Malunited Forearm Fractures in Children

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Research Funding: none

Financial Support for the publication of this thesis was provided by:

- Department of Orthopedics & Sports Medicine, Erasmus MC
- Department of Orthopedics, Reinier Haga Orthopedic Centre
- Stichting Research Orthopedie Delft
- Stichting FORCE (Foundation for Orthopedic Research Care & Education)
- Nederlandse Orthopaedische Vereniging (NOV)
- Nederlandse Vereniging voor Handchirurgie (NVvH)
- Annafonds |Nederlands Orthopedisch Research en Educatie Fonds
- Materialise BV
- Pro-Motion Group
- Manometric
- VREST
- Oldelft Benelux
- MediNL
- Oldelft Benelux
- ETB-BISLIFE
- Tromp Medical BV
- Bergman Media
- Oudshoorn Chirurgische Techniek
- Link en Lima
- Trauma Platform
- Bauerfeind
- Chipsoft
- ABN AMRO

ISBN : xxxxxxxxx

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Malunions na onderarm breuken bij kinderen

Proefschrift

ter verkrijging van de graad van doctor aan de
Erasmus Universiteit Rotterdam
op gezag van de
rector magnificus

Prof. dr. A.L. Bredenoord

en volgens besluit van het College voor Promoties.
De openbare verdediging zal plaatsvinden op

donderdag 7 september 2023

om 13:00 uur

door

Kasper Roth
geboren te Eindhoven

Erasmus University Rotterdam

Promotoren:	prof. dr. D. Eygendaal
	prof. dr. F. Stockmans
Overige leden:	prof. dr. M.H.J. Verhofstad
	prof. dr. E.H.G. Oei
	prof. dr. I.B. Schipper
	Prof. dr. J. N. Doornberg
Copromotor:	dr. J.W. Colaris

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Chapter 1

General introduction and goal of the thesis

Introduction

Before the age of 16, about one-third of children will sustain a fracture [1]. Fractures of the distal metaphyseal forearm account for 33% of pediatric fractures, whereas diaphyseal both-bone forearm fractures account for 5.4% [1]. A distal metaphyseal fracture is located in the distal fifth of the forearm, whereas the diaphysis is defined as the segment of the bone between 20% and 80% of its entire length [2]. In children aged 10-14 years, every year, 1.3% of boys and 0.9% of girls sustain a distal metaphyseal forearm fracture in the Netherlands [3]. The incidence of diaphyseal forearm fractures in children is increasing, often due to high-energy traumas such as trampoline injuries [4].

A malunion occurs when a fracture heals in a non-anatomical position. The healing of a fracture in a growing child is very different from the adult skeleton. The growing skeleton of children has a remodeling capacity and will correct angular deformity in time. Therefore, a distal forearm fracture with some degree of displacement or angulation can be safely accepted in the expectance that remodeling will occur.

Two well-known biological laws contribute to the remodeling process in pediatric fractures: The ‘Hueter-Volkmann Law” contributes to 75% of remodeling and states that epiphyseal growth is decelerated by increased mechanical compression of the growth plate and is accelerated by reduced loading of the growth plate. Wolff’s law contributes to the remaining 25% of remodeling and states that new bone is formed where it is mechanically necessary and reabsorbed where it is unnecessary [5].

The remodeling capacity depends on various factors:

- The proximity of fracture to the physis; the nearer the fracture to the physis, the greater the potential for spontaneous correction [6].
- The activeness of the physis: the distal radial physis provides 75% of radial growth, while the proximal radial physis provides 25% of radial growth [7].
- Age at trauma: The younger the child, the more angulation one can accept [8].
- Sex of the patient: The mean age for closure of the physis differs between boys and girls: 14.5 and 12.9 years, respectively [9]. Hence, boys have greater remaining growth potential than girls of the same age.
- The severity of angular deformity: Greater angulated fractures tend to remodel faster, and the remodeling speed decreases as remodeling progresses. Distal radius fractures in children with angulations ≥15° remodel with a mean remodeling speed of 2.5° per month [10].
- Plane of angular deformity: dorso-volar angulation remodels better than radio-ulnar angulation because deformity in the sagittal plane occurs in the main plane of movement of the wrist [11]. Rotational deformity does not resolve spontaneously at all.

Due to this remarkable fracture remodeling potential seen in children, the assessment of the long-term follow-up after displaced forearm fractures in children is essential to determine the optimal treatment strategy for pediatric forearm fractures.

Although orthopedic surgeons worldwide encounter pediatric forearm fractures very frequently, we still do not know the best treatment strategy. This thesis aims to provide a backbone when opting for the best treatment strategy when you find yourself in another classic, ever-returning treatment dilemma regarding a child with a displaced or malunited forearm fracture.

PART I: Distal metaphyseal forearm fractures in children

Chapter 2: Think twice before re-manipulating distal metaphyseal forearm fractures in children

In 1962 Gandhi et al. stated that *"Angular deformity of the distal third of the forearm usually corrects fully with the growth of the bone within five years, provided the physis does not fuse in the meanwhile"* [6]. However, despite the remarkable potential for remodeling seen in pediatric distal forearm fractures, surgeons still naturally tend to try to make each fracture radiographically more anatomic [12]. Perhaps this trend has arisen to avoid anxious parents demanding an answer to the question "How long will this persist?" when there is a visible clinical or radiographic deformity.

Rockwood stated: *"Modern parents have become very sophisticated and expect a perfect outcome for their child. They inspect the radiographs and expect the alignment to be perfect, pressuring the surgeon toward operative intervention to obtain a perfect alignment."* [13]

Thus, closed reduction under anesthesia for moderately displaced or angulated forearm fractures is often performed unnecessarily. This leads to a prolonged time in the emergency department (ED) and cost increases. In contrast, acceptance of the angulation can potentially lead to the same result if sufficient years of growth remain [12].

Furthermore, closed reduction is not always successful, and re-displacement is seen in 45% of distal metaphyseal forearm fractures in children during the first few weeks after reduction [14].

Recently, the Dutch "Children's Fractures" Guideline was published, which recommends performing a closed reduction in children aged 0-5 years if angulation exceeds 25°, in children aged 5-10 years if angulation exceeds 15°, and in children older than 10 years if angulation exceeds 10-15° [15]. Also, fracture translation over 50% warrants a closed reduction. Despite these guidelines, the decision as to whether accept, manipulate or operate traumatic pediatric forearm fractures is often based on gut feeling and rarely on objective criteria [16].

Therefore, we designed a retrospective study to determine whether re-manipulation of re-displaced fractures in children improves long-term outcome or if re-displacements can be accepted, deeming current treatment guidelines too strict.

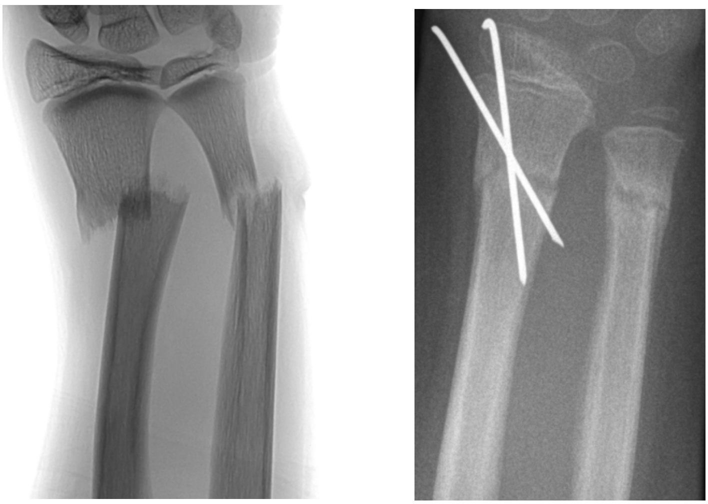
Figure 1. Example of remodeling of a re-displaced distal both-bone forearm fracture

Chapter 3: Do we need to stabilize all reduced metaphyseal both-bone forearm fractures in children with K-wires?

Fractures of both the radius and the ulna are often considered highly unstable, and therefore re-displacement is a recognized frequent complication. For example, in a series by Zamzam et al., both-bone forearm fractures were 23 times more likely to re-displace than isolated distal radius fractures [17]. Although these fractures are generally reduced and stabilized in a cast, additional K-wire fixation can be considered to prevent re-displacement.

In 2013, a randomized controlled trial (RCT) was published, which randomized children with a displaced metaphyseal, both-bone forearm fracture to closed reduction with or without additional K-wire fixation. Children treated without K-wire fixation had more re-displacements and greater limitation in forearm rotation at short-term follow-up than those treated with K-wire fixation [18]. Thus, K-wire fixation was recommended. Some authors even recommend the standard use of K-wires if a perfect anatomical reduction cannot be achieved [19]. A trend toward more operative management has been observed, despite no long-term outcomes studies showing superior results following operative treatment [20]. This zero-tolerance approach takes away the opportunity for spontaneous angulation correction, seen in pediatric fractures. The long-term follow-up of re-displaced fractures should be evaluated because it could change the treatment strategy. Hence, we analyzed the extended follow-up of an RCT with randomization between closed reduction with or without K-wire fixation to unravel if K-wire fixation is essential to prevent long-term sequelae or if nature is forgiving.

Figure 2. Example of K-wire fixation



PART II: Diaphyseal forearm fractures in children

Chapter 4: Long-term follow-up shows that early conversion to a below-elbow cast for reduced diaphyseal both-bone forearm fractures in children is safe

Some believe that in diaphyseal forearm fractures in children, early conversion to below-elbow casting (BEC) carries an increased risk of fracture re-displacement resulting in malunion. In contrast, others state that prolonged elbow immobilization in an above-elbow cast (AEC) might lead to soft tissue contractures. Regarding distal forearm fractures in children, Monga et al. previously advised changing practice and avoiding the discomfort and morbidity of unnecessarily immobilizing the elbow [21]. In 2013, a randomized controlled trial (RCT) was performed, including 127 children who underwent closed reduction due to a displaced diaphyseal forearm fracture. They were randomized to six weeks of above-elbow casting (AEC) or early conversion to below-elbow casting (BEC) after three weeks [22]. This study revealed no significant differences in re-displacement rates nor forearm rotation at short-term follow-up.

To verify if early conversion to BEC is safe for midshaft both-bone forearm fractures, even at long-term follow-up, we analyzed the extended follow-up of this RCT with a minimum 5-year follow-up.

Chapter 5: Both-bone forearm fractures in children: the outcomes of a prospective cohort of 316 patients with a mean follow-up of 7 years

Patient, fracture and treatment-related factors can be associated with long-term functional outcomes after both-bone forearm fracture in a child. Treatment can vary from cast immobilization alone to closed reduction with or without internal fixation by K-wiring or intramedullary pinning. During treatment, complications such as re-displacements, re-fractures, or re-operations can occur, which might deteriorate the clinical outcome. The clinical outcome after a both-bone forearm fracture is mainly influenced by pro-supination (forearm rotation) capability. The loss of pro-supination is correlated with the maximum angulation of the radius seen at the final follow-up [23].

Because the growing skeleton in children has remodeling capacity, assessing the long-term follow-up after both-bone forearm fractures in children is essential to determine the optimal treatment strategy. Reports on the long-term clinical and radiological outcomes after pediatric forearm fractures are rare. [24]. Therefore, we studied the long-term outcomes after pediatric both-bone forearm fractures to answer the following questions:

1) Which factors are associated with a persisting pro-supination limitation after pediatric both-bone forearm fractures?
2) Do accepted re-displacements of pediatric both-bone forearm fractures lead to inferior long-term outcomes?

PART III: Malunited forearm fractures in children

Chapter 6: Factors determining outcome of corrective osteotomy for malunited pediatric forearm fractures: a systematic review and meta-analysis

Re-displacement occurs in 34% of both-bone forearm fractures treated after closed reduction in a cast without additional fixation [25]. Such a re-displacement might result in a malunion with functional impairment. In 1962 Hughstone et al. quoted, *"In midshaft forearm fractures, growth will not correct angular deformity as it does in distal fractures"* [26]. Malunions in older children have less potential for remodeling, which can lead to disappointing clinical outcomes, especially a restriction in forearm rotation. A corrective osteotomy, a surgical intervention to restore normal bone alignment, may be considered for these patients. However, few articles have been published on the outcome after corrective osteotomy for malunited forearm fractures in children.

Therefore, we performed a meta-analysis of individual participant data (IPD) to provide the best available evidence on factors associated with a superior functional outcome after corrective osteotomy for malunited forearm fractures in children.

Figure 3. Example of a malunited forearm fracture

Chapter 7: Outcomes of 3D corrective osteotomies for pediatric malunited diaphyseal both-bone forearm fractures

A corrective osteotomy is often challenging due to angular deformities of the radius and the ulna involving all three dimensions [27]. Three-dimensional (3D) planning of the osteotomy and 3D printing of patient-specific instruments (PSIs) can potentially simplify the surgical procedure [28]. A 3D corrective osteotomy using PSIs is performed according to the following steps: A CT-based 3D model of the malunited forearm bones is superimposed on a mirrored version of the unaffected contralateral forearm bones; the location and degree of deformity are determined, and virtual cutting planes to perform the osteotomy are selected to match the contralateral side best; patient-specific drilling and cutting guides are designed, 3D-printed, sterilized and used during surgery.

Few studies examined the outcomes after 3D corrective osteotomy for pediatric diaphyseal forearm malunion. They were all heterogeneous, including both post-traumatic and congenital deformities or varying indications for surgery, such as distal radio-ulnar joint (DRUJ) instability.

We hypothesized that 3D corrective osteotomy could be essential in a patient who sustained a diaphyseal both-bone forearm fracture during childhood, leading to a symptomatic malunion with functional impairment in pro-supination. Therefore, we designed a prospective study on the outcomes after 3D corrective osteotomy for malunited pediatric diaphyseal both-bone forearm fracture. These inclusion criteria were chosen:

(1) Malunions after fractures sustained during childhood because of re-displacements during conservative management of forearm fractures in children;
(2) Diaphyseal forearm malunion because malunions located in the middle third cause more impairment in forearm rotation than the distal third and possess less remodeling capacity [29].
(3) Both-bone forearm malunion because complex 3D deformities of both forearm bones are more difficult to correct than radius deformity only;
(4) Limitation in pro-supination as an indication for surgery to ensure the comparability of the clinical outcomes after 3D corrective osteotomy.

To investigate if this innovative technology will improve clinical results, we determined what gain in forearm rotation can be achieved after 3D corrective osteotomy. Furthermore, we assessed which factors are associated with a superior outcome.

