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Use of Paley Classification and SUPERankle Procedure in the Management of Fibular Hemimelia

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Background: Fibular hemimelia is the most common deficiency involving the long bones. Paley classification is based on the ankle joint morphology, identifies the basic pathology, and helps in planning the surgical management. Reconstruction surgery encompasses foot deformity correction and limb length equalization. The SUPERankle procedure is a combination of bone and soft tissue procedures that stabilizes the foot and addresses all deformities.

Methods: We retrospectively reviewed 29 consecutive patients (29 limb segments), surgically treated between December 2000 and December 2014. Among the 29 patients, 27 were treated with reconstructive procedures. Type 1 (8 patients) cases were treated with only limb lengthening, and correction of tibial deformities. Type 2 (7 patients) cases were treated by distal tibial medial hemiepiphysiodesis or supramalleolar varus osteotomy. In type 3 (10 patients) cases, the foot deformity was corrected using the SUPERankle procedure. Type 4 (2 patients) cases were treated with supramalleolar osteotomy along with posteromedial release and lateral column shortening. In a second stage, limb lengthening was performed, using the Ilizarov technique. In the remaining 2 patients (type 3A and type 3C), amputation was performed using Syme technique as a first choice of treatment.

Results: The results were evaluated using Association for the Study and Application of Methods of Ilizarov scoring. Excellent results were obtained in 15 of 27 (55%) patients. Six (22%) patients had good results, 4 (14.8%) had fair results, and 2 (7%) had poor results. Mean limb length discrepancy at initial presentation was 3.55 cm (range: 2 to 5.5 cm) which significantly improved to 1.01 cm (range: 0 to 3 cm) after treatment ($P = 0.015$).

Conclusions: Our results and a review of the literature clearly suggest that limb reconstruction according to Paley classification, is an excellent option in the management of fibular hemimelia. Our 2-staged procedure (SUPERankle procedure followed by limb lengthening) helps in reducing the complications of limb lengthening and incidence of ankle stiffness. Performing the first surgery at an earlier age (below 5 y) plays a significant role in preventing recurrent foot deformities.

Level of Evidence: Level IV.

Key Words: fibular hemimelia, SUPERankle procedure, Paley classification, Ilizarov

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Fibular hemimelia is partial or total aplasia of the fibula.[1] It is associated with defects of the femur, tibia, ankle, and foot.[2] It is the most common deficiency involving the long bones and presents clinically as, limb length discrepancy (LLD), anteromedial bowing of the tibia, valgus deformity of the knee, equinovalgus deformities of the foot and ankle, and absent lateral rays.[3–5]

The classifications[4–6] are descriptive of the fibular pathologic abnormality, which does not require reconstruction, and lack descriptions of tibial and ankle pathologic abnormalities, which are the focus of reconstruction.[7,8] The decision to recommend either reconstructive or ablative surgery to the parents of children with fibular hemimelia is difficult and debatable in the orthopedic literature.[3,7,8] Moreover, persistent or recurrent foot and ankle deformities and recurrent valgus deformities of the knee, remained unsolved[9] and are the primary reason for unsatisfactory results.[2] The primary aim in the management of fibular hemimelia is a stable plantigrade foot, with normal limb length and alignment,[7] achieving the most effective gait. Thus, Paley classification, which is based on ankle joint morphology and stability, is helpful in planning surgical treatment.[10]

Reconstructive surgery involves foot deformity correction and limb length equalization. SUPERankle (SUPER is an acronym for Systematic Utilitarian

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R.M.K: All surgeries were performed by R.M.K. She has a major role in initial data collection, right from the beginning. N.A: Assisted R.M.K. in many surgeries in his interim period since last 3 years. He has collected all the previous data from R.M.K. and current data from patients. He was involved in preparation and analysis of the final data chart and preparation and submission of the whole manuscript in agreement with all the authors. S.S: Taught the corresponding author, how to collect data and analyze the data and prepare the manuscript. S.M.K: He has assisted few of the surgeries in the last 3 years and has verified the data. Y.S. and R.N.: Involved in blindly analyzing the data and the manuscript.

The authors declare no conflicts of interest.

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Procedure for Extremity Reconstruction) is a combination of bone and soft tissue procedures developed by Dr Paley in 1996 that stabilizes the foot and addresses all of the deformities.[7,8] In addition, the introduction of the Ilizarov method for limb lengthening has provided an attractive alternative to amputation. Some surgeons prefer this method because it can provide simultaneous correction of the LLD and tibial deformities.[11–13]

Herein, we review our total experience of deformity correction followed by Ilizarov lengthening in patients with fibular hemimelia.

