joint.

Before index operation					After index operation				
Case	**Fibula CP**	**Fibular migration**	**Physis wedging**	**Ankle valgus**	**Fibular migration**	**Physis wedging**	**Ankle valgus**	**Fibula fixation**	**Fibular union**
1	Yes	Mild	Yes	Yes	Moderate	Yes	Yes	No	No
2	Yes	Moderate	Closed	Yes	Sever	Closed	Yes	No	No
3	Yes	No	No	Yes	No	Yes	No	No	Yes
4	Yes	Mild	Yes	Yes	Mild	Yes	Yes	No	No
5	Yes	Sever	Yes	No	Sever	Yes	Yes	No	No
6	No	No	No	No	No	No	No	Yes	Yes
7	No	Mild	No	No	Mild	Yes	No	Yes	Yes

Fig. 2. Case 2 – El-Rosasy-Paley type II CPT.

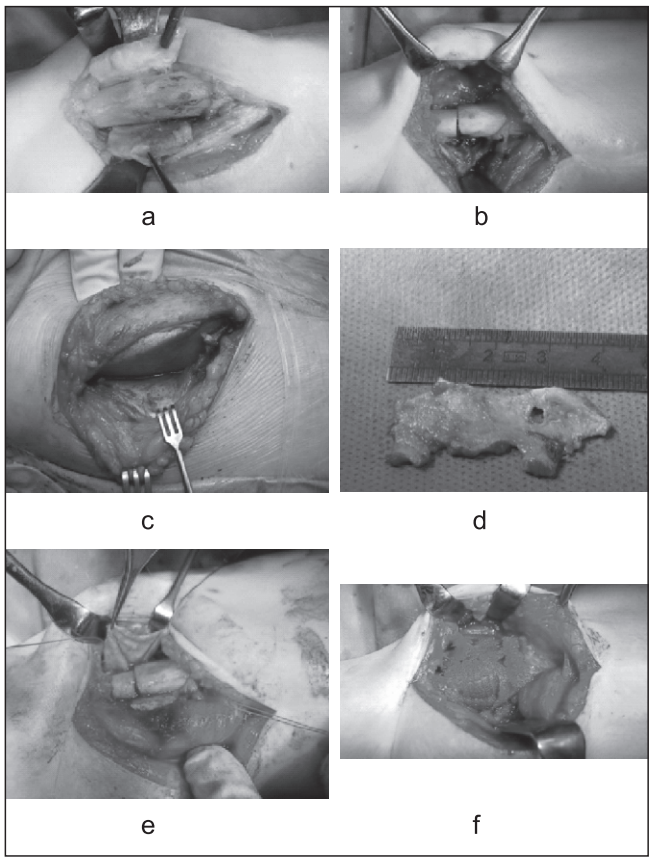

Fig. 3. Periosteal and pseudarthrosis resection (a, b); periosteal graft from the external surface of the iliac bone (c, d); pseudarthsosis site enveloped by periosteal and cancellous graft (e, f).

so the insertion of the Kirschner wire was impossible (fig. 4). Cancellous bone graft from the external surface of the iliac bone was placed around the pseudarthrosis site of the tibia and enveloped by free periosteal graft, which was harvested from the external side of the iliac bone. The average of tibial resection was 3.0 cm (range 1.5-4.0).

Fig. 4. Tibia and fibula fixation with intramedullary rods.

After the surgery, the operated limb was immobilized in above-knee posterior slab, which was changed after 2-3 weeks to a complete long cast and weight bearing was allowed. The cast was used till the bone union was observed. Usually, after four to six months, the immobilization was finished and the limb was protected with ankle-foot orthosis and walking with full weight bearing was continued.

The patients had radiological examination every six weeks. Both anterioposterior and lateral radiographs were evaluated for maintains of bone segments axis at the pseudarthrosis site and for bone union signs.

