



## Three-dimensional correction of fibular hemimelia using a computer-assisted planning : technical report and literature review

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The purpose of this study is to investigate a stepwise approach to translate the principles of deformity correction from 2D plain radiographs to a 3D computer assisted pre-operative planning (CAP), when treating a complex case of fibular hemimelia.

Computed tomography slices were used to perform a 3D reconstruction of the deformity. CAP determined the different axes and apex of deformity based on geometrical functions of the software. The obtuse angle was computed and used to determine the concomitant osteotomy angle. An additional review of the literature was performed, allowing comparison towards the current treatment approaches.

The pre- and post-operative clinical and radiographic follow up is reported. The computer assisted planning was applicable in a complex fibular hemimelia deformity. The literature review demonstrated no previous use of a 3D computer assisted planning.

This case report provides a feasible and effective method to convert the principles of deformity planning from a 2D to a 3D setting. The added value of this technique for clinical practice should be confirmed in further prospective studies.

**Level of evidence : Level V, Case report**

**Keywords :** fibular hemimelia ; hindfoot deformity ; computer assisted planning ; corrective osteotomy.

## INTRODUCTION

Fibular hemimelia (FH) is a congenital condition characterized by foot deformity and leg length discrepancy (LLD) (39). FH occurs in between 1:350,000 and 1:50,000 births and can either be uni- or bilateral (17,32,38). Because the physical disability imposed by FH markedly reduces quality of life by limiting or preventing physiological gait, selecting and carefully planning the appropriate treatment is crucial (28,29).

Diagnosis is performed clinically and confirmed on plain radiographs. Upon examination, the

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Table I. — 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 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)  |

severity and characteristics of the condition can be classified based on criteria proposed by Paley et al (21,32). While commonly used FH classification schemas like Achterman and Kalmchi or Coventry and Johnson focus on fibular absence, Paley noted fibular absence does not strongly correlate with the deformity (1,13,18,29,30). His classification thoroughly details the complete deformity to better inform reconstruction options (Table 1) (32).

Conservative treatment can be initiated, but eventually a surgical correction will be needed to realign the ankle and hindfoot. Currently, both a direct surgical correction using internal fixation with angular stable plates as well as an indirect correction using a circular frame (Ilizarov Technique) are described (10, 32). Both techniques have their own advantages and disadvantages (11,22,27,32) but both depend on detailed pre-operative planning. The latter is still based on 2D radiographs despite correcting a 3D deformity.

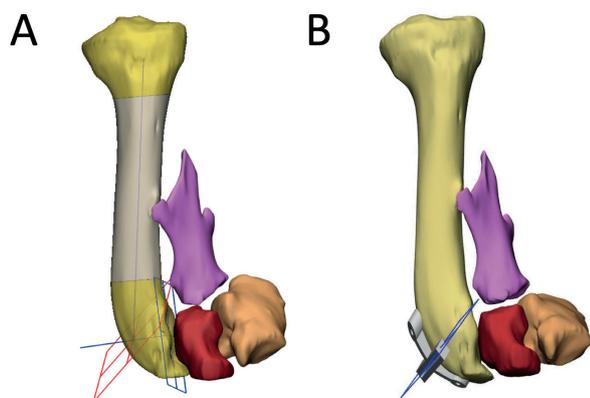
Therefore, the aim of this case report is to depict a stepwise approach to performing a pre-operative 3D computer assisted planning (CAP) when treating a complex case of fibular hemimelia. This approach overcomes one substantial weakness of current techniques: pre-operative planning's dependence on plain 2D radiographs. The stepwise computed 3D preoperative planning, perioperative implementation, and post-operative follow up are reported.



Figure 1. — Pre-operative bilateral fibular hemimelia deformity (A) Clinical (B) Radiographical Full leg (C) Radiographical A/P and lateral.