METHODS

A retrospective study was conducted, after obtaining approval from our institutional ethical committee, with consent from all of the patients. All patients with fibular hemimelia surgically treated at our institution between December 2000 and December 2014 were reviewed. Patients lost to follow-up or initially surgically treated elsewhere were excluded. The final sample included 29 patients (12 boys and 17 girls). The right side was affected in 14 limb segments and the left side in 15 limb segments. Patients were classified according to the Paley classification system[7,8,10] for fibular hemimelia (Table 1 and Figs. 1, 2). The number of patients for within each classification is shown in Table 2.

TABLE 1. Paley Classification

Paley Type	Deformities
Type 1	Stable normal ankle joint
Type 2	Dynamic valgus at ankle joint
Type 3	Fixed equinovalgus deformity
According to the ankle-subtalar pathoanatomy, type 3 is subdivided into:	
Type 3a-ankle type	The ankle joint is maloriented into procurvatum and valgus deformity
Type 3b-subtalar type	The subtalar joint has a coalition which is malunited in the equinovalgus with lateral translation
Type 3c-combined ankle and subtalar type	Combination of ankle and subtalar deformities. Both distal tibial malorientation and malunited subtalar coalition are present
Type 3d-talar type	Malorientation of the subtalar joint
Type 4	Fixed equinovarus at the ankle (clubfoot type)

OPERATIVE TECHNIQUES

Among the 29 patients, reconstruction procedures were performed in 27 patients. Two patients (1 type 3A and 1 type 3C) underwent Syme amputation as a first choice of treatment. The procedures used for the different Paley types are shown in Table 2. Type 3 patients were treated using an a la carte SUPERankle procedure[7–10] in 2 stages (Figs. 6–10).

In the first stage, a longitudinal lateral incision was made and the interosseous membrane and fibular anlage were dissected up to the level of the diaphyseal procurvatum deformity of the tibia. The fibrous fibular anlage was partially removed, up to the level of the apex of the tibial deformity, and the peroneal and Achilles tendons were lengthened. The peroneal and posterior tibial nerves were decompressed through separate incisions. Osteotomy was then performed according to the different Paley types as shown in Table 2. Finally, once the foot deformity was corrected, lengthening and tibial deformity correction was performed using the Ilizarov technique in the second stage. The associated deformities and their management are shown in Table 3.

POSTOPERATIVE MANAGEMENT AND METHOD OF EVALUATION

Weight-bearing was permitted, with mobilization of the knee and ankle joint, the day after the Ilizarov procedure. Distraction was started on postoperative day 5 and continued at a rate of 1 mm/d. Once the desired length was achieved, the regenerate was allowed to consolidate. The Ilizarov fixator was removed after the consolidation phase. Once the deformity of foot was

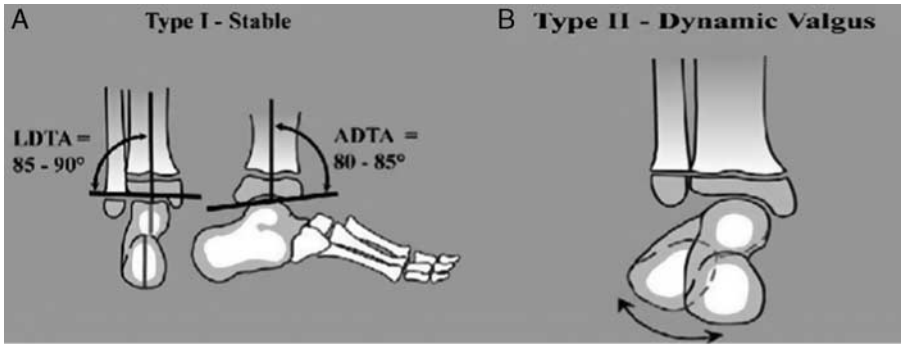

FIGURE 1. Paley's classification type 1: tibial shortening alone with a normal ankle (A); type 2: tibial shortening with dynamic valgus at the ankle (B). ADTA indicates anterior distal tibial angle; LDTA, lateral distal tibial angle. Reproduced with permission from Dr. Dror Paley and The Paley Advanced Limb Lengthening Institute (www.paleyinstitute.org/). The SUPERankle technique was developed by Dr. Paley in 1996. Copyright The Paley Advanced Limb Lengthening Institute, Florida. All permission requests for this image should be made to the copyright holder.