Chapter 8: Accuracy of 3D corrective osteotomy for pediatric malunited both-bone forearm fractures

In 2014 Jayakumar and Jupiter discussed the essential aspects regarding the reconstruction of malunited diaphyseal forearm fractures [30]. They stated that the entire forearm should be conceptualized as a single bicondylar articulation: the radio-ulnar joint. This means that a malunited fracture of the forearm should be considered intra-articular. Optimal treatment of a malunited (intra-articular) forearm fracture is aligned with the Arbeitsgemeinschaft für Osteosynthesefragen (AO) principles: restoration of bony anatomy, stable fracture fixation and preservation of blood supply with early mobilization. Angular deformities of the radius and ulna lead to bony impingement or tightness of the interosseous membrane. This can cause restriction in rotation of the radio-ulnar joint [30]. Likewise, axial rotational deformities lead to abnormalities in the proximal or distal radio-ulnar articulation, also causing restriction of forearm rotation. Forearm malunion is usually a combination of angular and rotational deformity. Due to the complex multiplanar deformity, a 3D corrective osteotomy can potentially help the surgeon to achieve a more accurate correction, which may result in a greater functional gain.

In 2015 Walenkamp et al. stated that for malunited distal radius fractures, numerous studies have shown that the accuracy of the anatomical reconstruction is essential to achieve an optimal outcome [31]. Several other authors have stated that anatomical correction of diaphyseal forearm malunions is highly desirable to achieve a good outcome[28,30].

However, for pediatric malunited forearm fractures, very few studies have examined the effectiveness of 3D corrective osteotomy using PSIs concerning the accuracy of the correction and its' relation to the gain in forearm rotation. None of these studies focused solely on diaphyseal both-bone forearm malunions after fractures sustained during childhood.

Therefore, we analyzed the radiographic accuracy of the achieved corrections in our series of 3D corrective osteotomies. Our primary outcome measures were the residual maximum deformity angle (MDA) and malrotation of the radius and ulna after 3D corrective osteotomy. A post-operative MDA > 5° or residual malrotation > 15° was defined as a non-anatomic correction. Our secondary outcome measure was the functional gain in pro-supination. Our main research questions were:

1) How often is an anatomic correction achieved by 3D corrective osteotomy for a pediatric malunited diaphyseal forearm fracture?
2) Does an anatomic correction provide a greater gain in pro-supination than a non-anatomic correction?

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PART I:
Distal metaphyseal forearm fractures in children

Chapter 2

Think twice before re-manipulating distal forearm fractures in children

Arch Orthop Trauma Surg. 2014 Dec;134(12):1699-707

Kasper Roth, Katharina Denk, Joost Colaris, Ruurd Jaarsma

ABSTRACT

Background:

Treatment of displaced pediatric distal forearm fractures is not always successful. Re-occurrence of angular deformity is a frequent complication. No consensus exists when to perform secondary manipulations. The purpose of this study was to analyze the long-term outcome of re-angulated pediatric forearm fractures to determine if re-manipulations can be avoided.

Methods:

Children who underwent closed reduction for distal forearm fractures and presented with re-angulation at follow-up were included in this retrospective cohort study. We compared those that were re-manipulated to those managed conservatively. Re-angulation was defined as ≥15° of angulation on either the AP or lateral view. Children were reviewed after 1-8 years post injury. Outcome measures were residual angulation on radiographs, active range of motion, grip strength, Visual Analogue Scales (satisfaction, cosmetics, pain), and the ABILHANDS-kids questionnaire.

Results:

Sixty-six children (mean age of 9.6 years) were included. Twenty-four fractures were re-manipulated and forty-two fractures had been left to heal in angulated position. At time of re-angulation, children <12 years in the conservative group had similar angulations to those re-manipulated. Children ≥12 years in the re-manipulation group had significantly greater angulations than children in the conservative group. At final follow-up, after a mean of 4.0 years, near anatomical alignment was seen on radiographs in all patients. Functional outcome was predominantly excellent. There was no significant difference in functional, subjective or radiological outcomes between treatment groups.

Conclusion:

Re-manipulation of distal forearm fractures in children <12 years did not improve outcomes, deeming re-manipulations unnecessary. Children ≥12 years in the conservative group achieved satisfactory outcomes despite re-angulations exceeding current guidelines. Based on observed remodeling, we now accept up to 30° angulation in children <9 years; 25° angulation in children aged 9 -<12; and 20° angulation in children ≥12 years, when re-angulation occurs. We conclude that clinicians should be more reluctant to perform re-manipulations.

INTRODUCTION

Distal forearm fractures are the most common injuries seen in pediatric traumatology, accounting for 40% of all fractures in children [1]. Severely angulated forearm fractures are generally reduced under general anesthesia or sedation and stabilized in a cast. Reduction is not always successful and re-displacement during the first few weeks after reduction is a frequent complication [2]. Rates of re-displacement have been reported to be between 7 and 91% [3]. A previous study revealed a re-displacement rate of 21.3% at our institute [4]. In case of re-displacement, especially re-angulation, the clinician is often confronted with a treatment dilemma: whether to perform a re-manipulation or to accept re-displacement and trust on correction by growth [3]]. This study focuses on the angular deformity of re-displacements and excludes the cases with solely a transitional or rotational aspect.

Earlier, Wilkins and O'Brien had suggested that dorsal angular deformities up to 30-35° will remodel adequately in children still having at least 5 growing years left [5]. More recently, it has been suggested that in children below 9 years, up to 20° of dorsal angulation or 15° of radial angulation will yield a good result. With increasing age, the degree of tolerable angulation decreases, recommending to accept up to 10-15° in children aged 9-13 years° and up to 5-10° in children aged 13-15 [6]. Controversy exists about the degree of angulation tolerable [7].

A recent trend toward increasingly more operative management has been observed, despite the fact that, to our knowledge, there have been no long term outcomes studies showing superior results following operative treatment [8-11]. Some authors even recommend the routine use of K-wires in cases where anatomical reduction cannot be achieved [12-15]. This zero-tolerance approach does not give the well-known spontaneous correction of angulation seen after fractures of long bones in children an opportunity [16; 17].

The long-term outcome of a re-displaced fracture has not yet been clarified [18]. Little attention has been paid to the outcome after re-manipulations [19]. Reports on clinical and radiological long-term results are altogether rare [20]. Due to the lack of consensus about and data on acceptable degrees of angulation, we developed a study with long-term follow-up. The purpose was to find whether re-manipulation of re-angulated fractures in children leads to an improved long-term outcome. We hypothesized that re-manipulations are often unnecessary.

MATERIALS AND METHODS

This retrospective cohort study was performed at a level 1 trauma institute. Ethics approval was obtained from the local medical ethics committee. A medical records search was performed to identify all children admitted with a distal forearm fracture between January 2005 and June 2012. Included in the study were: children who were ≤15 years old at the time of injury, who sustained a fracture of the distal third of the radius (with or without associated ulna fracture) which required closed reduction and subsequently presented with re-angulation at the initial follow-up. Re-angulation was defined as the progression of fracture angulation to greater than 15° on the lateral or posterior-anterior (PA) radiograph. Excluded were: non-displaced fractures; fractures that maintained satisfactory alignment after primary closed reduction; fractures initially treated by internal fixation; intra-articular fractures (Salter Harris); fractures treated by open reduction and open fractures. All included children were managed with an above elbow cast according to the institute's clinical management protocol.

Eligible patients were invited to revisit the orthopedic outpatients' clinic for long-term functional and radiographic assessment. Patients unable to attend were interviewed via telephone for subjective outcome. Informed consent was obtained from children's parents/guardians. All children voluntarily agreed to take part.

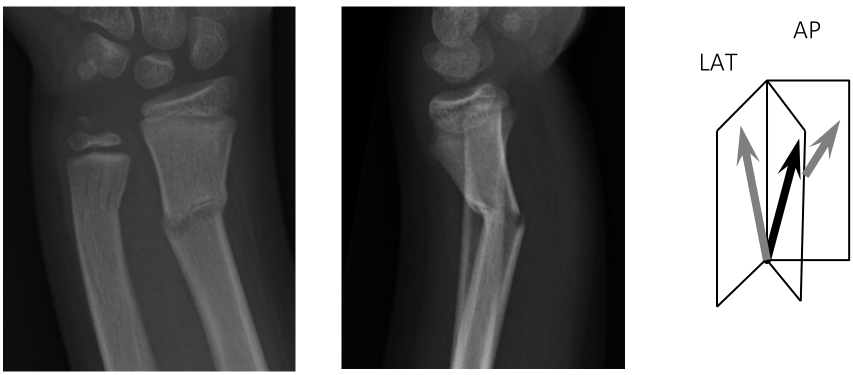
Patients were divided into two groups. The re-manipulation group consisted of patients, who underwent secondary closed reduction after re-angulation had occurred. The conservative group consisted of patients where re-angulations were accepted with the expectation that spontaneous correction by remodeling would occur. These patients were managed by casting alone and did not undergo a secondary closed reduction.

We classified our participants' angulated fractures into three categories of fracture types: A. Incomplete fractures, B. Complete fractures with bone contact and C. Complete fractures with 100% displacement. Presence of an associated ulna fracture was noted. We also investigated when re-angulation occurred, when re-manipulation was performed and what the total duration of treatment was in both treatment groups. Total duration of treatment was defined as time of injury until removal of cast.

One observer (first author) analyzed radiographs by measuring the degree and direction of angulation at the site of the fracture, using standard techniques [21]. Fracture angulation was analyzed at the time of trauma, re-angulation, post re-manipulation and final follow-up. The decision whether or not to re-manipulate was made at the time re-angulation was noticed. A method described by Ries et al. was used to determine the true angular deformity, which combines the findings of the PA and lateral radiograph [22; 23]. The maximum degree of angulation may occur in a plane other than the PA or lateral and the degree of true angulation can therefore be underestimated. True angulation was calculated with the formula given by Bär et al. [23]. Therefore, instead of presenting re-angulation as two findings (angular deformities on PA and lateral), radiographic results are presented as only one calculated finding. True angulation is demonstrated in Figure 1.

Figure 1 – True angulation

AP | **Lateral** | **True angulation**



13° radial angulation | 35° dorsal angulation | →37° true angulation

As remodeling potential decreases with increasing age [24; 25], radiographic results on angular deformity are subdivided into the following age categories: children <9 years old, children aged 9- <12 years and children aged ≥12 years.

To assess functional outcome, range of motion was measured using a goniometer and grip strength was measured using a JAMAR hydraulic hand dynamometer (Lafayette Instrument Company, Lafayette, IN, USA). To assess the subjective outcome, patient satisfaction regarding wrist function, cosmetic appearance and pain was documented using Visual Analogue Scales (VAS). The ABILHANDS-kids questionnaire was used to assess hand function in daily activities [26; 27]. Overall outcome was graded according to the criteria of Price et al. [8; 28]. A result was considered excellent if there were no complaints with strenuous physical activity and/or a loss of ≤10° of forearm rotation. A result was considered good if there were only mild complaints with strenuous physical activity and/or a loss of 11-30° forearm rotation. Fair results consisted of mild subjective complaints during daily activities and/or a 31-90° loss of forearm rotation. All other results were considered poor.

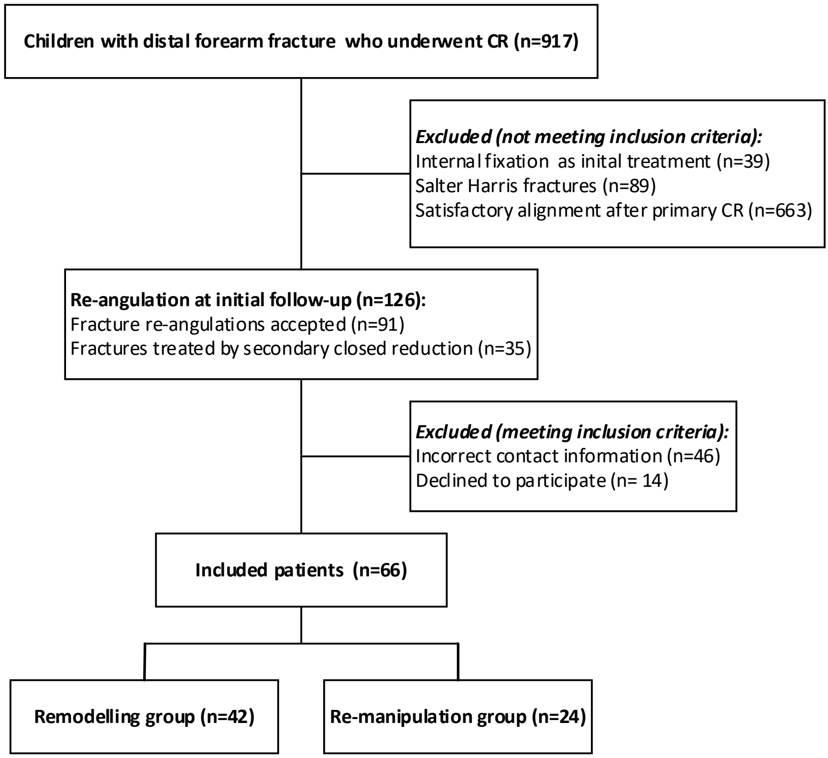
Statistical methods: Results are presented as means (± standard deviation). Chi-square test was used for analysis of patient demographics. Student's t-tests for independent samples, with equal variances not assumed, were performed to analyze differences in outcomes between groups. Fishers' exact test was used to compare overall outcome. One way analysis of variance (ANOVA) was performed to study the effect of age, after subdividing patients into age categories: <9 years, 9 - <12 years and ≥12 years. Remodeling capacity (re-angulation minus final residual angulation) was compared between age categories. Also, the effect of an associated ulna fracture was investigated.

Fracture angulation was re-measured in twenty cases by an independent trauma surgeon to confirm reproducibility of radiological assessment of fracture angulations (intra-class correlation). Statistical analyses were performed with Statistical Package for Social Sciences (SPSS) version 20.0 (SPSS Inc., Chicago, IL, USA). P-values < 0.05 were considered statistically significant.

RESULTS

Our search identified 917 children with a forearm fracture who underwent closed reduction. Re-angulation (≥15°) occurred in 126 patients (=14%), hereby meeting the inclusion criteria for enrolment (Figure 2).

Figure 2: Flowchart of Enrolment



Re-manipulation was performed in 35 children (=28% of 126), of whom 12 received additional internal fixation with K-wires. We included 66 children with a mean age of 9.6 years (±2.9) at the time of fracture. Table 1 shows the patient demographics, treatment chronology and fracture characteristics of the study population.

