

Combined Ilizarov Fixation and Intramedullary Nailing for Treatment of Congenital Pseudarthrosis of the Tibia

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Background: Congenital pseudarthrosis of the tibia (CPT) is one of the most challenging conditions in pediatric orthopedics. Multiple objectives must be achieved to correct this condition successfully, including obtaining union, prevention of refracture, management of limb-length discrepancy (LLD), and correction of deformities of the ankle and leg. This combined treatment conjoins the improved union rate and improved limb-length equalization of Ilizarov fixation and the controlled alignment and refracture prevention associated within intramedullary nailing.

Objective: To evaluate the outcome of combined Ilizarov fixation with intramedullary pinning in terms of union of pseudarthrosis and residual deformities.

Material and Method: Thirteen children who were treated by this technique were evaluated in this study. Clinical presentation and radiographs were evaluated and union time, secondary operations and time of follow-up were recorded. At final follow-up, condition of tibial and fibular pseudarthrosis, LLD, refracture and deformities of the tibia and ankle were documented.

Results: Union was achieved in 11 children. For the other 2 children, a secondary procedure using iliac bone graft was required to achieve union. Fibular pseudarthrosis persisted in 9 patients. A total of 30 secondary operations were performed for various indications. At final follow-up, mean anterior bowing was 8.5 degrees (range: 0-20°), mean medial bowing was 9.5 degrees (range: 0-25°), and ankle valgus was present in 5 patients. The mean LLD was 1.7 cm (range: 0-3). Two patients had limping gait. No donor site morbidity was observed in any patient. Refracture occurred in three cases after nail removal. In these three cases, secondary operations using the same technique were performed with favorable outcome.

Conclusion: The results of this study demonstrate that Ilizarov fixation combined with intramedullary nailing is a safe, effective and practical treatment for management of CPT. This combination technique achieves multiple objectives including rigid-stable fixation, management of LLD, correction of residual deformity and prevention of refracture. Moreover, this technique can be used in cases with previous operations that resulted in nonunion.

Keywords: Congenital pseudarthrosis, Tibia, Ilizarov technique, Intramedullary nailing, Fibular pseudarthrosis, Refracture

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One of the most difficult problems in pediatric orthopedics is congenital pseudarthrosis of the tibia (CPT). Neurofibromatosis-1 appears to be related to this condition, but the exact cause of CPT is unclear⁽¹⁻³⁾. Achieving union, correcting deformities of the ankle and leg, prevention of refracture and management of leg-length discrepancy (LLD) are the key objectives of treatment. Ilizarov fixation has been shown to be effective in the treatment of CPT⁽⁴⁾. In a multicenter study of 120 published cases of CPT that were treated by Ilizarov fixation, the European Pediatric Orthopedic Society (EPOS) reported healing in 94 pseudarthrosis patients with satisfactory results⁽⁵⁾. The

Ilizarov method concomitantly facilitates correction of complex deformity and treatment of LLD^(1,4,7,8). However, a high rate of associated complications like residual axial deformity and refracture has been reported⁽⁹⁾.

Refractures, growth disturbance, and stiffness that cause poor foot and ankle function, are frequent residual problems^(10,11). Re-emergence of pseudarthrosis after refracture is the most common and most serious complication after primary healing^(12,13). As such, a safe, effective and practical management method that minimizes complications after healing is needed to achieve the multiple goals of treatment.

The most commonly reported problem is lack of protection against refracture after Ilizarov removal⁽¹²⁾. To mitigate this problem, we combined Ilizarov fixation with intramedullary nailing of the tibia. This combined treatment conjoins the improved union rate and improved limb-length equalization of Ilizarov fixation and the controlled alignment and refracture prevention

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associated with intramedullary nailing. The objective of this study was to report the clinical and radiological results of this combined technique for the treatment of CPT.