## MATERIALS AND METHODS

We report the case of an eight-year-old girl who has bilateral FH with mainly a coronal deformity (Fig. 1A-C). The pre-op AOFAS hindfoot score was 32 and the feet were not plantigrade. Previously the patient was able walk until the age of seven years using a Kaye walker but last six months the patient was wheelchair bound as a result of deformity progression to an unbraceable hindfoot. The primary indication for surgical correction was marked pain situated on both medial sides of the ankles. The clinical complaints and radiographic deformity were more pronounced on the left side, type 3c according to Paley et al. (Table I) (21,32). For this reason, a left side deformity correction was planned first in agreement with the patient and



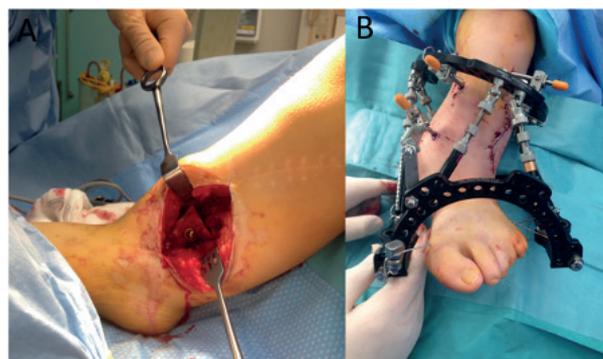
**Figure 2.** — Pre-operative deformity models (A) Converting the 2D principles of deformity planning to a 3D reconstruction. (B) Possibility to add a patient specific guide.

parents. The decision to reconstruct resulted from different discussions with both the parents and the patient. Ongoing discussions about different treatment options had been initiated from an early age. From the beginning, both parents have insisted on avoiding amputation, and their child recently expressed a similar desire, despite meeting amputation criteria (4,8).

Therefore, the goal of the procedure was salvage orientated: to have a shoe able and plantigrade foot, rather than a complete radiographic correction. Limb length was not the major concern in this case but could have been addressed as well by the same principles as described below.

#### a) Pre-operative planning

The osteotomy was planned in the coronal plane to run through the apex of the deformity and according to the line bisecting the obtuse angle as described in the principles of deformity correction by Paley and Herzenberg (31). This procedure was performed using computer aided design (CAD) software in a sequential manner (Fig. 2). Firstly, images were segmented out of CT slices to obtain a 3D volume (Mimics v20, Haasrode, Belgium) (Fig. 2A). Secondly, the best fitted longitudinal shaft axis of the tibia and the axis perpendicular to the tibiotalar joint surface were computed



**Figure 3.** — Per-operative setting (A) Application of the interference screw to maintain the ankle mortise (B) Application of the TSF foot frame.

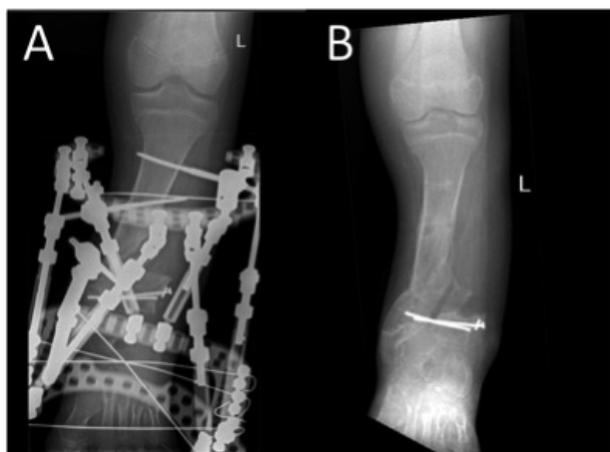
(3-matic v12, Haasrode, Belgium). Thirdly, the apex was computed as the intersection of both axes, determining the osteotomy plane. The obtuse angle was 93 degrees in the coronal plane. A proximal translation of the osteotomy plane needed to be performed, starting 5cm from the tip of the medial malleolus because of the nearby tibiotalar joint level. Three-dimensional models of this type allow for a patient specific guide (Fig. 2B).

#### b) Perioperative procedure

The patient was positioned prone under general analgesia. A tourniquet at 220mmHg was used. A lateral incision was used to attach the distal fibula to the tibia in order to maintain the ankle mortise (Fig. 3A). Afterwards, a longitudinal medial incision over the tibia was performed. A k-wire was drilled according to the 46.8 degrees angle measured in the coronal plane, starting 5cm from the tip of the medial malleolus and the intersection of the anatomical tibia axis of the proximal fragment to guide the osteotomy. The TSFO (Smith and Nephew, Memphis, TN, USA) 2-third circular frame and a U foot frame were used on the reference fragment and the moving fragment, respectively (Fig. 3B).

#### c) Post-operative follow up

The TSF software computed a lengthening schedule based on the mathematical iterations



**Figure 4.** — Post-operative radiographs (A) TSF frame in place (B) TSF frame removed after 3 months post op.

created by the struts. An initial lengthening was performed to disengage the fragments. A weekly clinical and radiographic post-operative follow-up was planned (Fig. 4A). After the achieved correction no compression of the fragments was needed, as osseous contact was already present.