Table 1: Patient demographics/fracture characteristics of the study population

	Total	Re-manipulation	Conservative
Number of children	66	24	42
Clinically reviewed	39	16	23
Subjectively reviewed	27	8	19
Gender (% male)	56%	54%	57%
Age at trauma in years (mean± SD)	9.6 (±2.9)	9.8 (± 2.7)	9.3 (± 3.1)
Days until re-angulation	15 (±9)	11 (±4)*	17 (±11)*
Total days of treatment	46 (±15)	57 (±19)*	40 (±7)*
Final follow-up (in years)	4.0 (± 1.7)	4.8 (±1.6) *	3.6 (±1.7)*
Fracture characteristics:			
A. Incomplete # (%)	9.1	0	14.3
B. Complete with contact (%)	56.1	62.5	52.4
C. Complete, 100% displaced (%)	34.8	37.5	33.3
Associated ulna fracture (%)	53	38	62
Dominant arm fractured (%)	48	44	50

* Significant difference (p<0.05)

We reviewed the functional, radiological and subjective outcome of 39 patients clinically and the subjective outcome of an additional 27 patients via telephone. There was a mean follow-up of 4.0 years; 4.8 years in the re-manipulation group, 3.6 years in the conservative group. There was no significant difference between the groups in terms of age, gender and side or dominance of the injured extremity. Re-angulation occurred after a mean of 15 (±9) days post injury. Re-manipulation was performed after a mean of 11 (±4) days post injury. A significant difference in total duration of treatment was found in favor of the conservative group with a mean of 17 days shorter total treatment duration.

Comparison of radiological results between the two treatment groups are presented in Table 2. At time of injury fracture angulations were similar between the two groups. When re-angulation occurred (±15 days post injury), in the age category of <12 years there was no significant difference in angulation between fractures of the two treatment groups. In the age category of children ≥12 years, the re-manipulation group had significantly greater re-angulations than the conservative group. Re-manipulation was initially successful in all cases, but fractures healed with a mean residual angulation of 12° due to secondary re-angulation. This was significantly less than seen in children <12 years in the conservative group. No significant difference in angulation was seen between groups in children ≥12 years post re-manipulation. At final follow-up, near anatomical alignment was achieved in all patients and no significant difference was found in degree of angulation.

Table 2: Patient demographics/fracture characteristics of the study population

True angulation (±SD):	N (=66)	Trauma	At time of re-angulation	Post re-manipulation	Final follow-up (n=39)
Children <9 years					
Re-manipulation	8	26°± 13°	31°± 9°	12°± 7°	3°± 2° (n =7)
Conservative	21	31°± 11°	25°± 6°	X	1°± 2° (n=13)
P-value	-	0.36	0.12	0.00*	0.51
Children 9-12 years					
Re-manipulation	10	28°± 9°	26°± 7°	12°± 6°	1°± 2° (n=7)
Conservative	12	33°± 15°	21°± 5°	X	3°± 3° (n=5)
P-value	-	0.24	0.12	0.00*	0.37
Children ≥12 years					
Re-manipulation	5	28°± 20°	25°± 6°	15°± 9°	2°±2° (n=2)
Conservative	9	26°± 10°	19°± 3°	X	6°± 4° (n=5)
Significance	-	0.85	0.04	0.45*	0.14
Total					
Re-manipulation	24	27° ±13°	27° ±8°	12° ±7°	2° ±2°
Conservative	42	31° ±12°	23° ±6°	X	3°±3°
Significance	-	0.27	0.01	0.00*	0.21

* compared to angulation of conservative group at time of re-angulation.

In terms of functional outcome, there were no significant differences between the two groups at final follow-up, likewise when subdivided by age. Limitations in functional outcome were minimal and are presented in Table 3. Following the criteria of Price [28], there were 18 excellent, 4 good and 1 fair outcomes in the conservative group and 12 excellent, 3 good and 1 poor outcomes in the re-manipulation group. The patient with a poor outcome in the re-manipulation group had a progressive loss of strength of >50%, which caused moderate to severe complaints during daily activities. The fracture of this child was re-manipulated and fixated with K-wires. The patient with a fair outcome in the conservative group had ulnar-sided wrist pain due to positive ulnar variance requiring ulna shortening osteotomy at skeletal maturity. Both children with inferior outcomes were above 12 years of age. All others had a near full to full range of motion and grip strength and all had returned to normal activities without restrictions. Overall outcome was not significantly different between treatment groups (p=0.81).

Patients' subjective assessment of pain, function and cosmetics (VAS) are presented in Table 3 and demonstrated no significant difference between groups. The ABILHANDS-kids questionnaire (n=66) revealed a score of 40.8 (±3.0) in the re-manipulation group and a score of 41.0 (±1.9) in the conservative group (maximal score: 42). Patients subjectively reviewed had no significant differences in patient demographics or fracture angulations when compared to those clinically reviewed. Associated ulna fractures did not influence outcomes significantly.

The inter-reproducibility of the radiological assessment of the degree of true angulation showed an intra-class correlation range of 0.88 – 0.98.

Table 3: Data on limitation of range of motion, grip strength* and VAS scores

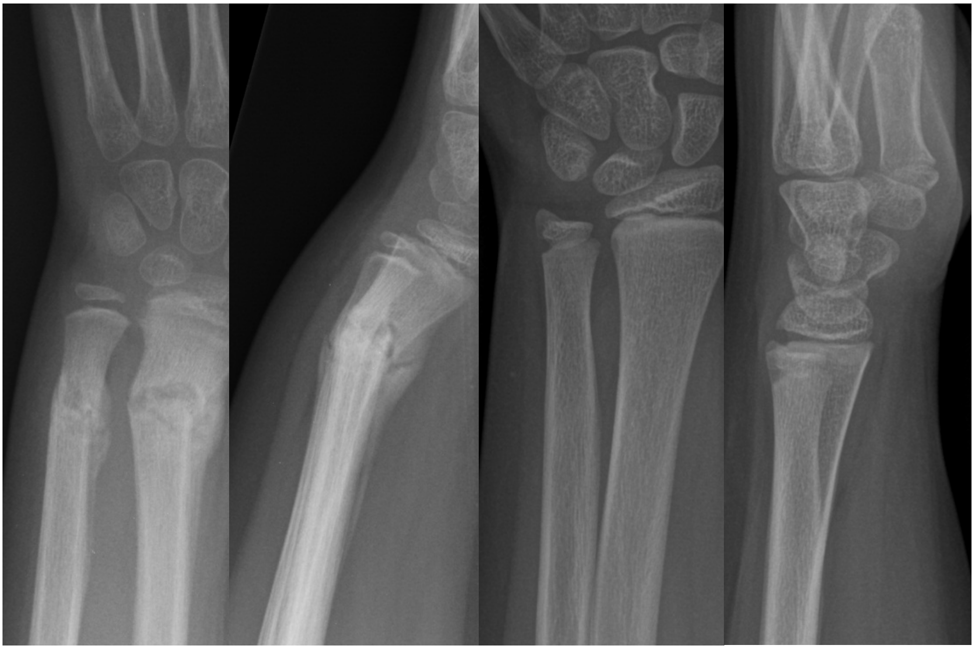
	Re-manipulation group (n=16)	Conservative group (n=23)
Loss of Pro-supination	4° (±5°)	6° (±6°)
Loss of Radial – ulnar deviation	5° (±7°)	5° (±7°)
loss of Wrist flexion/extension	2° (±4°)	2° (±6°)
Grip strength (in kg)	3 (±6)	1 (±3)
VAS Satisfaction**	8.8 (±2.0)	9.2 (±1.3)
VAS Cosmetic appearance**	9.4 (±1.1)	9.0 (±1.6)
VAS Pain**	0.8 (±1.4)	1.2 (±1.4)

* Limitation is in comparison to the contralateral arm.
** VAS scores (in cm) ranging from 0 – 10 cm (with 0 being the best and 10 the worst)
There are no statistically significant differences between the groups

DISCUSSION

The purpose of this study was to analyze the effect of manipulating re-angulations of initially reduced pediatric distal forearm fractures on the long-term outcome. We hypothesized that re-manipulations are often unnecessary. At final follow-up, near anatomical alignment was achieved in all patients and no significant difference was found in residual angulation between the treatment groups, despite the fact that the conservative group had greater residual angulation than the re-manipulation group. At final follow-up both groups performed just as well in terms of functional and subjective outcomes. Figure 3 demonstrates the power of remodeling over time.

Figure 3: Example of remodeling in radiological follow-up



Degree of remodeling (8 weeks) Degree of remodeling (2.5 years)

One of the factors affecting the decision whether or not to re-manipulate is evidently the degree of angulation. Our study revealed that children <12 years old did not differ significantly in degree of fracture re-angulation initially, when the decision whether or not to re-manipulate was made. However they still received different types of treatment (one being more invasive). This clearly indicates that when opting for re-manipulation, not only the severity of angulation, but also surgeons' preferences and parents' opinions are taken into account in the decision-making process whether to manipulate or not.

At final follow-up, our study did not demonstrate superior radiological, functional or subjective outcomes in the re-manipulation group. Therefore, re-manipulations in children <12 years would seem unnecessary, as fracture re-angulations did not vary significantly within this age category. In children ≥12 years, re-angulations were more severe in the re-manipulation group. This reveals that especially in the older children, severity of angulation plays an important part in the decision whether or not to perform secondary manipulation. As expected and in accordance with the literature, the capacity for remodeling at the fracture site was greater in the younger children than in the older children [29]. However, the degree of secondary angulation seen in the conservative group did exceed the amount considered tolerable and nevertheless, satisfactory results were still achieved. This deems guidelines too strict.

Only a few randomized controlled trials have compared functional outcomes following closed reduction and cast immobilization versus percutaneous pin fixation of angulated distal radius or both bone forearm fracture in children thus far [3; 30; 31]. Two RCTs found no significant difference in functional outcome after a mean period of approximately 3 months [30; 31] and one randomized controlled trial showed a significantly lower rate of loss of pronation/supination after percutaneous pin fixation of forearm fractures at 6 months follow-up [3], whereas our study shows predominantly excellent functional outcomes after a mean period of 4.0 years. This highlights that remodeling takes place over a long period of time and functional outcome can be restored in due time. Thereby, Zimmerman et al. also found that in children < 10 years large dislocations at the time of fracture healing do not influence the 10-year functional outcome and that repeated reduction of fractures produced significantly poorer results in the long term [20]. Furthermore, Price et al. studied the outcome of angulated pediatric forearm fractures after a mean follow-up of 5.8 years and found 32 excellent, 4 good, 3 fair and 0 poor outcomes [28]. Using the same grading system, we found similar results in overall outcome of fractures left to correct by remodeling.

Our findings suggest that the criteria of published guidelines recommending when to manipulate pediatric forearms fractures are too strict. This is supported by other studies: despite protocols suggesting to re-manipulate all fractures that fail to maintain these reduction parameters, only 51% of these children received secondary manipulation, found in two impartial studies [3; 18] and a recent study by Asadollahi et al. concludes that only a small number of fractures that lose reduction require a second intervention [32]. Reasons for clinicians to avoid (re-)manipulations of children's fractures are mainly based on risks associated with anesthesia [33-37]. Moreover, the treating surgeon may expect correction of the malunion by growth, may be reluctant to burden the child again and prolong the period of casting, or may find it difficult to accept failure of the initial treatment. In our study a delay of 17 days in total duration of treatment was seen in the re-manipulation group, causing extra discomfort and interference with daily activities without accomplishing superior outcomes.

Limitations

Our study had some limitations. In our study, all included children were managed with an above elbow cast according to our institute's former clinical management protocol. Recent literature [38-44] provides insights that below-elbow casting (BEC) is not inferior to above-elbow casting (AEC) and should be considered first-choice for conservative treatment. A recent meta-analysis by Hendrickx et al., updated by Bekerom et al. in 2012 including 5 randomized controlled trials comparing AEC versus BEC for the treatment of distal third forearm fractures in children had the following results: BEC had significantly fewer loss of reduction (OR 0.44 (0.24-0.82)); there was no significant difference in the number of performed re-manipulations (OR 0.64 (0.34-1.20)); there was no significant difference in plaster-related complications (OR 0.60 (0.42-1.12)) and children treated with BEC missed less school days and encountered less difficulties in daily living. In the interim, our protocol has been updated and we have implemented the use of below-elbow casting to treat metaphyseal distal radius or both-bone forearm fractures.

Due to the retrospective nature of this research, recruitment rates were modest. Geographic dispersion of the study population meant that 27 out of 66 children were unable to revisit clinics. Patients who could not revisit clinics did not significantly differ from those who were clinically reviewed in terms demographics, baseline fracture angulations and long-term subjective outcomes. Patients clinically reviewed thus represent a good sample of the homogenous total group of participants. Franklin et al. suggested that the ideal study to aid in evidence-based decision-making for pediatric distal forearm fractures would be a randomized controlled trial comparing cast immobilization and closed reduction versus operative management, in children aged older than 8 years with distal metaphyseal forearm fractures with angulation ≥20°, subdivided for fracture classification, with a minimum of 5 years of follow-up, studying the final functional outcome, defined as pronation and supination at final presentation [10] . In our opinion, the treatment option of below-elbow cast immobilization without closed reduction in children up to 12 years of age should be included in this ideal RCT.

The mean age for ossification of the physis differs between boys and girls (14.5 and 12.9 years, respectively) [45] which suggests a divergence in remodeling capacity especially in the oldest group. We did not detect a gender difference in remodeling capacity within this group, though statistical power might not have been strong enough. Numbers of males and females were however homogenous within all 3 groups, which balanced potential differences.

A difference in length of follow-up between the two groups was seen. Mean follow-up was 4.8 years in the re-manipulation group compared to 3.6 years in the conservative group. Yet, this reinforces our hypothesis, because the shorter follow-up period disadvantaged the conservative group in its remodeling potential.

Lastly, the clinical applicability of “true angulation” requires further investigation.

CONCLUSIONS

As a result of our findings, when re-angulation occurs at our institute, we now accept up to 30° true angulation in children <9 years; up to 25° true angulation in children aged 9 - <12; and 20° true angulation in children ≥12 years. We based these recommendations on our observed range of angulations within 1 SD from the mean of each age category which lead to predominantly excellent outcomes. If these recommendations would have been implemented beforehand, only three patients in the conservative group and nine patients in the re-manipulation group would have been re-manipulated. This would decrease the amount of re-manipulations performed by 50% without, to our beliefs, compromising outcomes. Our results provide yet another piece of evidence to justify this non-invasive management approach preferred by many clinicians.