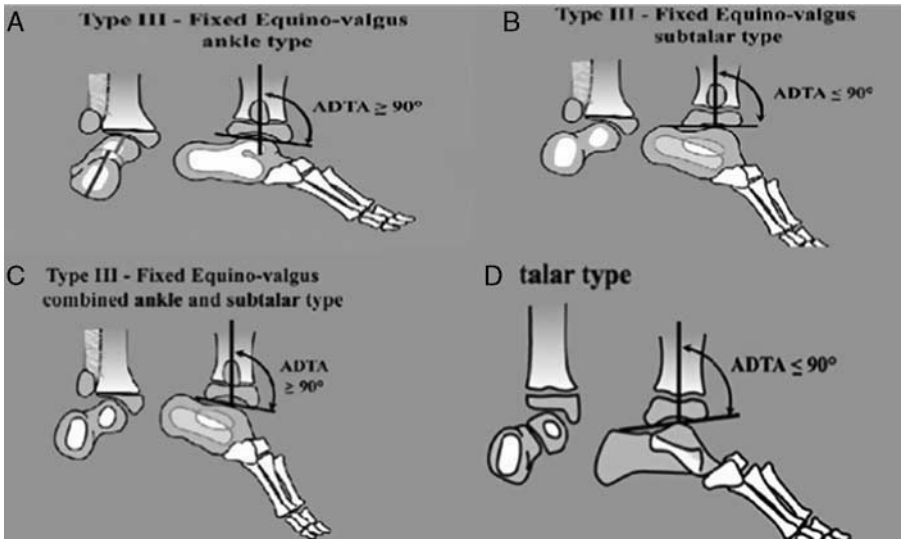

FIGURE 2. Paley classification type 3a: fixed equinovalgus at the ankle joint (A); type 3b: Fixed equinovalgus at the subtalar joint (B); type 3c: fixed equinovalgus at the combined ankle and subtalar joint (C); talar type (D). LDTA indicates lateral distal tibial angle. Reproduced with permission from Dr. Dror Paley and The Paley Advanced Limb Lengthening Institute (www.paleyinstitute.org/). The SUPERankle technique was developed by Dr. Paley in 1996. Copyright The Paley Advanced Limb Lengthening Institute, Florida. All permission requests for this image should be made to the copyright holder.

corrected, the patients used an ankle-foot orthosis brace until skeletal maturity to prevent recurrence. The final results were evaluated clinically, radiologically, and objectively using the ASAMI scoring system[1,14] (Table 4).

TABLE 2. The Number of Cases in Each Paley Type Category and the Associated Surgical Procedure

Paley Type	No. Cases	Treatment
Type 1	8	Foot and ankle surgery was not required, only a lengthening procedure was required with correction of the tibial deformity
Type 2	7	Required realignment of the ankle either by distal tibia medial hemiepiphysiodesis or by supramalleolar varus osteotomy (Figs. 3–5)
Type 3	10	SUPERankle procedure (described in the text)
In addition to the SUPERankle procedure, further management depends on Paley type		
3a	5	Required medial closing wedge supramalleolar osteotomy and fixation with k wires
3b	3	Subtalar coalition osteotomy (bone cuts between the talus and calcaneus for realignment) was required to correct the deformity (Figs. 6–10)
3c	2	Combined supramalleolar and subtalar osteotomies were performed to correct the deformity
3d	—	—
Type 4	2	Supramalleolar osteotomy with posteromedial release and lateral column shortening was performed to correct the deformity

SUPER indicates Systematic Utilitarian Procedure for Extremity Reconstruction.

Statistical Analysis

After verifying the normality of the data, parametric tests of significance were performed. A paired t test was performed to compare the LLD before and after surgery. χ^2 tests were performed to evaluate associations between qualitative variables.

RESULTS

The mean age at the first surgery was 4.06 ± 2.40 years (range: 1 to 9 y). The study demonstrated a statistically significant relationship ($P = 0.03$) between the age at the first surgery and recurrent foot deformities.

The mean follow-up duration was 9.37 ± 2.02 years (range: 5 to 14 y). The mean LLD at the initial presentation was 3.55 ± 1.08 cm (range: 2 to 5.5 cm), which was significantly improved to a mean of 1.01 ± 1.09 cm (range: 0 to 3 cm) at the latest follow-up ($P = 0.015$). Limbs were equalized to within 1 cm in 18 of 27 (66%) patients. Two patients had a shortening of 1.5 cm. Seven patients had a shortening of >2 cm. The mean external fixator duration was 7.74 ± 2.62 months (range: 3 to 14 mo). The mean healing index was 1 cm/mo.

The results in terms of the ASAMI scoring system are shown in Table 5.

Complications

A pin tract infection was the most common complication, followed by recurrent foot deformity (Table 6 and Fig. 11). Two type 2 patients and 4 type 3 patients had recurrence of an equinovalgus deformity.