## RESULTS

The final follow up at our outpatient clinic was at six months. At three weeks post-operative, the patient developed a pin-tract infection which resolved after one week of oral antibiotics. The correction stopped at five weeks due to skin tensioning. At that time, the treating physician, parents, and patient were satisfied with the correction obtained and thus treatment was not restarted.

The patient was allowed partial weightbearing at six weeks and the frame was removed at three months post-operative when sufficient consolidation appeared. The AOFAS score improved from 32 to 61. The magnitude of the deformity improved from 90 to 20 degrees of valgus, leaving a residual valgus (Fig. 4B), but was well tolerated by the patient in a post-operative ankle foot orthosis.

## DISCUSSION

This case report demonstrates an effective and feasible method to correct fibular hemimelia in

Table 2. — Technique for computer-assisted correction of fibular hemimelia

| Technique/Steps   |
|---|
| 1. Segmentation of CT images in Mimics® and generation of STL (stereolithography) files   |
| 2. Import STL files into 3-matic®   |
| A. Define a coordinate system on the proximal tibia, modified to Grood and Suntay (Grood and Suntay, 1988 approx):  |
| - The Medial/Lateral axis is based on the center of the medial and lateral condyle of the tibial plateau.   |
| - The Superior/Inferior axis is located between the tibial knee center and the center of diaphysis proximal to the deformity.   |
| - The Anterior/Posterior axis is defined by the cross product of the mediolateral and superoinferior axis.  |
| B. Define the Center of Rotation of Angulation (CORA) by intersecting the computed best fitted axis of the proximal and distal shaft of the tibia (Fig 2A)  |
| C. Computed the bisecting axis of the obtuse angle obtained from the CORA in the coronal plane to determine the direction of the osteotomy and to fit a potential patient specific guide (Fig 2B) |
| D. In this case, the osteotomy would be to close to the tibiotalar joint, hence a proximal translation was performed of 2cm   |
| E. Perform a virtual osteotomy with the cutting tool and position the distal segment to the desired correction.   |
| 3. Computed translational and angular displacements of the distal segment   |
| - Translations are based on the shift of the proximal point of the distal segment along the three prior defined axes.   |
| - Rotations are based on the angular displacement of the longitudinal axis of the distal segment pre-osteotomy versus post-correction.  |

clinical practice using computer assisted pre-operative planning based on stepwise approach (Table 2). This finding is in accordance with other papers demonstrating the advantages of 3D pre-operative planning (19,41).

Current treatments of fibular hemimelia can grossly be divided into three groups : amputation, acute correction, and gradual correction. The latter two groups depend more heavily on an accurate pre-operative and peri-operative plan.

Initial studies describing FH often recommended amputation for severe cases as few alternatives existed. In one of the earlier studies describing FH, Thompson *et al.* reviewed 31 cases (25 patients) of congenital absence in which eight patients underwent a Syme's amputation due to a limb discrepancy of greater than 5 inches (40). Subsequent FH studies commonly indicate that amputation should be performed for feet with a LLD greater than 20%, absence of three or more rays, and/or severe valgus foot deformities or deformities which would preclude a plantigrade, weightbearing foot (4,8). Syme's and Boyd's amputation are preferred as the primary form of amputation instead of over the knee amputation as the resultant heel pad allows for good weightbearing, greater control, and better fitting with a prosthesis. A tibial osteotomy is recommended for tibial angulation greater than 30 degrees and may be performed either before or during amputation for FH patients to ameliorate associated deformities of FH including genu valgum (4) and tibial bowing (42). Tibial osteotomies can improve alignment of the limb, result in a more cohesive prosthesis fit, and reduce the chance of deformity reoccurrence during growth (42).

The popularization of the Ilizarov method for gradual correction of FH has made amputation less popular and enhanced methods based on limb salvage and complex reconstructions. Concerns have been raised about whether this treatment should be applied for severe cases in which the technique itself becomes more complex and prone to complication, and the functional outcome might be disappointing. In one of the earliest studies on the topic, Catagni *et al.* studied 61 FH patients treated by the Ilizarov method who were divisible into three grades of FH severity based on the Dalmonte classification (1