We conclude that re-manipulation of re-angulated pediatric distal forearm fractures in children <12 years does not provide an improved 4-year outcome as compared to conservative management. Children ≥12 years also demonstrated to exceed the expected remodeling capacity and achieved satisfactory outcomes. Therefore, we recommend to accept up to 30° true angulation in children <9 years; up to 25° true angulation in children aged 9 -<12; and 20° true angulation in children ≥12 years. We believe that the clinician's reluctance to perform re-manipulations can be justified and suggest thinking twice before re-manipulating children's forearm fractures in clinical practice.

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Chapter 3

Do we need to stabilize all reduced metaphyseal both-bone forearm fractures in children with K-wires?

Clin Orthop Relat Res. 2022 Feb 1;480(2):395-404.

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The first two authors contributed equally to this manuscript.

ABSTRACT

Background:
Short-term follow-up studies have shown that reduced metaphyseal both-bone forearm fractures in children should be treated with K-wires to prevent re-displacement and inferior functional results. Minimum 5-year follow-up studies are limited. Range of motion, patient-reported outcome measures and radiographic parameters at minimum 5-year follow-up should be evaluated because they could change insights into how to treat pediatric metaphyseal forearm fractures.

Questions/purposes:
1) Does K-wire stabilization of reduced metaphyseal both-bone forearm fractures in children provide better forearm rotation at minimum 5-year follow-up?
2) Do malunions (untreated re-displaced fractures) of reduced metaphyseal both-bone forearm fractures in children induce worse functional results?
3) Which factors lead to limited forearm rotation at minimum 5-year follow-up?

Methods:
We analyzed the extended minimum 5-year follow-up of a randomized controlled trial in which children with a reduced metaphyseal both-bone forearm fracture were randomized to either an above-elbow cast (Casting group) or fixation with K-wires and an above-elbow cast (K-wire group). Between January 2006 and December 2010, 128 patients were included in the original randomized control trial: 67 in the Casting group and 61 in the K-wire group. For the current study, based on an a priori calculation it was determined that, with an anticipated mean limitation in pro-supination (forearm rotation) of 7° ± 7° in the Casting group and 3° in the K-wire group, a power of 80% and a significance of 0.05, the two groups should consist of 50 patients each. Between January 2014 and May 2016, 82% (105 of 128) patients were included with a mean follow-up of 6.8 ± 1.4 years: 54 in the Casting group and 51 patients in the K-wire group. At trauma, patients had a mean age of 9 ± 3 years and had mean angulations of the radius and ulna of 25° ± 14° and 23° ± 18°, respectively. The primary result was limitation in forearm rotation. Secondary outcome measures were radiologic assessment, patient-reported outcome measures (QuickDASH and ABILHAND-kids), handgrip strength and VAS score for cosmetic appearance. Assessments were performed by the first author (unblinded). Multivariable logistic regression analysis was performed to analyze which factors led to a clinically relevant limitation in forearm rotation.

Results:
There was a mean limitation in forearm rotation of 5° ± 11° in the Casting group and 5° ± 8° in the K-wire group with a mean difference of 0.3° (95% CI -3° to 4°; p = 0.86). Malunions occurred more often in the Casting group than in the K-wire group: 19% (13 of 67) versus 7% (4 of 61) with an OR of 0.22 for K-wiring (95% CI 0.06 to 0.80; p = 0.02). In patients in whom a malunion occurred (Malunion group), there was a mean limitation in forearm rotation of 6° ± 16° versus 5° ± 9° in patients who did not have a malunion (Acceptable alignment group), with a mean difference 0.8° (95% CI -5° to 7°; p = 0.87). Factors associated with a limited forearm rotation ≥ 20° were a malunion after above-elbow casting (OR 5.2 [95% CI 1.0 to 27] ; p = 0.045) and a refracture (OR 7.1 [95% CI 1.4 to 37] ; p = 0.02).

Conclusions:
At a minimum of 5 years after injury, in children with a reduced metaphyseal both-bone forearm fracture, there were no differences in forearm rotation, patient-reported outcome measures nor radiographic parameters between patients treated with only an above-elbow cast compared with those treated with additional K-wire fixation. Re-displacements occur more often if treated by an above-elbow cast alone. If fracture re-displacement is not treated promptly, this leads to a malunion, which is a risk factor for a clinically relevant (≥ 20°) limitation in forearm rotation at minimum 5-year follow-up. Children with metaphyseal both-bone forearm fractures can be treated with closed reduction and casting without additional K-wire fixation. Nevertheless, a clinician should inform parents and patient about the high risk of fracture re-displacement, (and therefore malunion) with risk for limitation of forearm rotation if left untreated. Weekly radiographic monitoring is essential. If re-displacement occurs, re-manipulation and fixation with K-wires should be considered based on gender, age and direction of angulation. Future research is required to establish the influence of (skeletal) age, gender, and the direction of malunion angulation on clinical outcome.

Level of Evidence:
Level I, therapeutic study.
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INTRODUCTION

Background

Reduced metaphyseal both-bone forearm fractures have been shown to re-displace in a cast in up to 46% [9, 26] and have a 3.6 to 23 times higher risk for re-displacement than isolated distal radius fractures [16, 31]. In 2013, we published [9] a randomized controlled trial (RCT) that included 128 children with a reduced stable metaphyseal both-bone forearm fracture who were randomized to an above-elbow cast with or without percutaneous K-wire fixation. Children treated with an above-elbow cast alone had a higher risk of re-displacement and a higher risk of limiting pro-supination (forearm rotation) than children who had additional K-wire fixation, after a mean follow-up of 7 months. Thus, pinning of apparently stable both-bone distal forearm fractures in children was recommended to prevent re-displacement [9].

Rationale

There has been a recent increase in operative management to treat fractures in children, despite the fact that there have been no long-term outcome studies showing superior results following operative treatment [12, 14]. As mentioned, the goal of operative treatment is to prevent re-displacement. If re-displacement of a metaphyseal forearm fracture occurs after conservative treatment, a clinician has two options: to reduce the fracture again (with or without K-wire fixation) or to accept malunion and hope that the remodeling that occurs during growth will result in acceptable cosmetics and function (Fig. 1) [25]. Tremendous remodeling is especially apparent in young children (younger than 10 years) with a distal fracture near the most active growth plate [19, 27, 30]. Treatment discussion is ongoing about what degree of malunion results in an acceptable long-term clinical result [23, 32, 33]. Minimum 5-year follow-up should be evaluated because it could change insights into the treatment of pediatric metaphyseal forearm fractures.

Therefore, we asked:

1) Does K-wire stabilization of reduced metaphyseal both-bone forearm fractures in children provide better forearm rotation at minimum 5-year follow-up?
2) Do malunions (untreated re-displaced fractures) of reduced metaphyseal both-bone forearm fractures in children induce worse functional results?
3) Which factors lead to limited forearm rotation at minimum 5-year follow-up?

Figures 1A-F

These sagittal radiographs are from a patient with a displaced metaphyseal both-bone forearm fracture, including:

(Fig. 1-A) an initial radiograph of the fracture,

(Fig. 1-B) after reduction,

(Fig. 1-C) re-displacement after 10 days,

(Fig. 1-D) 25 days after trauma,

(Fig. 1-E) 5 months after trauma, and

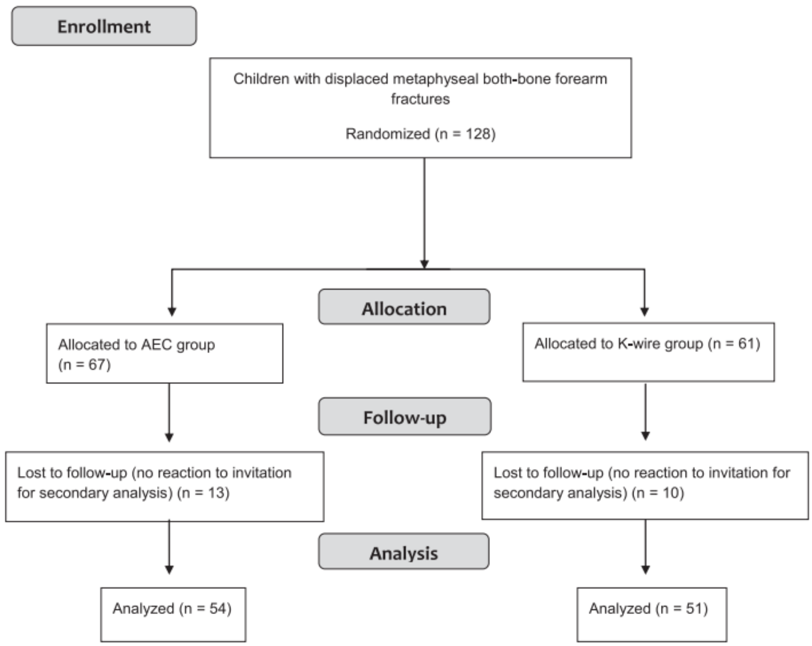
(Fig. 1-F) 7.5 years after trauma.

PATIENTS AND METHODS

Study Design and Setting

We report the extended follow-up of a published RCT with a minimum follow-up of 5 years. Children with a displaced metaphyseal both-bone forearm fracture were included in one of four participating Dutch hospitals: Erasmus Medical Center (Rotterdam), Haga Hospital (The Hague), Reinier de Graaf Hospital (Delft) and Franciscus Hospital (Rotterdam). Our initial institutional review board protocol did not specify another follow-up moment 5 years later. However, after finishing data collection of the original RCT, we thought this would be informative and initiated the current extended follow-up study. In the published RCT, between January 2006 and December 2010, 128 patients were included (67 in the Casting group and 61 patients in the K-wire group). For the current study, we invited all 128 patients to revisit the outpatient department. Between January 2014 and May 2016, 82% (105 of 128) patients were included: 54 of the Casting group and 51 of the K-wire group (CONSORT flowchart of enrollment is supplied in Fig. 2).

Figure 2: This CONSORT study flow diagram demonstrates the selection and flow of patients:



Participants

In the original RCT, we included children younger than 16 years who had a displaced metaphyseal fracture of the distal radius and ulna. We included only children with a displaced forearm fracture that was stable after closed reduction in the operating room. The criteria for fracture reduction were defined a priori: a fracture was reduced if radius and/or ulna showed displacement on a posteroanterior and/or lateral radiograph. Fracture displacement was based on angulation (> 15° for children aged younger than 10 years and > 10° for children between 10 to 16 years) and/or translation (more than half bone diameter) and/or any rotation. Fracture re-displacement was defined by the loss of reduction (angulation and/or translation) according to these primary reduction criteria [9]. Based on the occurrence of re-displacement, we divided all included patients into two additional groups (Malunion and Acceptable alignment group). Malunion was defined as the occurrence of fracture re-displacement, meeting the above-mentioned criteria for reduction, but was left untreated (contrary to RCT protocol) and thus consolidated in a malunited position.

Description of Treatment

All included patients underwent closed reduction. Thereafter, the fracture was tested for stability. The fracture was defined as unstable if full pronation and supination of the forearm caused re-displacement [9]. Unstable fractures were excluded and were treated with K-wire fixation. The remaining fractures were defined as stable and were randomized between above-elbow casting alone (Casting group) or K-wire pinning with an above-elbow cast (K-wire group), both for 4 weeks.

Randomization

In the published RCT, participants were randomly assigned and treated in the Casting or K-wire group. An independent clinician randomized the children by sealed envelopes with varied block sizes. The children, parents and clinicians were not blinded for randomization. For the current RCT, we obtained informed consent from all parents and all children aged at least 12 years. Patients unable to attend were, if possible, interviewed via telephone to complete patient-reported outcome measure questionnaires.

Variables, Outcome Measures, Data Sources, and Bias

Our primary outcome measure was limitation in pro-supination (forearm rotation) compared with the contralateral side. Secondary outcome measures were radiologic assessment; patient-reported outcome measures, including the Dutch version of the QuickDASH questionnaire and ABILHAND-kids questionnaire [2, 3, 6] handgrip strength percentage of the contralateral side; and the VAS score for cosmetic appearance.

One unblinded orthopedic surgeon (JWC) examined all patients during short-term follow-up (mean of 7.1 months) after the initial trauma for the original RCT [9]. A second independent orthopedic surgeon (LWD) examined all patients at minimum 5-year follow-up (unblinded). Forearm rotation was evaluated using a standardized procedure: visual estimation and a two-increment goniometer [8]. Handgrip strength was measured using a JAMAR dynamometer (Lafayette Instrument Company, Lafayette, IN, USA). Cosmetic appearance (forearm morphology and possible scars) was assessed by the first author (LWD) and either by the patient or by the parent, if the patient was younger than 17 years. This VAS was scored in the traditional way on a 10-cm line [13]. A score of 10 was defined as cosmetically best. Radiographic examination was performed. One of the authors (PE) measured the radiologic intramedullary angulation of the radius and ulna on posteroanterior and lateral radiographs taken at the time of cast removal (consolidation) and at final follow-up [17]. Radiographic angulation was remeasured in 25 patients by the primary author (LWD) to assess reproducibility.

Ethical Approval

Our institutional review board approved this post-trial follow-up study, which was registered under protocol number NL41839.098.12. The original RCT [9] was registered in ClinicalTrials.gov with registry identifier NCT 00397852.

Statistical Analysis, Study Size

In the previous RCT, after a mean follow-up of 7 months, a limitation of forearm rotation of 14° ± 14° was seen in the Casting group and 7° ± 9° in the K-wire group [9]. We expected that over time, limitation of pro-supination would decrease with approximately 50% at minimum 5-year follow-up. With an a priori calculation, we determined that with an anticipated mean limitation in pro-supination of 7° ± 7° in the Casting group and an anticipated mean limitation in pro-supination of 3° ± 5° in the K-wire group, a power of 80% and a significance of 0.05, the two groups should consist of 50 patients each.

It was established whether the variables had a normal distribution using the normality Shapiro–Wilk test. Based on these analyses, the results are presented as means ± SD, mean difference (95% confidence interval and p values. Patient demographics included for minimum 5-year follow-up were compared between the study groups (Casting group versus K-wire group) using the independent samples t-test (Table 1).

Radiographic and functional results were analyzed using independent samples t-test comparing the Casting group versus K-wire group (Table 2) and comparing patients in whom a malunion occurred (Malunion group) with patients who did not have a malunion (Acceptable alignment group) (Table 3). To assess the interrater reproducibility of the radiographic assessment, we calculated the intraclass correlation coefficient (Type C).