The last follow-up focused on the union of the tibia and fibula, migration of lateral malleolus, distal epiphysis of the tibia wedging, the valgus deformity of the ankle joint and the limb length discrepancy (LLD).

RESULTS

The primary union of tibial pseudarthrosis union was achieved in six patients, at a mean time of 4.5 months (range 4-6) (fig. 5a-5d). Failure of obtaining the bone union was observed in the youngest patient after 13 months. The reoperation with the same technique is needed (fig. 6 a-e).

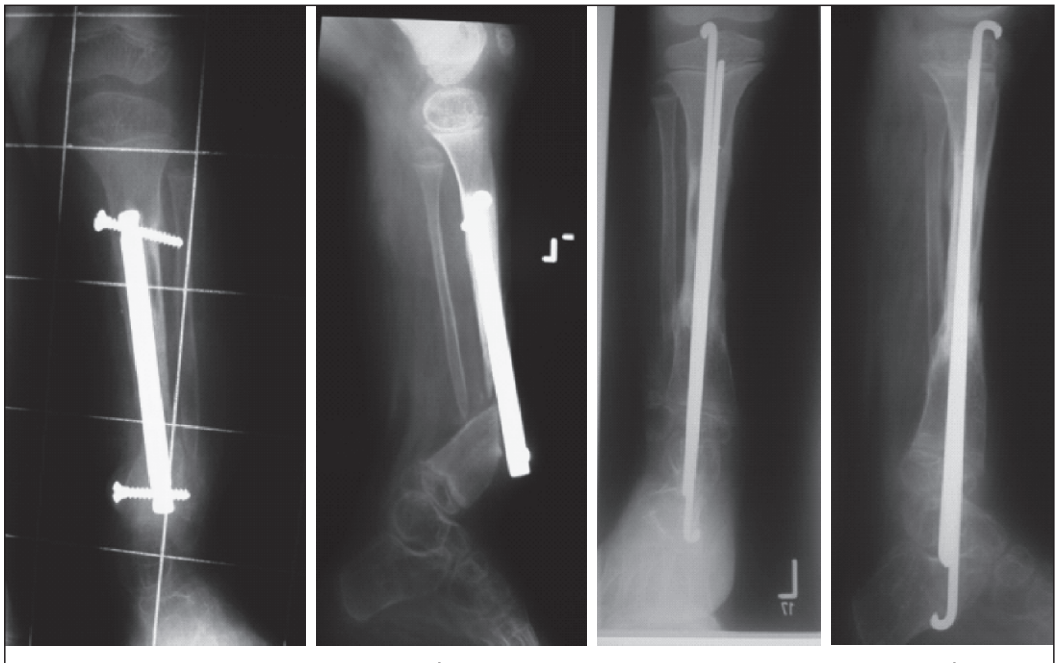

Fig. 5. Case 1. Type II CPT before index operation (a, b). Pseudarthrosis primary union (c, d).