Material and Method

Thirteen pediatric patients were treated for CPT using the combined technique of Ilizarov fixation with intramedullary nailing at the Department of Orthopedic Surgery, Faculty of Medicine, Siriraj Hospital, Mahidol University during the 1996 to 2005 study period. Our study population consisted of 7 girls and 6 boys, with age range of 1 to 14 years (mean 8 years). Patients with pseudarthrosis found to be associated with neurofibromatosis were also evaluated by a pediatric geneticist and a pediatric ophthalmologist.

Surgical procedure

Fibrous tissue at the pseudarthrosis site was excised. The margin of excision was measured pre-operatively and confirmed intra-operatively by bleeding at the osteotomy cut under direct visualization. Bone end preparation was performed at the pseudarthrosis site, followed by intramedullary nailing with titanium elastic nails (TEN) (De Puy Synthes, West Chester, PA, USA). In cases where the pseudarthrosis was located at the very distal part of the tibia (less than 4 cm from the tibial plafond) nail insertion through the talus and calcaneus was necessary to achieve stable fixation.

The Ilizarov apparatus was then inserted and

assembled, with two whole rings above the non-union site and one whole ring below, with compression of the bone end in cases where the resection of the pseudarthrosis did not exceed 3 cm.

Axial correction, end-to-end compression and lengthening via proximal tibial metaphyseal osteotomy were all performed during the same operation (Fig. 1B). In cases where resection of the pseudarthrosis site was more than 3 cm. Ilizarov bone transport with simultaneous intramedullary nailing was performed.

At 1 week after surgery, lengthening adjustments were performed by parents until equal leg length was achieved. Distraction speed was 1 mm/day. When lengthening site union and leg length became equal, the Ilizarov apparatus was removed but the titanium elastic nails were kept in place. A patellar tendon-bearing (PTB) brace was then applied to protect against refracture until skeletal maturity (Fig. 1A-C). Intramedullary pinning was left in place until skeletal maturity to minimize the risk of refracture. If radiograph showed evidence of rod migration that jeopardized stabilization of the tibia, then revision of IM pinning was performed.

Radiographs were taken at 6-week intervals to assess and evaluate bone union and consolidation of the lengthening site. 'Primary union' referred to bone union at the pseudarthrosis site and 'secondary union' referred to additional surgical procedures required to create bone union.

Secondary procedures included pseudarthrosis resection, preparation of bone edge and iliac