Multivariable logistic regression analysis was performed to analyze which factors led to a clinically relevant limitation in forearm rotation at minimum 5-year follow-up, defined as a limitation of forearm rotation ≥ 20° (as dependent variable), a cutoff point which has been used previously [10]. The following factors were included in our exploratory analysis (univariate logistic regression): intervention (Casting group versus K-wire group); occurrence of a malunion (Malunion versus Acceptable alignment group) and occurrence of a refracture (group versus non refracture group), age at trauma (age younger than 10 years versus age 10 years or older) and sex (male versus female). A p value < 0.05 during univariate logistic regression was used as a threshold to determine which factors progressed to the more definitive multivariable logistic regression analysis. Statistical analyses were performed using IBM SPSS Statistics, version 23 (SPSS Inc., Chicago, IL, USA).

Patient Demographics

Of the patients who were included in the original RCT [9], 82% (105 of 128) participated in the current study. Fifty-four of the original 67 participants who were allocated to the Casting group and 51 of the original 61 participants who were allocated to K-wire fixation participated. Eighteen percent (23 of 128) of patients were lost to follow-up. The mean length of follow-up was 6.8 ± 1.4 years. Baseline characteristics were similar between the groups (Table 1). At trauma, patients had mean angulations of the radius and ulna of 25° ± 14° and 23° ± 18°, respectively. The interrater reliability of the radiologic measurement had an intraclass correlation of 0.83 (95% CI 0.57 to 0.94).

In the original RCT, in the Casting group, re-displacement occurred in 30 patients in the first weeks after trauma, 17 of whom underwent re-manipulation (six received additional K-wire fixation) and 13 of whom accepted re-displacement (the Malunion group). Eighty-three percent (25 of 30) of patients with re-displacements were available for minimum 5-year follow-up. In this group of 25 patients, 14 patients underwent re-manipulation, and 11 patients accepted the re-displacement (the Malunion group). Refractures occurred in 11 of 128 patients, nine of whom were reevaluated at final follow-up.

Table 1. Patient demographics

Characteristic	Casting group (n = 54)	K-wire group (n = 51)	Mean difference (95% CI)	p-value
Age at trauma	9 ± 3	9 ± 3	-0.4 (-1.6 to 0.8)	0.49
Sex (% male)	61 (33)	69 (35)	7.5% (-11 to 26)	0.43
Dominant arm	52 (28)	45 (23)	6.8% (-13 to 26)	0.49
Fracture type radius				
Complete	76 (41)	84 (43)	19.1% (1 to 37)	0.04
Greenstick	24 (13)	16 (8)	-19.1% (-37 to -1)	0.04
Fracture type ulna				
Complete	44 (24)	47 (24)	1.3 (-1.8 to 2.1)	0.89
Greenstick	50 (27)	45 (23)	-4.9 (-24 to 15)	0.62
Torus	6 (3)	8 (4)	3.6 (-3.9 to 11)	0.34
Angulation radius, °	27 ± 16	23 ± 15	4.6 (-0.9 to 10)	0.10
Angulation ulna, °	25 ± 21	20 ± 13	5.0 (-1.7 to 12)	0.15

Data presented as % (n) or mean 6 SD, unless noted otherwise.

RESULTS

Does K-wire Stabilization of Reduced Metaphyseal Both-bone Forearm Fractures in Children Provide Better Forearm Rotation at Minimum 5-year Follow-up?

K-wire stabilization of reduced metaphyseal both-bone forearm fractures in children did not provide better forearm rotation at minimum 5-year follow-up. There was a mean limitation in pro-supination in the K-wire group of 5° ± 8° and a mean limitation of 5° ± 11° in the Casting group with a mean difference of 0.3° (95% CI -3° to 4°; p = 0.86) (Table 2). Radiographic results were similar. There was less residual angulation of the radius in the coronal plane in the Casting group (4° [95% CI 3° to 5°) than in the K-wire group (5° [95% CI 4° to 6°], mean difference -1° (95% CI -3° to -0.4°; p = 0.04). We found no differences in patient-reported outcome measures (QuickDASH and ABILHAND-kids), VAS score for cosmetics, and handgrip strength (Table 2).

Table 2. Radiographic and functional results (Casting vs K-wire group)

Radiographic outcomes		**Casting group (n = 67)**	**K-wire group (n = 61)**	**Mean difference (95% CI)**	**p value**
Consolidation[a]	Radius – PA	8° ± 7°	6° ± 4°	2° (-0.6° to 4°)	0.12
	Radius - Lateral	13° ± 10°	8° ± 4°	5° (2° to 9°)	0.01
	Ulna - PA	7° ± 4°	6° ± 4 °	1° (-0.7° to 3°)	0.25
	Ulna - lateral	7° ± 5°	7° ± 5°	0.5° (-2° to 3°)	0.67
		(n = 54)	**(n = 51)**		
7-year follow-up	Radius – PA	4° ± 3°	5° ± 3°	-1° (-3° to -0.4°)	0.04
	Radius - lateral	4° ± 3°	4° ± 3°	-0.4° (-2° to 0.9°)	0.52
	Ulna - PA	5° ± 3°	5° ± 3°	-0.3° (-2° to 1°)	0.68
	Ulna - lateral	3° ± 3°	4° ± 3°	-1° (-3° to 0.2°)	0.08
Functional outcomes					
7- year follow-up	Limitation in pro-supination	5° ± 11°	5° ± 8°	0.3° (-3° to 4°)	0.86
	QuickDASH	5.8 ± 11	3.4 ± 5	2.4 (-1.0 to 5.8)	0.16
	ABILHAND	41 ± 2	42 ± 1	-0.5 (-1.1 to 0.8)	0.09
	Cosmetics (patient)	8.3 ± 2	7.8 ± 3	0.5 (-0.4 to 1.4)	0.29
	Cosmetics (clinician)	8.7 ± 2	8.1 ± 2	0.6 (-0.2 to 1.4)	0.17
	Hand grip strength %	99 ± 21	100.0 ± 18	-1.8 (-9.6 to 6.0)	0.64

[a]Data in these rows are from a prior publication [9]; PA = posteroanterior.

Do Malunions of Reduced Metaphyseal Both-Bone Forearm Fractures in Children Induce Worse Functional Results?

Malunions of reduced metaphyseal both-bone forearm fracture in children occurred more often in the Casting group than the K-wire group at short-term follow-up: 19% (13 of 67) versus 7% (4 of 61) with an odds ratio of 0.22 for K-wires (95% CI 0.06 to 0.80; p = 0.02). At minimum 5-year follow-up, there was a mean limitation of forearm rotation of 6° ± 16 in the Malunion group versus 5° ± 9° in the Acceptable alignment group, with a mean difference of 0.8 (95% CI -5° to 7°; p = 0.87). Angulation of the ulna in the sagittal plane was less in the Malunion group (1° [95% CI -0.8° to 3°]) than in the Acceptable alignment group (4° [95% CI 3° to 5°]), with a mean difference of -3° (95% CI -5 to -0.4°; p = 0.02). Patient-reported outcomes (QuickDASH and ABILHAND-kids), cosmetic appearances scores, and grip strength were not different (Table 3).

Table 3. Radiographic and functional results (Malunion vs Acceptable alignment group)

		Malunion group (n = 13)	Acceptable alignment group (n = 115)	Mean difference (95% CI)	p value
Radiographic outcomes					
Consolidation[a]	Radius - PA	15° ± 7°	6° ± 4°	9.6° (4.0° to 15°)	< 0.001
	Radius - lateral	17° ± 6°	9° ± 8°	7.2° (1.5° to 13°)	0.01
	Ulna - PA	7° ± 5°	6° ± 4°	1.6° (-1.2° to 4.3°)	0.26
	Ulna - lateral	10° ± 7°	6° ± 5°	3.4°(-1.0° to 6.9°)	0.06
		(n = 11)	(n = 94)		
7-year follow-up	Radius - PA	5° ± 3°	5° ± 3°	-0.1° (-2.4° to 2.1°)	0.91
	Radius - lateral	4° ± 3°	4° ± 3°	0.3° (-1.7° to 2.3°)	0.76
	Ulna - PA	5° ± 3°	5° ± 3°	0.01° (-2.2° to 2.2°)	0.99
	Ulna - lateral	1° ± 2°	4° ± 4°	-2.8° (-5.2° to -0.4°)	0.02
Functional outcomes		(n = 11)	(n = 94)		
7-year follow-up	Limitation in prosupination	6° ± 16°	5° ± 9°	0.8° (-5.2° to 6.9°)	0.87
	QuickDASH	3.4 ± 6	4.6 ± 9	-1.3 (-6.8 to 4.2)	0.64
	ABILHAND	41 ± 2	41 ± 2	0.01 (-1.0 to 1.1)	0.98
	Cosmetics (patient)	8.0 ± 2	8.3 ± 2	-0.2 (-1.5 to 1.1)	0.58
	Cosmetics (clinician)	8.6 ± 1	8.7 ± 2	0.2 (-1.0 to 1.4)	0.76
	Hand grip strength %	98 ± 15	99 ± 20	-1.0 (-14 to 12)	0.88

Which Factors Lead to Limited Forearm Rotation of More than 20°?

At minimum 5-year follow-up, two factors were associated with a clinically relevant limitation in forearm rotation of ≥ 20°: occurrence of a malunion after above-elbow casting (OR 5.2 [95% CI 1.0 to 27]; p = 0.045) and a refracture (OR 7.1 [95% CI 1.4 to 37]; p = 0.02). Limitation in forearm rotation ≥ 20° was seen in the Malunion group in 27% (3 of 11) versus 7% (7 of 94) in the Acceptable alignment group. Also, this limitation was seen in 33% (3 of 9) of patients in whom a refracture occurred versus in 7% (7 of 96) of patients without a refracture (Table 4). Sex and age at trauma older than 10 years were not associated with a limitation in forearm rotation ≥ 20° at minimum 5-year follow-up during exploratory univariate logistic regression analysis (p values of 0.11 and 0.49, respectively).

Table 4. Multivariable logistic regression analysis

Subgroup	≥ 20° of limitation	Odds ratio (95% CI)	p-value
Malunion group	27% (3 of 11)	5.2 (1.0-27)	0.045
Nonmalunion group	7% (7 of 94)		
Refracture	33% (3 of 9)	7.1 (1.4-37)	0.02
No refracture	7% (7 of 96)		

Factors associated with limitation in forearm rotation of ≥20° at minimum 5-years follow-up.

DISCUSSION

Background and Rationale

Displaced metaphyseal both-bone forearm fractures in children, which are stable after closed reduction show high risk of re-displacement in a cast, which can cause malunion and limitation in forearm rotation [9, 26]. Re-displacement can be prevented by K-wire stabilization. To determine if K-wire stabilization is essential for all reduced metaphyseal both-bone forearm fractures in children or that such malunions will correct by growth, we reassessed ROM, patient-reported outcome measures, and radiographic parameters of patients included in our previous RCT after a minimum of 5-year follow-up.

Limitations

A key limitation is that we could not include enough patients to perform a powerful multivariable analysis including more potentially relevant factors, such as the direction of malunion angulation and degree of initial displacement, but also, we could not adequately control for patient's age and sex. Concerning direction of malunion angulation, Roberts et al. [24] demonstrated that radial deviation is more closely related to loss of forearm rotation than dorsal angulation. Zimmerman et al. [33] compared palmar versus dorsal displaced pediatric metaphyseal radius fractures and found no differences in remodeling capacity, but they did find a higher restriction of supination in palmar displaced malunions. Furthermore, the degree of initial angulation at trauma may be predictive for re-displacement risk after 1 or 2 weeks. Initial angulation may predict the degree of fracture stability. Although in our study female sex and being older than 10 years at trauma were not associated with a clinically relevant limitation in forearm rotation (≥ 20°), we still cannot assume the findings will apply equally to both sexes at any age. Girls can be more skeletally advanced than boys with the same age, as the mean age for ossification of the physis differs between boys and girls (14.5 and 12.9 years, respectively), which results in less remodeling potential [25]. Greater remodeling potential is generally found in patients with more residual growth, a smaller distance to the most active growth plate, and fracture angulation in the sagittal plane [16]. Therefore, in clinical practice, one should be cautious to apply our results especially to nearly skeletally mature girls with severe (radial or volar) re-displacement. A second limitation is that although the RCT protocol stated to perform re-manipulation in case of a re-displacement, 13 of 30 re-displacements were left untreated. This introduced a treatment bias because there may have been factors influencing a surgeon to accept the re-displacement (for instance younger age), which could skew the impact of that re-displacement on the ultimate clinical result. This indicates that the criteria for reduction possibly were too stringent.

Furthermore, functional and radiologic assessments were not blinded and were performed by only one investigator. Blinded assessment was not possible because of the assessment of cosmetic appearance (including scars). The measurements of forearm rotation could also have inter- and intraobserver variations, thus our conclusions based on these measurements would be stronger if repeated measurements had been performed. Finally, below-elbow cast (compared with above-elbow cast) has been shown to be sufficient in treatment of distal forearm fractures in children, but this became apparent after initiation of our original RCT [4, 11, 29].

Does K-wire Stabilization of Reduced Metaphyseal Both-bone Forearm Fractures in Children Provide Better Forearm Rotation at Minimum 5-year Follow-up?

Although this RCT showed superior results of stabilization with K-wires in addition to an above-elbow cast after 7 months of follow-up [9], a minimum 5-year follow-up stabilization with K-wires did not provide better forearm rotation, radiographic parameters, or patient-reported results. Therefore, children with a displaced metaphyseal both-bone forearm fractures can be treated with closed reduction and an above-elbow cast without K-wire fixation. Previously, one meta-analysis compared results of displaced distal radius fractures between children treated with an above-elbow cast versus K-wire fixation [26]. This meta-analysis included three RCTs [9, 20, 21], one prospective cohort study [15], and two retrospective cohort studies [22, 28]. In this meta-analysis, 76% (292 of 382) of included children had a both-bone forearm fracture. In the Casting group, the re-displacement proportion was 46% (90 of 197) patients versus 4% (7 of 185) in the K-wire group (OR 0.07 [95% CI 0.03 to 0.15]). Complications other than re-displacement occurred more often in the K-wire group than in the Casting group (15.7% versus 3.6%). In contrast to the study by Colaris et al. [9], the studies by McLauchlan et al. [20] and Ozcan et al. [22] found no differences in functional results between the two treatment groups at 3 and 20 months of follow-up, respectively. Based on the combined results of these three studies, Sengab et al. [26] concluded that K-wire fixation does not result in better ROM but leads to a lower re-displacement proportion and fewer reinterventions. This is consistent with our findings. Future research, such as a meta-analysis or a large prospective observational study, is required to establish the influence of (skeletal) age, gender, and the severity and direction of malunion angulation of both the radius and ulna on clinical result. Currently, we await the results of the comparison of intervention and conservative treatment for angulated fractures of the distal forearm in children (AFIC) RCT by Adrian et al. [1], in which children (younger than 11 years of age) with displaced distal forearm fractures with up to 30° angulation are randomized between: cast immobilization versus closed reduction with or without additional K-wire fixation.