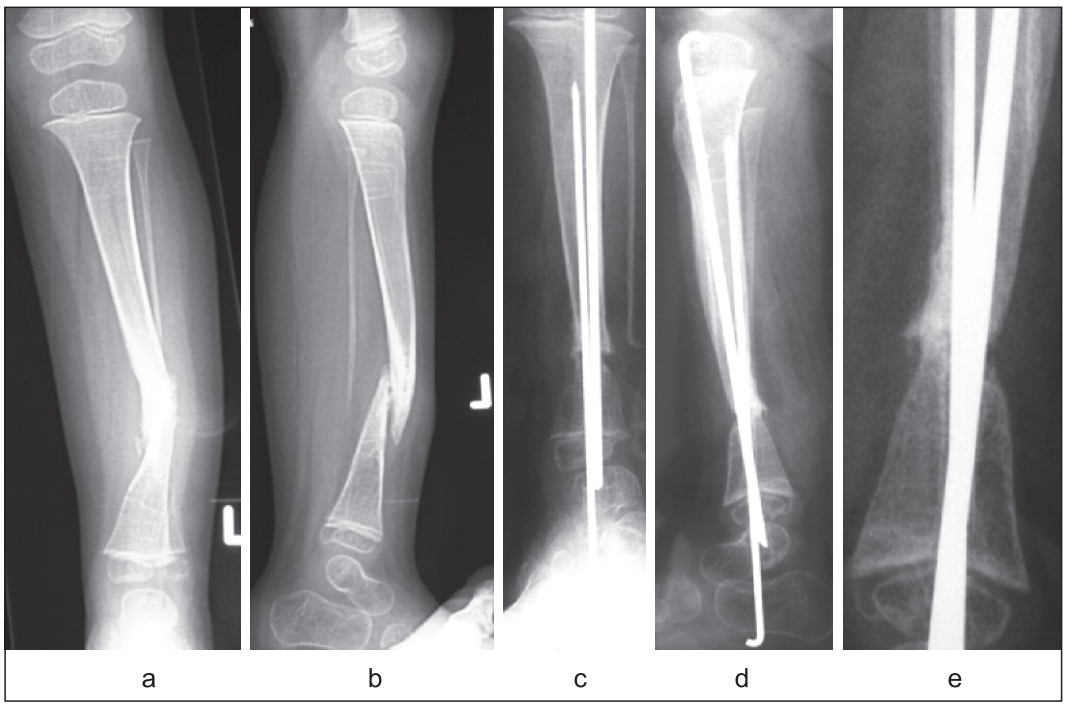

Fig. 6. Case 2. Type II CPT before index operation (a, b). 12 months after index operation no union (c, d, e).

Fibular union was observed only in three patients. Two of them had no fibular pseudarthrosis preoperatively (after osteotomy). In all of them, the fibulas were transfixed with Kirschner wire. The fibula in last follow-up remains non-union in the other four patients. Their fibulas were not fixed at the index operation. No fracture was noticed after the primary consolidation in our patient till last follow-up.

In one patient, the proximal Rush rod shifted proximally to the knee and was removed after two years from the index operation.

The ankle joints of all patients remained transfixed with the distal rod, which hooked in the plantar surface of the calcaneus in spite of tibial growth. The anteroposterior radiographs showed the wedging of distal tibial epiphyses in six patients. According to Malhotra classification, the severe migration of the lateral malleolus was observed in two patients, moderate in one, mild migration in two patients and two patients shows normal level of the lateral malleolus as preoperatively (tab. 2). Ankle joint valgus was revealed in four patients while the other three patients their joints stayed in the

neutral position. Bony ankylosis of the ankle joint developed in one patient in whom before the index operation moderate fibular migration, wedging of distal tibial epiphysis and ankle valgus were observed.

The average LLD is 5.0 cm (range from 0 to 12.5). The patient with 12.5 cm LLD has complete distal tibial growth plate closure, 10 cm shortening before the index operation needing prosthesis. Two patients with 7 and 8 cm LLD waiting for lengthening with Ilizarov method, the other two patients with LLD 1.0 and 1.5 cm treated with shoe lifting. In the patient with nonunion after the index operation, the limb shortening is 5 cm.

DISCUSSION

The treatment of CPT remains controversial and none of the treatment method shows clear superiority in obtaining and maintaining bone union (6, 12-14). Until recently, the outcome of any methods is still unsatisfactory. Both patients and surgeons are disappointed and frustrated by repeated failed operations, refractures, prolonged immobilization, poor function of the limb or even amputation (9, 15).

The primary treatment goal is to obtain and maintain union until the skeletal maturity in order to assure a functional limb. LLD and axis deformities should also be taken into consideration in CPT managements (6, 10, 13, 14).