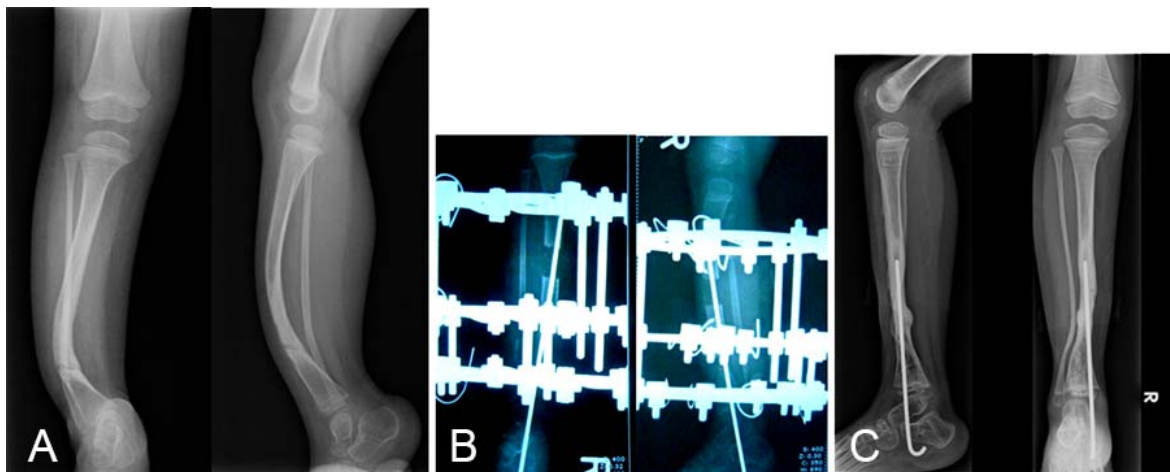


Fig. 1 (A) Preoperative radiograph of a 4-year-old girl with pseudarthrosis right tibia, (B) Postoperative radiograph showing combined Ilizarov and intramedullary nailing using titanium elastic nail (approximately 4 cm of bone lengthening was achieved), (C) AP and lateral radiographic at 2-year follow-up showed healing of pseudarthrosis with good AP and Lateral alignment.

bone graft. At final follow-up, 13 patients were evaluated for LLD, residual deformities, painful tibia and fibula pseudarthrosis status.

Results

Neurofibromatosis was associated with CPT in 5 cases. All patients were atrophic-type according to Crawford's classification⁽¹⁴⁾ and 12 cases had fibula pseudarthrosis. All of the nonunion sites were located at the distal third of the tibia. Four patients had multiple surgical procedures with persistent nonunion. Demographic and clinical data of the 13 patients evaluated in this study are summarized in Table 1. Mean age at presentation was 6.6 years (range: 11 months to 14 years). Mean age at surgery was 7 years (range: 1-14). Mean follow-up was 5.2 years (range: 2-11).

Bone union was achieved in 11 of 13 cases of CPT with a mean time to union of 11.7 months (range: 5-22). The other 2 cases achieved union after a secondary procedure using iliac bone graft. Lengthening ranged from 3 to 7 cm. Angular deformity was fully corrected in 4 cases. In the remaining 9 cases, mean anterior bowing was 8.5 degrees (range: 10-20) and mean medial bowing was 9.5 degrees (range: 0-25). Mean ankle valgus was 15 degrees (range: 0-20) (Table 2). Pseudarthrosis of the fibula showed nonunion in 9 cases at final follow-up.

Three patients had refracture due to rod migration when leg-length increased and all of those refractures occurred at the original level of the pseudarthrosis. All cases of refracture were treated by the same combined technique (Ilizarov and intramedullary nailing), with bone union being achieved in every case. All refractures healed in 3 to 6 months.

Thirty secondary operations were performed for various indications, as follows; refracture (n = 7), revision of rod due to bone growth (n = 7), correction of deformity (n = 7), management of LLD (n = 7), and repositioning of migrating rod (n = 2).

There were no intra-operative complications and no donor site morbidity at the iliac crest in patients requiring bone graft. Pin tract infections were successfully treated by oral antibiotic and local wound care. Mean limb-length discrepancy was 1.7 cm (range: 0-3).

At final follow-up, all patients had achieved union of the tibia and could walk with full weight-bearing on the affected limb. All patients were prescribed patellar tendon-bearing (PTB) brace until skeletal maturity. Although none reported having any ankle pain, ankle stiffness was reported 3 patients with

degenerative changes of the ankle joint observed from radiographs. No patients reported ankle instability and all cases had normal knee function.

Discussion

Achieving union, preventing refracture, correcting residual deformity, angulation, and limb-length discrepancy are the key treatment objectives in the management of congenital pseudarthrosis of the tibia^(12,15).

Ilizarov fixation has been shown to be effective in the treatment of CPT⁽⁴⁾. Adding intramedullary nailing to Ilizarov fixation as a combined treatment would improve prevention of refracture.