Do Malunions of Reduced Metaphyseal Both-bone Forearm Fractures in Children Induce Worse Functional Results?

Malunions lead to a higher risk (27% versus 7%) of a clinically relevant limitation in forearm rotation (≥ 20°) at minimum 5-year follow-up. Our results, however, show no differences in mean limitations between the two groups (Malunion versus Acceptable alignment group). This may seem contradictory, but it can be explained by the fact that most patients with a malunion (73%) still showed good forearm rotation at minimum 5-year follow-up, leading to a low mean limitation in forearm rotation of the whole malunion group. In clinical practice if fracture re-displacement occurs 1 or 2 weeks after the initial trauma, we advise to (based on sex, age and direction of angulation) consider re-manipulation and K-wire fixation promptly to decrease the risk of developing a persistent limitation in forearm rotation. Earlier, Colaris et al. [7] showed that pediatric metaphyseal both-bone forearm malunions angulated ≥ 16° developed a clinically relevant limitation in forearm rotation in 60% after a mean follow-up of 7 months.

Which Factors Lead to Limited Forearm Rotation?

At minimum 5-year follow-up, factors associated with a clinically relevant limitation in forearm rotation were malunion after above-elbow casting and a refracture. A study performed by Zimmerman et al. [32] revealed that children older than 10 years whose fractures healed with an angular deformity of more than 20° had the poorest long-term results, while in children younger than 10 years of age, angular deformity did not influence long-term results. The occurrence of a refracture was also associated with limited forearm rotation of ≥ 20°, possibly explained by repeated immobilization in a cast leading to soft tissue contractures [10]. Refractures are eight times more likely to reoccur in diaphyseal fractures as in distal forearm fractures [5]. Diaphyseal fractures behave vastly different to metaphyseal forearm fractures. In 1962, Hughston [18] claimed that in diaphyseal fractures “growth will not correct angulation deformity as it does in metaphyseal fractures”. Because of the relatively long distance between a diaphyseal fracture and the growth plates, only minimal correction of malalignment by growth can be expected.

CONCLUSIONS

At minimum 5-year follow-up in children with metaphyseal both-bone forearm fractures that were stable after closed reduction, we found no differences in forearm rotation between treatment with only an above-elbow cast and treatment with additional K-wire fixation. Re-displacement occurs more often if treated by an above-elbow cast alone. If fracture re-displacement is not treated promptly, a malunion may occur which is a risk factor for a clinically relevant limitation in forearm rotation at minimum 5-year follow-up. Children with a displaced metaphyseal both-bone forearm fracture can be treated with closed reduction and an above-elbow cast without additional K-wire fixation. The clinician should inform parents and patient about the high risk of fracture re-displacement, which, if left untreated, results in malunion with risk for forearm rotation limitations. Weekly radiographic monitoring is essential. If re-displacement occurs, re-manipulation and K-wire fixation should be considered based on sex, age and direction of angulation. Future research is needed to establish the influence of (skeletal) age, sex, severity of initial displacement and the direction of malunion angulation on clinical result.

Acknowledgments
We thank head of departments L. U. Biter MD, PhD, M. R. de Vries MD, PhD, Dr. C. P. van de Ven MD, PhD, and R. M. H. Wijnen MD, PhD, for their collaboration to make this research project possible at their departments. We thank Dr. N. M. C. Mathijssen MSc, PhD, for her scientific advice and support. Finally, we are grateful to all members of the administrative staff and outpatient departments of the four participating hospitals (Haga Hospital, Erasmus Medical Centre, Franciscus Hospital, and Reinier de Graaf Hospital).

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PART II:
Diaphyseal forearm fractures in children

Chapter 4

Long-term follow-up shows that early conversion to a below-elbow cast for reduced diaphyseal both-bone forearm fractures in children is safe

As submitted

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ABSTRACT

Background:

For distal forearm fractures in children, it has been shown that a below-elbow cast (BEC) is an adequate treatment that overcomes the discomfort of an above-elbow cast (AEC) and unnecessary immobilization of the elbow. For reduced diaphyseal both-bone forearm fractures, our previous randomized controlled trial (RCT) which compared AEC with early conversion to a BEC revealed no significant differences in re-displacement rates or functional outcomes at short-term follow-up. Although long-term results after diaphyseal both-bone forearm fractures in children are scarce, they are essential to finding out the effect of growth on clinical outcomes. Therefore, we conducted this long-term follow-up study to answer the following questions:

1. Is early conversion to a BEC safe for reduced stable diaphyseal forearm fractures in children, based on the long-term follow-up findings?
2. Does an accepted secondary displacement leading to a malunion result in inferior clinical outcomes at long-term follow-up?

Methods:

In this study we did a long-term follow-up of children who were included in a previous RCT. The original RCT was registered in ClinicalTrials.gov with registry identifier NCT NCT00398242. Ethics approval was obtained for this post-trial follow-up study with protocol number NL41839.098.12. Eligible patients were invited for long-term functional and radiographic assessment. The primary outcome was the difference in forearm rotation compared to the uninjured contralateral arm. Secondary outcomes were loss of flexion and extension of the elbow and wrist compared to the contralateral forearm, the ABILHAND-kids and quick-DASH questionnaire, JAMAR grip strength ratio, and radiological assessment of residual deformity.

Results:

The mean duration of follow-up was 7.5 (range 5.2 to 9.9) years. Out of 127 participants, 97 were included (76%). Loss of forearm rotation was 7.9^0 (SD 17.7^0) for the AEC group and 4.1^0 (SD 6.9^0) for the AEC/BEC group, with a mean difference of 3.8^0 (95% CI -1.7 to 9.4; p=0.2). The long-term follow-up showed significant improvement in forearm rotation in both groups compared to the rotation at 7 months.

Thirteen patients with persisting malunion at 7 months follow-up showed no clinically relevant differences in functional outcomes at long-term follow-up compared to children without malunion. The loss of forearm rotation was 5.5^0 (SD 9.1^0) for the malunion group compared to 6.0^0 (SD 13.9^0) in the non-malunion group, with a mean difference of 0.4^0 (95 % CI of -7.5^0 to 8.4^0 p=0.9).

Conclusions:
Early conversion to a BEC in reduced diaphyseal both bone forearm fractures in children is safe at long-term follow-up and should be the treatment of choice. Moreover, this study shows that remaining growth behaved like a friend in children with reduced diaphyseal both bone forearm fractures, as patients in which secondary fracture displacement occurred showed good to excellent long-term results.

Keywords: fracture, forearm, pediatric orthopedics, trauma, long-term results, casting

Level of evidence: I

INTRODUCTION

Background

Diaphyseal forearm fractures are far less forgiving than distal forearm fractures in the growing skeleton. Almost half of pediatric fractures are forearm fractures of both bones of which 20% is located in the diaphysis [5,6,29]. Although there is an increasing tendency of treating diaphyseal forearm fractures with intramedullary nails, stable fractures after reduction can also be treated in an above elbow cast (AEC) [32]. The disadvantage of treatment in a cast remains fracture re-displacement which has been described in up to 7-39% [22,30,32]. Re-displaced fractures, that are 'accepted as is' and which are not treated with re-manipulation or surgical stabilization often result in a malunion [2,6,9,10,12,15,19]. These diaphyseal malunions show, in general, a lower tendency to correct by growth in comparison to distal forearm fractures. Such a malunion can result in rotational impairment caused by either collision of the forearm bones or tightness of the soft tissues as the central band of the interosseous membrane [2,10,14,15,20,23,28,31].

To find out if early conversion to BEC is safe, our group conducted a randomized controlled trial (RCT) in 2013 that included 127 children who sustained a displaced diaphyseal both-bone fracture of the forearm which was stable after reduction. These children were randomized into 2 groups. Group 1 was immobilized in an AEC for 6 weeks, group 2 was immobilized in an AEC for 3 weeks followed by three weeks of a BEC [19]. After 7 months no statistically significant difference in loss of forearm rotation between both groups was found: 17.6 (16)° in the AEC group and 12 (12.4)° in the AEC/BEC group. There was a similar re-displacement rate: 23 out of 62 (37%) in the AEC group and 20 out of 65 (31%) in the AEC/BEC group. A total of 22 malunions were accepted 'as is', based on previously set criteria (Figure 2), and did not receive any further treatment [7]. Cast comfort was significantly better in the AEC/BEC group. However, we believe that treatment recommendations should be based on the occurrence of complications and functional outcomes in the long term.

Rationale

Therefore, we conducted a long-term clinical and radiological follow-up of the patients previously included in this RCT to answer the following questions:

1. Is early conversion to a BEC safe for reduced stable diaphyseal forearm fractures in children, based on the long-term follow-up findings?
2. Does an accepted secondary displacement leading to a malunion result in inferior clinical outcomes at long-term follow-up?

PATIENTS AND METHODS

Trial design and participants

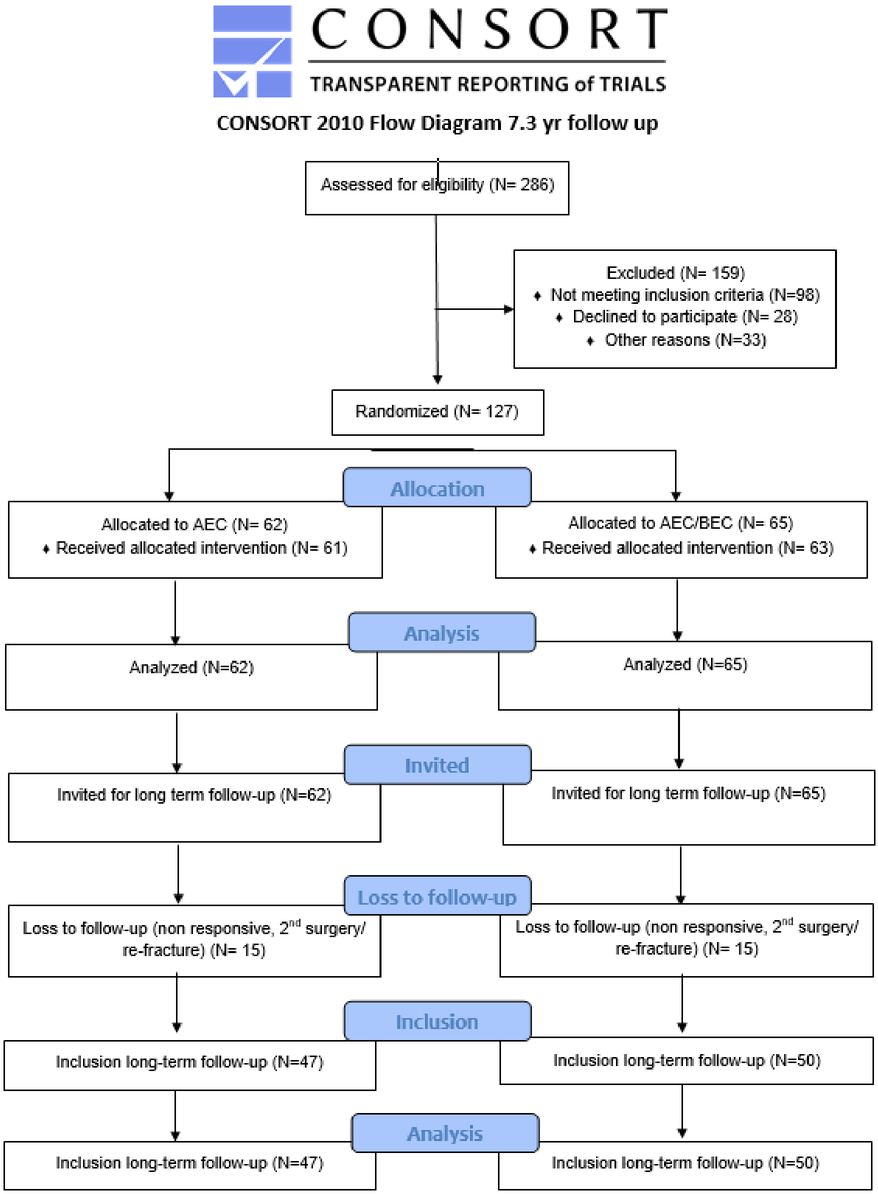
The study design is the long-term follow-up (with a minimum of 5 years) of a previous RCT by Colaris et al.[7]. We approached the 127 patients who were included between January 2006 and August 2010. All patients were invited to visit the outpatient clinic for clinical and radiological reassessment in the period between January 2014 and April 2017. Children who visited the emergency department of one of 4 participating Dutch hospitals: Erasmus Medical Center (Rotterdam), HAGA Hospital (The Hague), Reinier de Graaf Hospital (Delft), and Franciscus Vlietland Hospital (Schiedam), were eligible for participation. Inclusion criteria for the initial RCT were: children who presented with a displaced diaphyseal both-bone forearm fracture that was stable after reduction. The exclusion criteria were: no response to our invitation for follow-up, refracture, or secondary surgery of the affected forearm. At seven years, follow-up measurement and informed consent were reobtained from all children and parents of children aged <12.

The original RCT was registered in ClinicalTrials.gov with registry identifier NCT NCT00398242. For this post-trial follow-up study, ethics approval was obtained at the regional medical ethical committee with protocol number NL41839.098.12. This study complies with the CONSORT statement (Figure 1).

Outcomes measures

Our primary outcome was a difference in forearm rotation compared to the contralateral uninjured arm at long-term follow-up. This primary outcome was also used in the initial RCT. We compared the outcomes of long-term follow-up to the outcomes at seven months of follow-up. Secondary outcome measures were loss of flexion-extension of the elbow and wrist compared to the contralateral forearm, the quickDASH, ABILHAND-kids questionnaire, grip strength (using a JAMAR Dynamometer) displayed as a ratio of affected forearm / contralateral side and radiological assessment of the angulation of radius and ulna [7,16,24,31,33]. One orthopedic surgeon (LD) performed the standardized physical examination. Finally, we performed a radiological assessment on X-rays at the final follow-up, in which we measured the coronal and sagittal angulation of the radius and ulna. Different cut-off values were used to define a malalignment for different ages (Figure 2). Radiological measurements were conducted blinded by one of the co-authors (PE)[13,17,26,34]. Analyses were done using locally-available analysis programs (PACS and JiveX).