DISCUSSION

Congenital deficiency of the fibula and associated anomalies of the lower limb represent a well-recognized

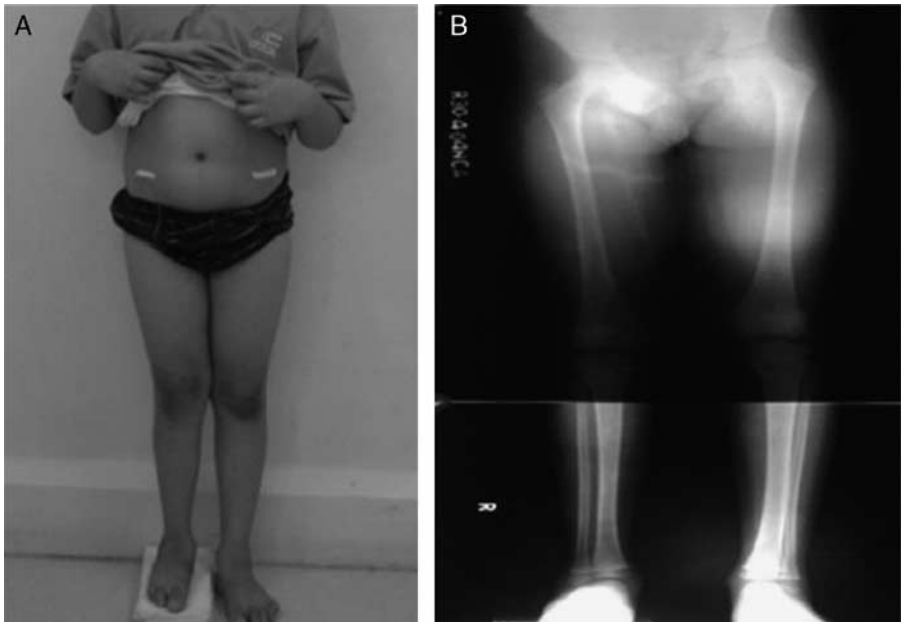

FIGURE 3. Clinical picture (A) and orthoradiogram (B) of a 5-year-old male child with Paley type 2 fibular hemimelia with 3 cm of tibial shortening.

clinical entity. Previous results[2,15,16] have shown that lengthening and deformity correction is the preferred treatment for patients with mild to moderate leg length discrepancy with mild foot deformities (Paley types 1 and 2). Controversial cases include those with more severe foot deformities (Paley types 3 and 4) and greater leg length discrepancies.[8]

The techniques used in the present study had 2 variations from the original SUPERankle procedure as described by Paley.[8] Paley practiced a combined SUPERankle procedure with lengthening in a single surgery, with complete resection of the fibular anlage. The current authors advocate a 2-staged procedure: deformity correction with the SUPERankle procedure followed by a lengthening procedure. The lengthening procedure was usually performed 1 to 2 years after the first procedure and was never performed before the age of 3 years. This variation helps in reducing limb lengthening complications and the incidence of ankle stiffness.[7,8,17] In addition, complete resection of the fibular anlage does not

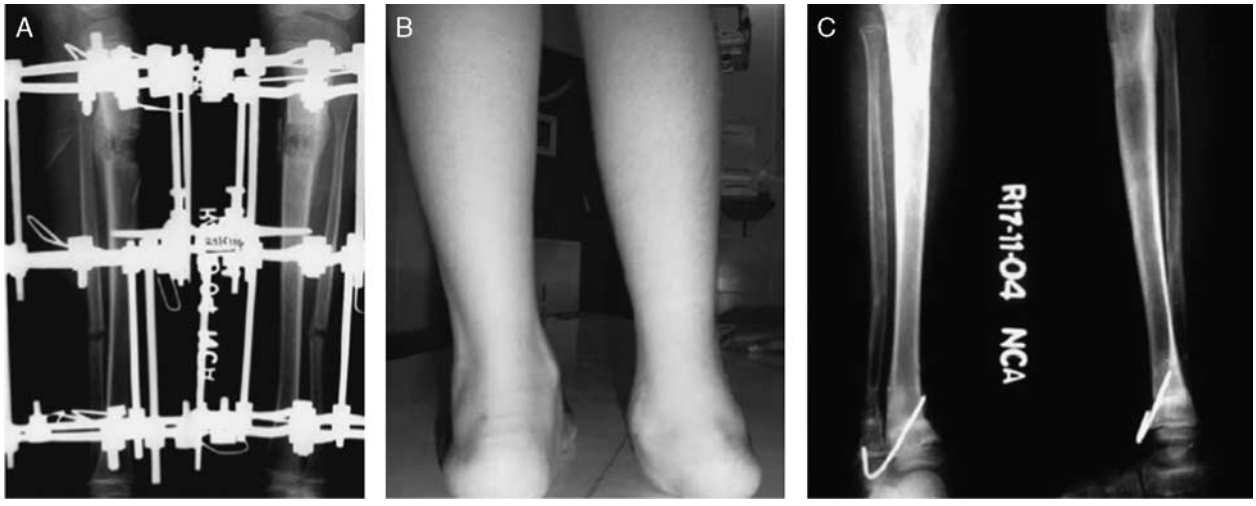

FIGURE 4. X-ray of the same patient as in Figure 3 after treatment with tibial lengthening using the Ilizarov method as an external fixator (A). Clinical picture showing dynamic equinovalgus at the ankle (B), corrected with supramalleolar osteotomy and k wire pinning (C).