In our study, the unequivocal primary union was achieved in six patients (85.7%) which is higher than other reports. Johnston (14) used Charnley-Williams procedure (without periosteal grafting) and reported successful union in 11 of 23 patients (47.8%). In other two reports used intramedullary rod, the primary union was obtained in 18 of 23 patients (78.3%) (13) and 4 of 12 patients (33.3%) (16). Joseph and Mathew (12) used intramedullary rod and cortical graft reported the primary union in 21 of 26 patients (80.7%). Recently, Nguyen (18) reported the primary union in 16 of 21 patients (76.2%) by using an intramedullary Kirschner wire for tibial stabilization. The rate of union did not improved using BMP with rodding, where primary union achieved in 4 of 5 patients (80%) (19).

The overall primary union rate with the Ilizarov methods were 82 of 108 patients (75.5%) in multicenter EPOS study (20), and was 14 of 21 patients (66,7%) in Boero et al study (21). Paley et al. (22) reported the primary union in 15 of 16 limbs (93.8%). Thabet et al. (10) reported 100% primary union after index operation in series of 20 patients using combined approach including free periosteal grafting, bone grafting, IM nailing of both the tibia and fibula, and Ilizarov fixation. They recommended this strategy as the best combination of biological and mechanical stimulation of healing. This combined method looks to be the most promising solution up to date since it guarantees obtaining and maintaining union in addition correcting the associated axial deformity and LLD.

LLD is frequently associated with CPT as a result of primary bone segments resorption or resection of pseudarthrosis at the operation. Our patients with extensive shortening before index operation refused to have Ilizarov frame as they have been treated by this method before without achieving union. For this reason, we chose the two-stage approach. First stage, to obtain CPT union with our protocol of treatment. In the second stage, the limb lengthening is performed.

The Ilizarov method increases our armamentarium in the CPT management. However, in most of cases, the basic pathophysiology of CPT continues unaltered even after healing and associated with high rate of refracture and complication (6, 15, 21, 23).

After achieving primary union, we did not have any refracture probably because of maintaining of longitudinal axis, internal protection with IM fixation and wide bone section at the pseudarthrosis site. Mathieu et al. (24) reported the association of persistence of the fibular pseudarthrosis and tibia refracture. Four patients in our study had no fibular union but no one of them develops tibia refracture as reported by Paley et al. (22). This patient also shows the ankle joint valgus probably due to proximal migration of the fibula and wedging of distal tibial epiphysis, which also described by many authors (12, 17, 20, 22, 24, 25). Dobbs et al. (13) speculated that valgus deformity of the ankle is the natural outcome of initial deformity, not iatrogenic complication of the distal tibia epiphysis damage with centrally positioned rod. Because of this, they recommended the placement of syndesmosis screw once the deformity was corrected. In most of our patients, the valgus deformity occurred before the index operation and sustained with non-union fibula.

Our method of intramedullary fixation with two Rush rods inserted through the proximal epiphysis and the calcaneus gives suitable longitudinal fixation, relative rotational stability and behaves as telescope with growth. However, it penetrate the growth plate of the tibia that may anticipate the longitudinal growth but its effect is still not clear, especially in our two patients whose lower limb lengths were almost equal after the CPT union.

The Rush rod crossed ankle and subtalar joint in all our patients as the CPT site is located in the distal part of the tibia in most of them. This makes the fixation of the distal short segment difficult without crossing the ankle joint.

The disadvantage of this type of fixation is ankle joint stiffness as reported by many authors (13, 15, 16, 20, 24). Although Dobbs et al. (13) reported that no negative effect in long-term functional results if the trans-ankle fixation is removed after 2 years. Rods removing to regain the ankle motion may leads to refracture of pseudarthrosis site. To avoid this complication, using the interlocking nails or introducing the rods from the medial malleolus is recommended by El-Rosasy et al. (6).

CONCLUSIONS

The primary union of CPT can be obtained and maintained with limited pseudarthrosis site resection, periosteal and cancellous bone grafting and intramedullary rod fixation.

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