Ilizarov and Gracheva reported 16 cases of pseudarthrosis union without grafting, but 5 refractures occurred resulting from residual deformities of the tibia⁽¹⁶⁾. From a multicenter retrospective study by Paley et al tibial union was achieved in 15 of 16 cases with 5 cases of refracture⁽⁴⁾. Ghanem et al reported 10 of 14 patients successfully treated with Ilizarov technique, with 3 of 10 patients achieving bone union after bone graft procedure and refracture occurring in 1 patient⁽⁷⁾. Our results are comparable to those of other studies regarding rates of bone union, refracture, limb-length discrepancy and residual deformities (Table 2). Factors in refracture prevention include; (1) angular deformities correction that can be achieved by Ilizarov fixation combined with intramedullary nailing; (2) achievement of fibular union; (3) prevention of refracture by intramedullary nailing and external orthosis. Stress on a dystrophic bone due to residual deformities after healing of the pseudarthrosis can result in refracture. Ilizarov and Gracheva recommended the correction of residual angular deformities to decrease risk of refracture⁽¹⁶⁾.

Fibular pseudarthrosis can influence union of the pseudarthrosis tibia, refracture rate, and ankle valgus deformity. Cho et al reported that the tibial refracture rate was higher when the fibula remained ununited^(12,17). Dobbs et al reported that concomitant fibular pseudarthrosis occurred with ankle valgus deformities in most patients in their long-term follow-up study, even after the fibular lesion had been treated⁽¹⁸⁾. Cho et al reported that fibular stabilization to create union has significantly better refracture-free survival^(12,17). Cho et al also reported that maximization of cross-sectional diameter at the healing level of the pseudarthrosis can safeguard against refracture. This is explained by the fact that wide cross-sectional healing is effective in resisting mechanical stresses. Cho

Table 1. Demographic and clinical data of 13 patients with congenital pseudarthrosis of tibia

Case	Gender	Side	Evidence of neurofibromatosis	Fibula pseudarthrosis surgeries	Priorsurgery/ No. of pseudarthrosis surgeries	Age at presentation (yr)	Age at index surgery (yr)	Age at final follow-up (yr)	Time to union (mo)	Refracture	Age at refracture (yr)	LLD (cm)	Anterior bowing (degree)	Medial bow (degree)
1	M	R	+	+	-	1	4	7 yr 8 mo	12	-	-	1	-	10
2	M	R	-	+	-	11 mo	1	9	18	-	-	1	-	25
3	M	R	-	+	-	8	8 yr 6 mo	11 yr 1 mo	10	-	-	2	10	-
4	M	R	+	-	-	9 yr 11 mo	10 yr 3 mo	17	5	+	11	3	20	25
5	M	L	+	+	-	4	5	14	8	+	8	2	15	15
6	F	L	+	+	-	+/IM nailing 2	2 yr 6 mo	6	7	-	-	1	20	-
7	F	R	-	+	-	14	14	17	20	-	-	1	10	10
8	F	R	-	+	-	13	13 yr 5 mo	16	5	+	14	1	10	10
9	F	R	+	+	-	7	7	16	22	-	-	0	-	-
10	F	L	-	+	-	6	6	9	10	-	-	0	-	-
11	F	L	-	+	-	4	4	8	10	-	-	1	5	5
12	M	R	+	+	-	5	6	8	8	-	-	1	5	10
13	M	R	+	+	-	5	8	12	8	-	-	0	5	10

Table 2. A review of the literature (including this study) for demographic, clinical, and outcome data in patients with congenital pseudarthrosis of tibia

References, No. of treatment patients method	Age at final follow-up (yr)	Follow-up period (yr)	Primary union	Sound union at final follow-up	Frequency of amputation	Refracture		Ankle valgus Frequency (degree)	Mean limb-length difference, (range)(cm)	Tibial deformity mean, (range) (degree)	
						No. of patients	Severity (degree)			Anterior bowing	Valgus
Grill et al ⁽⁷⁾ (IL)	>16	-	-	22	3	-	15	-	-	-	-
Shah H ⁽¹⁾ (IMN/BG)	18.4 (16-21.6)	15.2 (12.8-17.4)	9	11	0	5	6	15	2.6 (0-8)	15 (0-43)	8 (0-28)
Nguyen Ngoc ⁽⁹⁾	-	6.4	16	16	0	17	17	9 (5-25)	3.5 (2.0-5.5)	-	-
Hung (IP)	- ¹²	4 (2-7)	15	16	0	16	3	1	4 (1-8)	-	-
Dror Paley ⁽¹⁾ (IL)	-	4 (1.2-6.6)	9	9	0	9	1	3	-	-	-
Mathieu L ⁽¹⁰⁾ (IMN+IL)	(5-16)	5	8	10	0	3	3	5	1.2(0-3)	8.5(0-20)	9.5(0-25)
This study (IL+IMN)											