FIGURE 5. Clinical picture showing the follow-up of the patient in Figures 3 and 4 at 15 years of age (A), with a stable ankle shown from the posterior (B) and anterior (C) views.

completely eliminate proximal tibia valgus and is not necessary as 10 to 15 degrees varusization of the tibia at the end of the lengthening procedure can balance out the proximal tibia valgus that may occur with incomplete anlage resection.[8] Moreover, complete anlage resection has a theoretical risk of injury to the common and deep peroneal nerves. The present study had only 1 case of recurrent genu valgum (Figs. 8, 9), which was treated during subsequent lengthening. Therefore, we recommend a partial resection of the fibular anlage up to the level of the apex of the deformity.

Outcomes associated with the reconstruction procedures are favorable. For example, Paley et al[18] compared 22 patients treated with the SUPERankle

FIGURE 6. Clinical picture (A) of a 5-year-old male child with Paley type 3b fibular hemimelia with tibial shortening and anteromedial bowing (B). A magnetic resonance imaging shows that the os calcis is translated laterally to the talus (C).

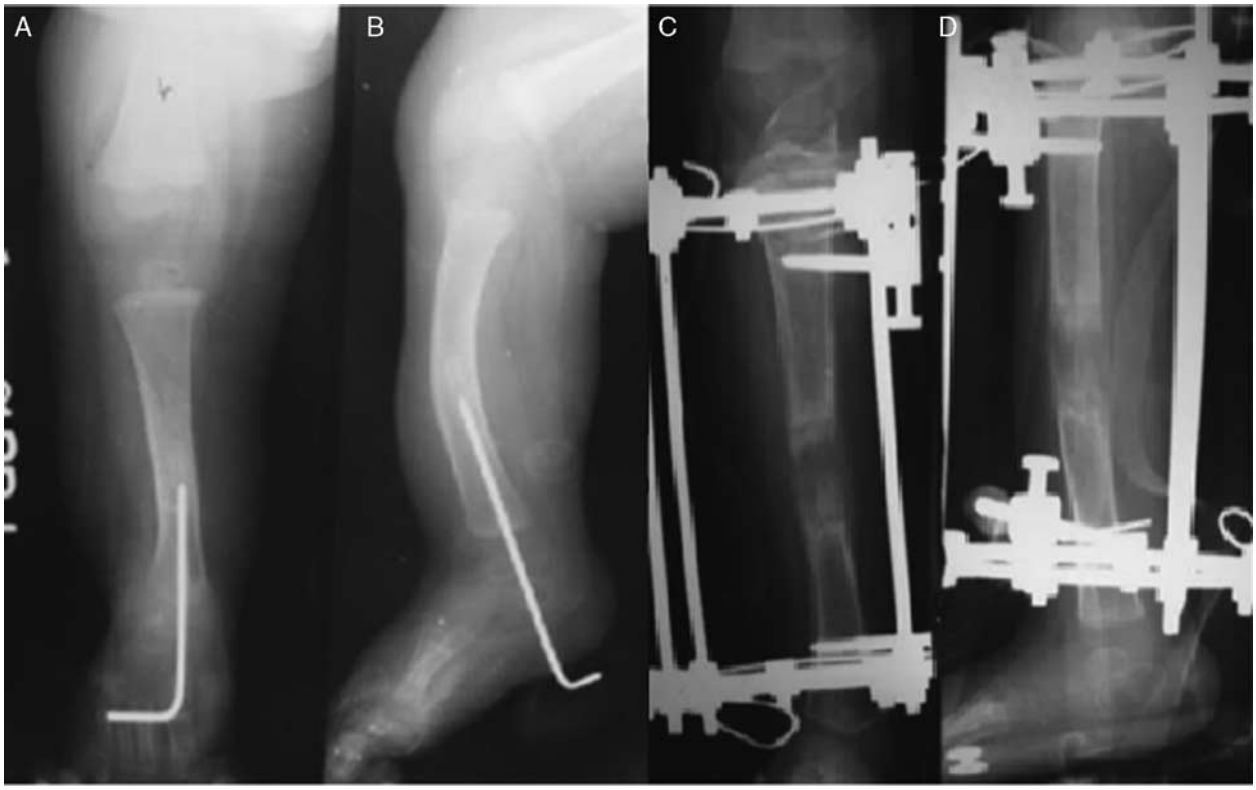

FIGURE 7. The deformity in Figure 6 was treated with partial resection of the fibular anlage, peroneal lengthening, and tendo-Achilles lengthening. An x-ray showing the anteroposterior (A) and lateral view (B) of the tibia, wherein a subtalar osteotomy was performed and fixed with k wires. Further treatment with lengthening using the Ilizarov method as an external fixator was performed. An x-ray showing regenerate callus formation at the corticotomy site in anteroposterior (C) and lateral views (D).

procedure, combined with lengthening, to an age-matched group of patients who underwent Syme amputation; the results demonstrated no difference in function between the 2 groups. The big advantage of reconstruction surgery is that patient retains a sensate foot that can feel the ground, thereby providing balance and proprioception.[7,8] However, Birch et al[19] classified fibular hemimelia according to the number of rays of the foot and recommended amputation for most cases with less than 3 rays. The present study had 2 patients with 2 rays who underwent primary amputation as there was not enough surface area for a stable plantigrade foot.