Figure 1: Consort Flow Diagram 7.3 years follow-up



Statistical methods

Of the initial 127 children who were analyzed at seven months in the previous RCT, 97 patients (76% response) were reevaluated at this long-term follow-up. To evaluate whether the included patients in the current study are representative of the total initial study population and address the potential effects of attrition, we performed a sensitivity analysis. We compared the baseline characteristics, functional outcomes, and complications at short-term follow-up (7 months) between the included patients (responders) and the patients lost to follow-up (non-responders).

Long-term results of the primary and secondary outcome measures of the two treatment groups (AEC vs. AEC/BEC) were compared. Differences between both groups were analyzed using independent T-tests and crosstabs. Results are presented as mean with standard deviation (SD) and p-values. In addition, Levene's test for equality was performed to compare means. Finally, a linear mixed model analysis was conducted for multiple follow-up moments (moment of trauma, 6 weeks post-trauma, 7 months post-trauma, and 7.5 years post-trauma) in time to address possible missing data.

To assess the inter-rater reproducibility of radiographic assessment, two authors (PE and LD) measured radiological angulations of forty-five cases (at cast removal and final follow-up). The intra-class correlation coefficient was calculated in the initial RCT (ICC, two-way mixed and absolute agreement).

Statistical analyses were performed using IBM SPSS Statistics version 27.

RESULTS

Of the initially 127 patients included in the initial RCT 97 (76%) patients participated in the long-term follow-up measurements. The mean follow-up was 7.5 (range 5.2 to 9.9) years. The study population characteristics are presented in Table 1. We found no statistically significant differences in the baseline characteristics or functional outcomes at short-term follow-up (7 months) between the loss to follow-up group (non-responders) and the included population (responders) (Table 2).

Table 1. Baseline characteristics of the study population

Baseline	Total	AEC	AEC/BEC	P value
Number of patients (N)	97	47	50	
Age at time of fracture, years (range)	7.9 (1.3-14.9)	8.3 (3.2-14.9)	7.5 (1.3-13.5)	0.2
Age at FU, years (range)	15.4 (8.7-24.2)	15.8 (9.3-24.2)	14.9 (8.7-21.6)	0.2
Length FU, years (range)	7.5 (5.2-9.9)	7.6 (5.2-9.9)	7.4 (5.2-9.8)	0.6
Male sex, % (N)	64 (62)	64 (30)	64 (32)	1.0
Fracture type, radius in % (N)				0.008
Buckle	0 (0)	0 (0)	0 (0)	
Greenstick	46(45)	32 (15)	60 (30)	
Complete	54 (53)	68 (32)	40 (20)	
Fracture type, ulna in % (N)				0.2
Buckle	0 (0)	0 (0)	0 (0)	
Greenstick	55 (53)	47 (22)	62 (31)	
Complete	45 (44)	53 (25)	38 (19)	

AEC *= above elbow cast;* ***BEC*** *= below elbow cast; CI = Confidence interval; N= number of patients*
Data is presented as mean with standard deviation between parentheses unless reported otherwise.

Table 2. Representability of the lost to follow up and included population.

	Loss to follow up	Included	Mean difference with 95% CI
Number of patients	30	97	/
Age at trauma, years(range)	7.8 (1.5-14.9)	7.9 (1.3-14.9)	-0.04(-1.4-1.3)
Male sex, % (N)	81 (25)	64 (61)	/
Loss of forearm rotation at 7 months, degrees	14.7 (13.7)	14.6 (14.8)	0.04(-6.0-5.9)
Arc of motion at 7 months, degrees	132 (23)	132 (18)	-0.3(-8.3-7.6)
ABILHAND-kids questionnaire *	41.3 (1.5)	40.1 (8.1)	1.3 (-1.9-4.4)
VAS-cosmetics parents/child **	7.6 (2.4)	8.3 (2.0)	-0.7 (-1.6 -0.2)
VAS-cosmetics surgeon ***	8.1 (2.0)	8.5 (1.9)	-0.4 (-1.3- 0.4)

* ABILHAND-kids questionnaire score 0-42/ 42 is the optimal score, ** VAS cosmetic parents/child sore 0-10/10 is optimal score, *** VAS cosmetic surgeon score 0-10/ 10 is optimal score

Is early conversion to a BEC safe for reduced stable diaphyseal forearm fractures in children, based on the long-term follow-up findings?

After long-term of follow-up, no statistically significant difference in loss of forearm rotation between both groups was found, respectively 7.9° (SD 17.7) in the AEC/BEC group and 4.1° (SD 6.9^0) in the BEC group, with a mean difference of 3.8° (95% CI -1.7^0 to 9.4^0, p=0.2) (Table 3). The AEC group improved from a mean loss of rotation of 27.2° (SD 21.6°) at two months to 17.6° (SD 16.0°) at seven months to 7.9° (SD 17.7°) at 7.5 years. For the AEC/BEC group, this was 21.8° (SD 18.7^0) at two months, 12.0° (SD 12.4^0) at seven months, and 4.1° (SD 6.9°) at 7.5 years. A mixed linear model analysis also showed a significant improvement in forearm rotation over time for both groups.

Secondary outcomes showed no statistically significant differences between the AEC and the AEC/BEC groups at long-term follow-up (Table 4). Less ulnar angulation in the coronal view and more ulnar bowing (p<0.001) were found in the AEC/BEC group (p<0.001) (Table 5). When comparing all time points, we found a significant increase in radial angulation over time in the coronal view for the AEC/BEC group (p=0.003) but not for the BEC group.

Table 3. Loss of forearm rotation of the fractured arm, subgroup analysis in percentages

	AEC	AEC/BEC	Mean diff with 95%CI
2 months after trauma	N=62	N=65	
None	7	13	
1-10^0	20	31	
11-20^0	26	19	
21-30^0	16	8	
>31^0	31	29	
Mean limitation in degrees (SD)	27.2 (21.6)	21.8 (18.7)	5.3(-1.9-12.6)
7 months after trauma	N=62	N=65	
None	20	32	
1–10^0	20	28	
11–20^0	31	22	
21–30^0	12	11	
> 31^0	17	8	
Mean limitation in degrees (SD)	17.6(16.0)	12.0 (12.4)	5.7(0.6-10.7)
7.5 years after trauma	N=47	N=50	
None	48	59	
1–10	24	27	
11–20	22	12	
21–30	2	2	
> 31 degrees	4	0	
Mean limitation in degrees (SD)	7.9 (17.7)	4.1 (6.9)	3.9(-1.7-9.4)

Results in percentage. Mean limitation with Standard Deviation between parentheses in degrees. AEC= above elbow cast. BEC= below elbow cast. CI= Confidence Interval.

Table 4. Data on primary and secondary outcomes at 7.5 years of follow-up

	AEC (N = 47)	AEC/BEC (N = 50)	Mean diff 95%CI
Age at follow-up, years (range)	15.8 (9.3-24.2)	14.9(8.7-21.6)	0.9(-0.5-2.3)
Follow-up length, years (range)	7.6 (5.2-9.9)	7.4 (5.2-9.8)	0.1(-0.4-0.7)
Loss of forearm rotation, degrees	7.9 (17.7)	4.1 (6.9)	3.8(-1.7-9.4)
Arc of motion, degrees	152 (21)	155 (11)	-2.5(-9.3-4.4)
Loss of wrist flexion-extension, degrees	1.0 (5.0)	0.6 (4.2)	0.4(-1.5-2.2)
Loss of elbow flexion-extension, degrees	0 (/)	0 (/)	/
ABILHAND-kids questionnaire *	41.0 (2.4)	41.7 (0.7)	-0.7(-1.4-0.04)
quick DASH score **	5.8 (9.6)	2.9 (6.0)	2.9(-0.5-6.2)
JAMAR score (ratio) ***	0.95 (0.2)	0.99 (0.2)	-0.04(-1.1-0.03)

AEC= Above elbow cast. BEC= below elbow cast, CI= confidence interval. N=number of patients.
Data is presented as mean with standard deviation between parentheses unless otherwise stated.
** ABILHAND-kids questionnaire score 0-42/ 42 is optimal score,*
*** DASH score 0-100/100 being the worst score,*
**** JAMAR ratio= grip strength affected wrist/ collateral side*

Table 5. Radiological analysis of angulation at 7 months compared to 7.5 years follow-up

	AEC	AEC/BEC	Mean with 95% CI
7 months follow-up	N=62	N=65	
AP radius, degrees	6.4 (3.9)	5.3 (4.0)	1.1 (-0.6-2.7)
AP ulna, degrees	5.4 (3.9)	5.4 (3.9)	-0.44 (-1.6-1.6)
Lateral radius, degrees	7.9 (4.8)	7.7 (5.1)	0.2 (-1.8-2.2)
Lateral ulna, degrees	5.5 (4.5)	4.5 (3.6)	0.8 (-0.6-2.7)
Bowing radius, %	11.6 (2.5)	12.6 (2.1)	-1.0 (-2.1-0.1)
7.5 years follow-up	N=47	N=50	
AP radius, degrees	9.0 (2.1)	8.7 (4.1)	0.3 (-1.0-1.7)
AP ulna, degrees	6.4 (3.1)	4.6 (2.3)	1.8(0.7-3.0)
Lateral radius, degrees	4.8 (3.3)	4.9 (3.6)	-0.1 (-1.5-1.4)
Lateral ulna, degrees	4.7 (2.6)	4.5 (2.1)	0.2 (-0.8-1.2)
Bowing radius, % *	11.8 (2.3)	13.4 (2.8)	-1.6(-2.7 to -0.5)

*AEC= Above elbow cast. BEC= below elbow cast, CI= confidence interval. N=number of patients. Data is presented in degrees with standard deviation between parentheses or reported otherwise. *=r/Y*100, (Firl and Wunsch 2004), see Figure 2*

Does an accepted secondary displacement leading to a malunion result in inferior clinical outcomes at long-term follow-up?

Accepted secondary displacement in the cast resulted in malalignment in 34 patients during the cast treatment, of which 22 still had a radiological malunion based on the previously set criteria and 12 had remodeled at 7 months follow-up (see Figure 2). Of these 22 patients with malunions only one case was lost to follow-up for the long-term measurements. At long-term follow-up 13 of these 22 patients still had a remaining radiologic malunion.

Figure 2. Criteria for reduction of the fracture of radius and/or ulna based on anteroposterior and/or lateral radiographs.

Type of displacement	Age in years	Displacement
Angulation	< 10	>15°
	10-16	>10°
Translation	<16	>half of bone diameter
Rotation	<16	>0°

The group (n = 13) with a persisting malunion showed a mean loss of rotation of 5.5^{0} (SD 9.1^{0}), compared to 6.0^{0} (SD 13.9^{0}) in the non-malunion group, with a mean difference of 0.4^{0} (95 % CI of -7.5^{0} to 8.4^{0} p=0.9). Secondary outcomes showed no significant differences between the malunion and non-malunion groups. The JAMAR ratio in the non-malunion group was 0.97 (SD 0.2) compared to 0.94 (SD 0.2) in the malunion group, with a mean difference of 0.04 (95% CI of -0.06 to 0.14 p=0.4). The ABILHAND-kids questionnaire score was 41.4 (SD 1.9) in the non-malunion group compared to 41.7 (SD 0.5) in the malunion group, with a mean difference of -0.3 (95% CI of -1.4 to 0.7 p=0.5). The QuickDASH was 4.2 (SD 8.2) in the non-malunion group compared to 5.0 (SD 6.4) in the malunion group, with a mean difference of -0.8 (95% CI of -5.6 to 4.0 p=0.7) (Table 6). Linear mixed analyses showed significant improvement in rotation over time (p=0.002).

Radiological analysis comparing the malunion with the non-malunion group only showed a significant difference in lateral radial angulation, 8.2^{0} (SD 4.0^{0}) in the malunion group, compared to 4.3^{0} (SD 3.1^{0}) in the non-malunion group, mean difference -4.0^{0} (95 % CI of -5.9^{0} to -1.9^{0} p=<0.001) (see Table 6).

The interrater reproducibility of the radiological assessment showed an ICC of 0.81 (95% CI: 0.68 to 0.89) and 0.89 (95% CI: 0.81 to0.94) for the radioulnar angulation of the ulna and radius, respectively. The ICC of sagittal angulation was 0.92 (95% CI: 0.85 to 0.95) for the ulna and 0.87 (95% CI: 0.77 to 0.92) for the radius [8].

Table 6. Outcome of subgroup with malunion at final FU compared to those without malunion

Primary/secondary outcomes	Malunion (N=13)	No malunion (N=84)	Mean diff 95%CI
Loss of forearm rotation, degrees	5.5 (9.1)	6.0 (13.9)	0.4 (-7.5-8.4)
ABILHAND-kids questionnaire *	41.7 (0.5)	41.4 (1.9)	-0.3 (-1.4-0.7)
quick DASH score **	5.0 (6.4)	4.2 (8.2)	-0.8 (-5.6-4.0)
JAMAR score (ratio) ***	0.94 (0.2)	0.97 (0.2)	0.04 (-0.06-0.14)
Radiologic analysis			
AP radius, degrees	8.9 (4.4)	8.8 (3.1)	-0.1 (-2.1-1.9)
AP ulna, degrees	5.3 (3.0)	5.5 (2.9)	0.3 (-1.5-2.1)
Lateral radius, degrees	8.2 (4.0)	4.3 (3.1)	-4.0 (-5.9—1.9)
Lateral ulna, degrees	4.6 (2.6)	4.6 (2.3)	-0.01 (-1.5-1.5)
Bowing radius, % ****	13.1 (2.8)	12.5 (2.6)	-0.5 (-2.2-1.1)

CI= confidence interval. N=number of patients. Data is presented in degrees with standard deviation between parentheses or reported otherwise ;
** ABILHAND-kids questionnaire score 0-42/ 42 is optimal score,*
*** DASH score 0-100/100 being the worst score,*
**** JAMAR ratio= grip strength affected wrist/ collateral side*
*****=r/Y*100, (Firl and Wunsch 2004), see Figure 2.*

DISCUSSION

The short-term outcomes of the previous randomized controlled trial (RCT) which randomized 127 children with a stable reduced displaced diaphyseal both-bone forearm fracture to either six weeks of AEC or early conversion to BEC found a similar rate of fracture re-displacement and comparable functional outcomes after seven months, but a higher cast comfort in the AEC/BEC group [8]. The current long-term follow-up study of this RCT shows that early conversion to a below-elbow cast is a safe and effective treatment for pediatric forearm fractures without any significant long-term functional limitations. So both short and long-term follow-up supports early conversion to below elbow cast as the recommended treatment strategy for stable reduced pediatric both-bone forearm fractures. Accepted re-displaced fractures resulting in a malunion even showed excellent long-term clinical outcomes despite that 62% of the malunions were not fully corrected by growth.