IL = Ilizarov; IMN/BG = intramedullary nailing and bone grafting; VFG = vascularized fibularized fibular graft; IP = intramedullary pinning

reported that patients with narrow cross-sectional healing area had significantly higher rates of refracture. They reported a hazard ratio for re-fracture of 3.997 (95% CI: 1.218-13.119) for fibular stability, but only 0.986 (95% CI: 0.977-0.994) for tibial cross-sectional area. Cho recommends performing tibio fibular synostosis in every case. In contrast, Joseph and Mathew reported that fibular surgery is unnecessary when an intramedullary rod is used. They reported union rates of 83% without fibular osteosynthesis⁽¹⁹⁾.

For management of fibular pseudarthrosis, tibial osteosynthesis and fibular osteosynthesis performed during the same operation is preferred for restoring normal ankle mortise. However, in cases where the pseudarthrotic fibula is too dysplastic, alternative methods (osteosynthesis or tibiofibular fusion) for stabilizing the ankle should be used. In the present study, rates of refracture and ankle valgus were comparable to other studies even though 12 cases of fibular pseudarthrosis had nonunion (Table 2). Further studies to investigate fibula procedures in CPT are needed.

We support the use of titanium elastic nails for intramedullary nailing to help realign tibial segments during the Ilizarov operation and to minimize the shearing force at the pseudarthrosis site during healing. Nails should be kept in place in the tibial medullary canal at the time the Ilizarov frame is removed. The justification for positioning a rod across the pseudarthrosis site is for the biomechanical benefits, even though the rod cannot change the biological process underlying refracture. A total contact short leg brace (PTB) is recommended after bone union due to the persistent risk of refracture until or after skeletal maturity.

Conclusion

The results of this study demonstrate that Ilizarov fixation combined with intramedullary nailing is a safe, effective, and practical treatment for management of CPT. This combination technique achieved multiple objectives, including rigid-stable fixation, management of LLD, correction of residual deformity, and prevention of refracture. Moreover, this technique can be used in cases with previous operations that resulted in nonunion.

What is already known this topic?

Ilizarov fixation has been shown to be effective in the treatment of CPT. However, a high rate of associated complications like residual axial deformity

and refracture has been reported.

What this study adds?

Ilizarov fixation combined with intramedullary nailing is a safe, effective, and practical treatment for management of CPT. This combination technique achieved multiple objectives, including rigid-stable fixation, management of LLD, correction of residual deformity, and prevention of refracture. Moreover, this technique can be used in cases with previous operations that resulted in nonunion.

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Funding disclosure

None.

Potential conflicts of interest

None.

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การรักษาภาวะกระดูกหน้าแข้งไม่ติดแต่กำเนิดด้วยวิธีการใส่โลหะตามแกนกระดูกร่วมกับอิลิซาลอฟ