The present study did not compare limb reconstruction to amputation because the current challenge involves improving the results of lengthening.[7,8] An analysis of literature[15,20,21] suggests that unsatisfactory results after a salvage procedure are mainly related to recurrent or residual foot deformities.[8] For example, Naudie et al[2] achieved satisfactory results in only 4 of 10 cases after lengthening and the reason for unsatisfactory outcomes involved residual or recurrent foot and tibial deformities.[8] Cheng et al[22] reported a similar experience in a small prospective group of 4 cases of lengthening, with unsatisfactory results secondary to recurrent tibial and foot deformities.

The current study revealed a statistically significant relationship between the age at the first surgery and recurrence of foot deformities. Hence, early treatment is important in reducing recurrence and unsatisfactory results. Five of 12 patients with an age at the first surgery of more than 5 years had recurrence, while only 1 of 15 patients with an age at the first surgery of less than 5 years had recurrence. Paley[8] prefers to perform this procedure when the patient is between 18 and 24 months of age, performing lengthening at the same time. We have performed SUPERankle as early as 12 months, with lengthening performed as a next-staged procedure. Furthermore, genu valgum at the knee contributes to recurrent deformities at the ankle. As there is usually no subtalar joint present, genu valgum cannot be compensated by a mobile subtalar joint.[7,8] It is therefore important to identify and treat knee valgus to improve the results of the foot correction and to help prevent recurrent ankle valgus.[7,8] Despite the above precautions, recurrence is still a problem. The theoretical explanation is that by reorienting the ankle joint via supramalleolar osteotomy, the distal tibial growth plate is maloriented. This could lead to gradual recurrence, which can be addressed at the time of next staged lengthening.[7,8]

FIGURE 8. Clinical picture (A) of the same patient as in Figures 6 and 7 at 12 years follow-up. Orthoradiogram (B) showing internal rotation of the tibia, genu valgum, and a shortening of 5 cm.