being the least severe and 3 the most) (9). Treatment was largely successful with an increase in bone lengthening ranging from between 4 and 37 cm and ultimately resulting in better angulation. However, complications varied drastically between the groups. All 24 grade I patients had no significant complications and had satisfactory joint function while grade III patients often had to undergo two or three stages of correction and faced such major complications as foot deformity relapses, fractures, edema, and limited range of motion. Despite varying degrees of complication, Catagni's only recommended limitation was that the patients should not be 15 years or older with greater than 20% LLD and a long standing equinovalgus foot deformity. Literature has since indicated that older FH patients who undergo the Ilizarov method suffer from a greater amount of complications which often are also more severe (18). As such, the initial limb lengthening surgery is recommended at a young age (12,18). Subsequent literature has failed to reach a consensus on treatment for severe cases. Changulani *et al.* recommended the Ilizarov method for patients with at least three rays and amputation for those with less (12), while Catagni *et al.* has continued to recommend surgery for patients with severe Type III FH (Dalmonte classification), achieving a plantigrade foot and nearly equal limb length for half of his patients (16 of 32 patients) (11).

Numerous studies have followed both amputation and limb lengthening cohorts to address the controversy surrounding severe FH treatment. After comparing amputation (12 patients) and limb lengthening (10 patients) cohorts, Naudie *et al.* concluded amputation was still the preferred method for severe FH. Patients treated with the Ilizarov method were subject to more extensive complications, a greater number of surgeries, an extended time in the fixation device (average 7.8 months), and more frequent (4.1 vs 2.3) and longer hospital admissions (49.0 vs 19.3) (29). Mixed cohort studies by Oberc *et al.* and McCarthy *et al.* offered similar treatment recommendations and outcome data, additionally noting that patients who underwent Ilizarov treatment had higher pain scores and lower perceived functionality and satisfaction with their treatment (27,30). However, the methodology of

some such studies has been criticized for an absence of reliable outcome measurements, a small sample size, and overemphasis of the severity of negative outcomes ; critics of such studies have advocated that amputation should be a last resort (32,33).

Novel acute correction techniques have been developed to supplement limb lengthening procedures and reduce complications rates and deformity severity. Paley *et al.* successfully performed his SUPERankle procedure on 22 type III or IV (Paley classification) FH patients (32). This cohort had a limited amount of pain and equal functionality as a cohort of patients who underwent Syme's amputation. In contrast to contraindications for limb lengthening procedures, Paley indicated surgery can be performed with only one functional ray and a predicted LLD of 26 cm (femur lengthening can be performed for higher LLD cases). Kulkarni *et al.* evaluated SUPERankle patients using the ASAMI score : six patients achieved excellent or good results, 2 fair, and 2 poor (22). Disagreement exists over whether concurrent lengthening procedures may increase incidence of ankle stiffness and limb lengthening complications (22,32).

The use of Ilizarov external fixators for lengthening and axis correction requires mechanical hinges and translation mechanisms to build a custom-made frame for each patient (22,32). Complex deformities might require frequent structural changes to the frame during the course of lengthening. Hexapod fixators allow simultaneous correction of multiplanar complex deformities and are quicker to construct than the Ilizarov external fixator (22,32). This made their application for correction of complex congenital deformities such as FH more popular. However, one should realize that the outcome of deformity correction remains dependent on the application of basic osteotomy rules. This does require a perfect description of the deformity. Until the advent of this technique, pre-operative planning has depended on plain 2D radiographs to treat a 3D deformity.

The novelty of this study is the application of 3D methods in the pre-operative planning of a complex fibular hemimelia deformity. To our knowledge and in accordance with the above review no reports has been published previously using a similar approach.

On the other hand, computer assisted surgery has been widely used in the foot and ankle (35). However, the emphasis is mostly on peri-operative achievements, rather than on an exact pre-operative plan.

This study has several limitations. First, only one patient is presented, which can be considered the main shortcoming of a case report. Second, an incomplete correction of the valgus deformity was achieved due to the magnitude of the pre-operative deformity. Third, a pre-operative planning was performed on a non-weightbearing CT, possibly underestimating the hindfoot deformity. The recent advent of weightbearing cone beam CT scans could improve the accuracy of present 3D pre-operative planning of the hindfoot and can involve assessment towards the complete lower limb alignment (2,3,5-7,14-16,20,23-26,34,37,36).

## CONCLUSION

This technical report provides a feasible and effective method to convert the principles of deformity correction from a 2D to a 3D setting. A stepwise approach is provided, which can be of use for similar complex hindfoot deformities. At present, this process remains time consuming, but recent advances in computed imaging allow for automated segmentation and measurements, potentially improving the current workflow (23). The added value of this technique for clinical practice should be assessed in further prospective studies.

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