พีระจิตร เอี่ยมโสภณา, ธเนศ อริยวัตรกุล, กมลพร แก้วพรสวรรค์

ภูมิหลัง: ภาวะกระดูกหน้าแข้งไม่ติดแต่กำเนิดเป็นปัญหาที่ยากทางกุมารศัลยศาสตร์ออร์โธปิดิกส์ การรักษาต้องมุ่งเป้าหลายประการ คือ รักษาให้กระดูกติด ป้องกันการหักซ้ำรักษาภาวะขาสั้นยาวไม่เท่ากันและการผิดรูปของกระดูกขา การรักษาภาวะกระดูกหน้าแข้งไม่ติดแต่กำเนิดโดยการตัดกระดูกส่วนที่ผิดปกติ จากนั้นใส่โลหะตามแกนกระดูกร่วมกับอิลิซาลอฟ ซึ่งวิธีการนี้มีข้อดีคือการใช้อิลิซาลอฟจะแก้ไขเรื่องภาวะขาสั้นยาวไม่เท่ากัน ซึ่งเกิดจากการที่ตัดกระดูกส่วนที่ผิดปกติออกไปและใส่โลหะตามแกนกระดูกเพื่อจัดแนวกระดูกให้ตรง อีกทั้งยังสามารถป้องกันภาวะการหักซ้ำ ซึ่งเป็นปัญหาสำคัญของภาวะกระดูกหน้าแข้งไม่ติดแต่กำเนิด

วัตถุประสงค์: เพื่อศึกษาผลการรักษาภาวะกระดูกหน้าแข้งไม่ติดแต่กำเนิดด้วยวิธีการใส่โลหะตามแกนกระดูกร่วมกับอิลิซาลอฟ โดยเน้นศึกษาอัตราการติดของกระดูกและภาวะความผิดรูปที่หลงเหลืออยู่

วัสดุและวิธีการ: ผู้ป่วยสิบสามรายที่ได้รับการรักษาด้วยเทคนิคนี้ ทำการวัดผลโดยการตรวจร่างกายและภาพถ่ายรังสี การวัดผลการศึกษาประกอบด้วยระยะเวลาตั้งแต่รักษาจนกระดูกติด การผ่าตัดเพิ่มเติมจากการรักษาหลัก ผลข้างเคียงของการรักษา การหักซ้ำ ภาวะขาสั้นยาวไม่เท่ากัน ความผิดรูปของกระดูกหน้าแข้งและข้อเท้า รวมถึงบันทึกภาวะกระดูกที่บวมไม่ติด และระยะเวลาที่ติดตามการรักษา

ผลการศึกษา: ภาวะกระดูกหน้าแข้งติดจากการรักษาหลักจำนวน 11 ราย ผู้ป่วยอีกสองรายกระดูกหน้าแข้งติดหลังได้รับการผ่าตัดใส่กระดูกเสริมจากบริเวณเชิงกราน มีผู้ป่วยกระดูกที่บวมไม่ติดมีทั้งสิ้น 9 ราย ในจำนวนผู้ป่วยทั้งหมด 13 คนได้รับการผ่าตัดเพิ่มเติมทั้งสิ้น 30 ครั้ง มุมการโค้งทางด้านหน้าเฉลี่ย 8.5 องศา (0-20 องศา) มุมการโค้งเข่าด้านในเฉลี่ย 9.5 องศา (0-25 องศา) ผู้ป่วยทุกรายพบมีข้อเท้า เข่าสั้นยาวไม่เท่ากันเฉลี่ย 1.7 ซม. (0-3 ซม.) ผู้ป่วยเดินกะเผลก 2 ราย การหักซ้ำเกิดในผู้ป่วย 3 รายและทุกรายกระดูกหน้าแข้งกลับมาติดดีหลังจากได้รับการรักษาโดยเทคนิคใส่โลหะตามแกนกระดูกร่วมกับอิลิซาลอฟ

สรุป: การรักษาผู้ป่วยกระดูกหน้าแข้งไม่ติดแต่กำเนิดโดยวิธีการใส่โลหะตามแกนกระดูกร่วมกับอิลิซาลอฟได้ผลดี แม้ผู้ป่วยจะได้รับการรักษาด้วยวิธีอื่นมาก่อนแล้วไม่ประสบผลสำเร็จในรายงานนี้ผู้ป่วยกระดูกหน้าแข้งติดทุกราย อีกทั้งเทคนิคนี้ยังสามารถแก้ไขภาวะความผิดรูปขาสั้นยาวไม่เท่ากันและการหักซ้ำได้