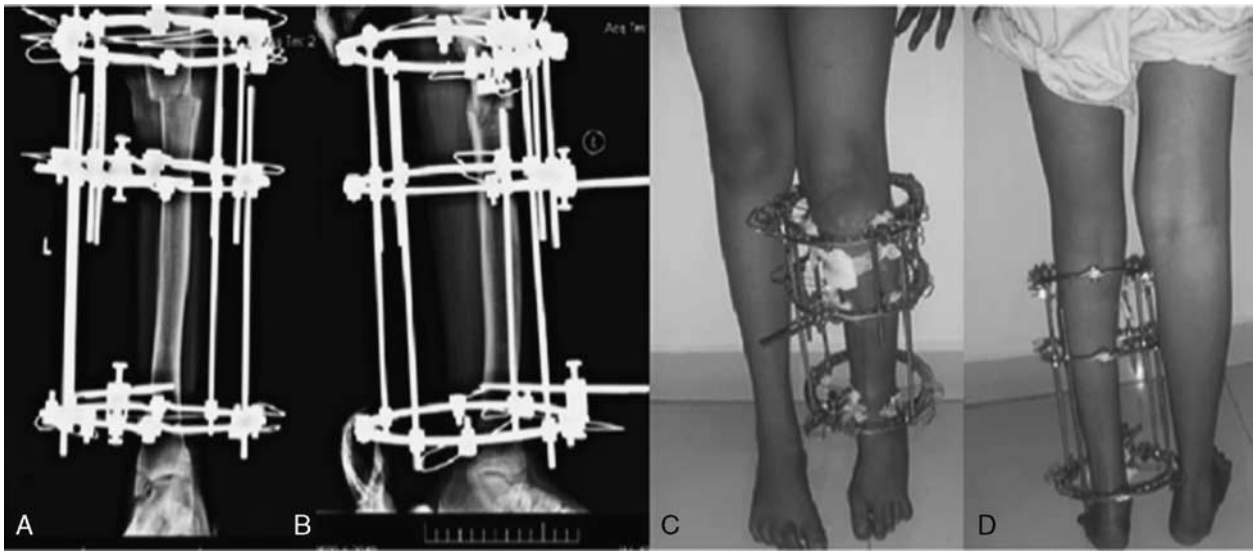

FIGURE 9. The rotation, genu valgum, and shortening in Figure 8 were treated with derotational osteotomy and lengthening of the tibia. An x-ray of the patient in the anteroposterior (A) and lateral views (B) with osteotomized tibia and stabilized using the Ilizarov method. Clinical pictures (C, D) of the patient showing both patellas facing forward with Ilizarov fixation in situ.

FIGURE 10. Orthoradiogram (A) of a patient at 14 years follow-up with 1 cm of shortening and 5 degrees of internal rotation deformity. Clinical picture of a patient at the same time showing internal rotation deformity (D) which is well compensated by external rotation at the hip joint (B). The patient is able to squat (E) and sit cross-legged (G) with a full range of motion at the knee and appreciably good motion at the ankle (C). The patient has 1 cm of shortening with a stable ankle (F).

Previous studies have shown the effectiveness of lengthening for fibular hemimelia. Using gait analysis Johnson and Haideri[23] showed that, successful lengthening resulting in plantigrade feet and well-aligned tibia, is associated with better ankle push-off strength and better knee flexion strength compared with that in patients who underwent Syme amputation.[8] Furthermore, Catagni et al[1] reviewed 32 patients with type 3 fibular hemimelia treated with successive lower limb lengthenings and deformity correction using the Ilizarov method; nearly equal limb length and a plantigrade foot were achieved in half of the patients. Similarly, Jawish and Carlioz[11] reported good correction of the foot in 60% of cases of fibular hemimelia treated by lengthening. In addition, Paley[7,8] (in his unpublished results presented at AAOS 1999, Anaheim, CA) was able to achieve good or excellent functional results, including the desired goal of lengthening, in 36 of 38 lengthened legs.[8]

Similarly, the present study had favorable results. According to the ASAMI scoring system, 15 of 27 cases had excellent results. Among patients classified as Paley type 1, the majority had excellent ASAMI scores and all had at least a good score. In contrast, among Paley type 3a cases, some patients had poor and fair scores. In addition, none of the type 3c cases had a score of more than fair. However, as the number of cases within each type was small, an association between Paley type and

TABLE 3. Associated Deformities and their Treatment

Associated Deformities	No. Cases	Treatment
Ankle subluxation	2	Ankle arthrodesis
Anteromedial bowing of the tibia	23	Deformity correction along with lengthening
Tibial torsion	1	Deformity correction along with lengthening
PFFD	1	Femoral lengthening
Congenital short femur	3	Tibial lengthening: 2 cases Femur lengthening: 1 case
Genu valgum	5	Distal femur varus osteotomy: 1 case Distal femur medial hemiepiphysiodesis: 4 cases
Opposite side CTEV	1	Posteromedial release
Postaxial hypoplasia	29	Two patients had < 3 rays; thus, primary amputation was performed

CTEV indicates congenital talipes equinovarus; PFFD, proximal focal femoral deficiency.

TABLE 4. ASAMI Scoring

Bone results using ASAMI scoring system	
Excellent	Union, no infection, deformity < 7, limb length discrepancy < 2.5 cm
Good	Union + any 2 of the following: absence of infection, < 7 deformity and limb length inequality of < 2.5 cm
Fair	Union + only 1 of the following: absence of infection, deformity < 7 and limb length inequality < 2.5 cm
Poor	Nonunion/re-fracture/union + infection + deformity > 7 + limb length inequality > 2.5 cm
Functional results using ASAMI scoring system	
Excellent	Active, no limp, minimum stiffness (loss of < 15 degrees knee extension/ < 15 degrees dorsiflexion of the ankle), no RSD, insignificant pain
Good	Active, with 1 or 2 of the following: limp, stiffness, RSD, significant pain
Fair	Active, with 3 or all of the following: limp, stiffness, RSD, significant pain
Poor	Inactive (unemployment or inability to perform daily activities because of injury)
Failures	Amputation

ASAMI indicates Association for the Study and Application of Methods of Ilizarov; RSD, reflex sympathetic dystrophy.

TABLE 5. Paley Type and the Resultant Distribution of Patients in Each Type, as Evaluated by ASAMI Scoring

Paley Type	No. Patients in Each Category as Evaluated by ASAMI Score
Type 1	6 patients had excellent results and 2 had good results
Type 2	3 had excellent results, 3 had good results, and 1 had fair results
Type 3a	3 had excellent results, 1 had fair results, and 1 had poor results
Type 3b	2 had excellent results and 1 had good results
Type 3c	1 had fair results and 1 had poor results
Type 4	1 had excellent results and 1 had fair results

ASAMI indicates Association for the Study and Application of Methods of Ilizarov.

TABLE 6. Complications and their Management

Complications	No. Cases	Treatment
Pin tract infection (grades 1 and 2)	7	Local antiseptics, oral and injectable antibiotics, wire tensioning, and wire exchange
Recurrent foot deformity	6	Revision TA lengthening, posterior soft tissue release, and supramalleolar osteotomy
Knee subluxation	3	Reduction of the knee subluxation and addition of a tibial ring
Rocker bottom foot	5	—
Fixed flexion deformity of the knee	5	—
Delayed union of corticotomy site	1	Debridement, wire exchange, and stabilization of frame
Recurrent genu valgum	1	Deformity correction along with lengthening (Figs. 8, 9)

TA indicates tendo-Achilles.

ASAMI scores could not be verified statistically. Two patients with ankle fusion were rated as fair. The ankle fusion was performed as a successful way of permanently stabilizing the foot[12] and the patients did not require any additional surgeries for the ankle or foot. In addition, the present study had excellent follow-up duration, with a mean of 9.37 years and a maximum of 14 years.

In the present study, 18 of 27 patients had limbs equalized to within 1 cm. Two patients had a shortening of 1.5 cm. These patients did not require any additional surgeries and were managed with the help of a shoe rise. Patients with a shortening of more than 2 cm were lengthened again after a minimum gap of 2 years.

Limb lengthening with an external fixator alone is fraught with complications during the long external fixator period.[24] Early removal of fixator will decrease most of the associated complications and can be done with the help of a lengthening over plate and recently designed slotted plates by the current author.[25] Delayed union of the corticotomy site was due to frame instability, associated with a grade 2 pin tract infection and wire loosening. This was treated with debridement and wire exchange, thereby stabilizing the frame. Three patients had knee subluxation and was associated with femoral lengthening.[24] This was treated by reducing the knee joint with the addition of a tibial ring.

The main limitation of the present study is that, due to relative rarity of this condition and small sample size, a comparison in functional outcomes and complications between the different Paley types was not possible.

CONCLUSIONS

Our long-term follow-up clearly suggests that limb reconstruction according to the Paley classification is an excellent option in the management of fibular hemimelia. Our 2-staged procedure of SUPERankle followed by lengthening with a minimum gap of 1 to 2 years helps in reducing the complications of limb lengthening and ankle stiffness. Performing the first surgery at an earlier age (below 5 y) plays a significant role in preventing recurrent foot deformities. Nevertheless, the surgeon must be thoroughly trained to get the desired result.

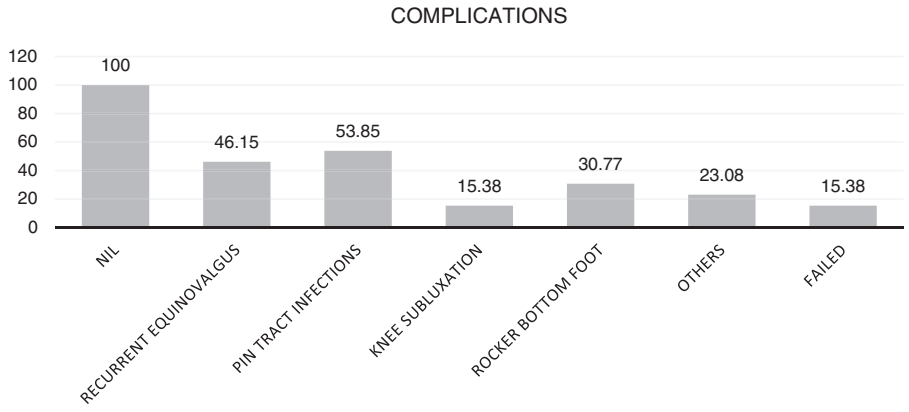

FIGURE 11. Thirteen of 29 cases did not experience complications. The percentile representations of all the complications are depicted with respect to cases without complications.

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