Clinical and radiological outcomes

In 1990, Price et al. studied the long-term functional outcomes of 39 skeletally immature patients with severe diaphyseal both-bone forearm fractures, which healed in a malunited position. At a mean follow-up of 5.8 years, they found good or excellent outcomes in 92% of cases. In their series, results were graded as excellent if there were no complaints with physical activity and/or a loss of ≤10 of forearm rotation [25]. Our study showed similar excellent results in the limitation of forearm rotation at long-term follow-up with respectively 7.9 degrees (SD 17.7) in the AEC/BEC group and 4.1 (SD 6.9) in the AEC group. In the above elbow cast group, 96% had good/excellent functional outcomes, and in the early conversion group, even 100% had good/excellent functional outcomes.

Our study showed some significant differences in radiologic angulation between the two treatment groups, but none of these were clinically relevant. Regardless of the initial treatment, the radiological outcomes were good.

Re-displacement

The literature shows that diaphyseal both bone forearm fractures treated non-operatively, either with a cast or with manipulating followed by a cast, have a high tendency to re-displace.

Bowman et al. retrospectively analyzed radiographs of 282 children with diaphyseal both-bone forearm fractures. As criteria for reduction, Bowman et al. accepted shaft angles up to 20 degrees, pending on the location of the fracture and sex of the patient. Of the 144 participants who failed closed reduction and casting within 4 weeks, 80 (56%) had their first radiographic evidence of re-displacement during the first-week post-reduction, 34 (24%) during the second week, 23 (16%) during the third week, and 7 (5%) during the fourth week. Bowman et al. stated that patients ten years or older and those with proximal-third radius fractures are at the highest risk for re-displacement [4].

Yang et al. also studied risk factors for re-displacement in diaphyseal forearm fractures in 57 children. They found that a poorer reduction (odds ratio of 8.5) and complete fracture (odds ratio of 9,6) were factors associated with re-displacement [32].

Jones et al. performed a retrospective study analyzing their treatment of 730 consecutive pediatric forearm fractures in children. For midshaft forearm fractures, the reduction was performed for any patient in the 0- to 8-year age group with >10 degrees of angulation. In children aged 9–17, the reduction was performed for any fracture with >8 degrees of angulation [19].

Malunion and functional outcome

It is interesting to know if re-displacement resulting in a malunion also results in an inferior functional outcome.

Eismann et al. retrospectively studied the radiographic outcomes of 31 children who were treated with re-reduction due to re-displacement of a displaced both-bone forearm shaft fracture. They stated that re-manipulation provided satisfactory radiographic outcomes and was 2.4 times less expensive than surgical stabilization. However, failure of conservative treatment was mainly seen in patients with apex ulnar angulation, which encroaches upon the interosseous space, contributing to impaired forearm rotation [11].

Zionts et al. prospectively studied the relationship between residual deformity and functional outcomes following closed treatment of displaced diaphyseal both-bone forearm fractures in 25 older children. They found that loss of forearm rotation was correlated with the maximum angulation of the radius seen on either the final PA or lateral radiograph. Of the 25, five patients (20%) had malunions with more than 15 degrees of angulation of either the radius or ulna of which 3 patients demonstrated >30° of loss of forearm rotation [35].

Together, Voto et al., Bochang et al., and Jones et al. reported functional outcomes of 103 pediatric patients with forearm shaft fractures who were re-manipulated after re-displacement and all patients had satisfactory functional results with no complications. The authors concluded that “re-manipulation provides a safe, effective means to obtain and maintain reduction” [3,19,30].

In line with these studies, our long-term follow-up shows that even patients ending up with a malunion due to re-displacement of the fracture and insufficient remodeling, generally have good/excellent functional outcomes after seven years.

Remodeling

Literature states that some degree of malunion of the forearm can be accepted in children because the remaining growth in pediatric bones enables remodeling capacity. The degree of correction by growth depends on the remaining growth and the location and plane of the malunion. Early studies have already demonstrated a significant relationship between age and the ability to correct the deformity. Moesner and Ostergaard suggested that children under nine years of age can achieve correction of 90 percent of their malunion, and remodeling capacity decreases with age >9 years [21]. Höström et al. showed that the age at the time of the fracture was correlated positively with late residual angulation, older children being less able to compensate for the fracture deformity. Also, they showed a significant correlation between the late residual angulation and limitation of pronation and supination [15]. Johari et al. showed that fractures located closest to the epiphysis have the highest remodeling potential. They concluded that midshaft fractures in children >10 years of age with angulation have a poor diagnosis if left uncorrected [18].

Price et al. studied the outcomes of 39 children with malunions after severe diaphyseal both-bone forearm fractures with a mean follow-up of 6 years. Complete remodeling occurred in only 12 out of 39 patients, almost all <10 years of age, but 92% showed good or excellent outcomes [25]. Thomas et al. showed that most malunions in children end in complete functional recovery or minimal function loss with no influence in daily use [27].

Our study can support this; While patients with accepted malunions after secondary displacements have more sagittal radial angulation at follow-up, all children remodeled to a clinically acceptable angulation, resulting in good to excellent functional outcome over time. However, this only includes children who had initially stable fractures with acceptable angulations, that were secondarily displaced in the cast. Therefore the moment for intervention had already passed. Although the long-term follow-up results are rather good even in children with a malunion, diaphyseal malunions will correct slow by growth and it might take years to gain full rotation and a cosmetic straight forearm. Therefore we support the use of intramedullary elastic nails in case of unstable diaphyseal both bone forearm fractures that are reduced in the operation room.

Study limitations

Primarily, the clinical assessment was not blinded. Blinding of patients was impossible because of the cast morphology. Radiological assessments, however, were blinded.

The second limitation is the number of patients lost to follow-up. The main reason is that young patients (and their parents) without any complaints are not very motivated to return to the hospital for an additional assessment. Therefore, in our opinion a follow-up of 76% is actually quite high and acceptable for this study population. To address the potential effects of attrition, we did a patient group analysis, showing that the follow-up group was representative of the original study group

CONCLUSIONS

This long-term follow-up study of patients included in a previously conducted RCT shows that early conversion to a BEC is safe in reduced stable diaphyseal both bone forearm fractures in children. Moreover, this study shows that remaining growth behaved like a friend in children with reduced stable diaphyseal both bone forearm fractures. Even in cases of malalignment, function improved over time resulting in excellent long-term outcomes.

Conflict of interest statement
None of the authors or the authors' institutions has a financial or other relationship with other people or organizations that may influence this work.

Ethical Review Committee Statement
The study has been performed in accordance with the ethical standards in the 1964 Declaration of Helsinki and has been carried out in accordance with relevant regulations of the US Health Insurance Portability and Accountability Act (HIPAA). Details that might disclose the identity of the subjects under study are omitted.

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Chapter 5

Both-bone forearm fractures in children: the outcomes of a prospective cohort of 316 patients with a mean follow-up of 7 years.

As submitted

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ABSTRACT

Background:
Some degree of fracture displacement can be safely accepted in pediatric forearm fractures due to their remodeling capacity. So far, no studies have determined which factors are associated with inferior long-term clinical outcomes. Therefore, this investigation prospectively determined the long-term outcomes of children with both-bone forearm fractures. Our research questions were:

1) Which factors are associated with a pro-supination limitation at long-term follow-up?
2) Do accepted re-displacements lead to inferior long-term functional and radiographic outcomes?

Methods:
A prospective cohort study was conducted analyzing pediatric patients with a distal metaphyseal or diaphyseal both-bone forearm fracture with a minimum four-year follow-up. Patients were primarily included in various randomized controlled trials: Non-displaced distal fractures were randomized between below-elbow and above-elbow cast (BEC/AEC); Displaced distal fractures were randomized between closed reduction with or without K-wires fixation; Stable diaphyseal fractures were randomized between AEC and early conversion to BEC; Unstable diaphyseal fractures were treated with 1 or 2 intramedullary nails.

Our primary outcome measure was the limitation in pro-supination. Secondary outcomes were patient-reported outcome measures, grip strength and residual angulation. Multi-variate linear regression analysis was performed to identify factors associated with a pro-supination limitation. Radiographic and functional outcomes were compared between patients with accepted re-displacements and good alignments.

Results:
In total, 316 participants with 149 diaphyseal and 167 distal metaphyseal fractures were included, with a mean follow-up of 7.2 years. Predictors for limitation in pro-supination at long-term follow-up were: complete ulnar fracture, diaphyseal fracture and older age at trauma. Accepted diaphyseal re-displacements led to greater residual angulation at long-term follow-up.

Conclusions:
Excellent spontaneous remodeling of angular deformity and functional outcomes are seen in distal metaphyseal forearm fractures in children with remaining growth potential. However, in midshaft forearm fractures, growth will not correct angular deformity as it does in distal fractures and more pro-supination limitation is seen.

Level of Evidence: Level II

INTRODUCTION

Although forearm fractures account for 38% of pediatric fractures, long-term follow-up studies are scarce, and the optimal treatment strategy is still unknown [1-3].

Treatment of forearm fractures in children generally varies from simple immobilization to closed reduction with or without stabilization by K-wiring or intramedullary pinning [4,5] . Re-displacement occurs in up to 46% of diaphyseal forearm fractures and 51% of displaced distal metaphyseal, even though they appear stable after closed reduction [6,7] . Because of remodeling potential, a forearm fracture with some degree of displacement or angulation can be safely accepted in the expectance that remodeling will occur [2]. However, the acceptable degree of residual deformity for both distal metaphyseal and diaphyseal forearm fractures in children remains ill-defined [3,8]. There is a trend toward more operative management, although no long-term outcomes studies have shown superior results following an operation [9] . *"Despite the remarkable potential for remodeling seen in pediatric forearm fractures, there is still a natural tendency to try to make each fracture radiographically more anatomic"* [10]. Angular deformity of the distal forearm usually entirely remodels within two to five years, provided the epiphysis does not fuse [11,12].

Therefore, the effects of re-displacement on long-term outcomes must be established. Previously, we reported the short-term outcomes of 410 children with both-bone forearm fractures [4,5,13-16]. The assessment of the long-term follow-up of this cohort is essential to evaluate and potentially adjust the treatment strategy for pediatric forearm fractures [17]. The clinical outcomes after pediatric forearm fractures are mainly influenced by pro-supination. The purpose of this prospective study was to investigate functional and radiographic outcomes after both-bone forearm fractures in children with a minimum follow-up of four years.

Our main research questions were:

1) Which factors are associated with a persisting pro-supination limitation after pediatric both-bone forearm fractures?

2) Do accepted re-displacements of pediatric both-bone forearm fractures lead to inferior long-term outcomes?

PATIENTS AND METHODS

Study design, setting, and participants

Between 2006 and 2010, 410 children with both-bone forearm fractures were prospectively included, and their short-term outcomes were reported with a mean follow-up of 7 months [4,5,13-16]. Currently, we report the long-term follow-up of this entire cohort, with a minimum follow-up of 4 years. The following inclusion criteria were used: children aged <16 years at trauma with a both-bone forearm fracture in the diaphysis or distal metaphysis. Exclusion criteria were torus fractures of both the radius and ulna and open fractures.

Description of Treatment

The criteria for performing closed reduction of a pediatric both-bone forearm fracture were: a closed reduction was performed in case of ≥50% displacement, ≥15° of angulation in children aged <10 years, and ≥10° of angulation in children aged 10-16 years. Re-displacement was defined as the re-occurrence of a displacement meeting the initial reduction criteria during cast treatment. The protocol stated to perform a re-manipulation for all re-displacements. An accepted re-displacement was defined as a re-displacement which was treated conservatively due to the treating surgeon's or parents' preference.

The included children were participants in several randomized controlled trials (RCTs) in which treatment protocol was based on the fracture location, need for reduction, and stability [4,5,14,15,18]. Distal metaphyseal fractures without the need for reduction were randomized to a below-elbow cast (BEC) or an above-elbow cast (AEC) [18]. Stable reduced distal metaphyseal fractures were treated with or without K-wires fixation [4]. Unstable reduced distal metaphyseal fractures were treated with K-wires fixation. Diaphyseal fractures without reduction or stable after reduction were treated with AEC for six weeks or early conversion to a BEC after three weeks [5,14] . Unstable diaphyseal fractures were treated with 1 or 2 intramedullary nails [15]. A fracture was defined as unstable if performing maximum pronation or supination after closed reduction caused re-displacement under fluoroscopy [5,19].

The short-term outcomes of these RCTs are summarized in short: A BEC is recommended to treat minimally displaced distal metaphyseal fractures [18]. Children with displaced metaphyseal fractures who underwent closed reduction alone had more re-displacements (45% vs 8%) and less pro-supination (14° vs 7°) than children who received K-wires [4]. Children with stable diaphyseal fractures can be safely treated with early conversion to BEC [5]. Unstable diaphyseal both-bone forearm fractures should be treated with two intramedullary nails [15].

Variable, Outcomes measures, Data Sources, and Bias

Our primary outcome measure was the limitation of pro-supination. Secondary outcomes were patient-reported outcome measures (ABILHANDkids, QuickDASH questionnaire, Numeric rating scale (NRS) cosmetic scores), grip strength (Jamar ratio) and angular deformity.

To investigate which factors are associated with a pro-supination limitation, we analyzed: age at trauma (≤10 versus >10 years), fracture location, fracture type (complete versus torus/greenstick), re-displacement, treatment for re-displacement and re-fracture. To investigate if accepted re-displacements lead to inferior outcomes at long-term follow-up, we compared outcomes between patients with accepted re-displacements and good alignments. We subdivided between diaphyseal and distal metaphyseal re-displacements.

One orthopedic surgeon examined patients at short-term follow-up. Another orthopedic surgeon examined patients at long-term follow-up. One author measured the intramedullary angulations, according to Bowman's method [7].

Statistical methods

Loss to follow-up was addressed by comparing the demographics of the included patients with those lost to follow-up. For categorical variables, the Chi-square test was used. For continuous variables, the independent samples t-test was used. Next, an exploratory analysis was performed to identify factors associated with a pro-supination limitation at long-term follow-up. A p-value of <0.10 was used as a threshold to determine which factors progressed to the more-definitive multi-variate linear regression analysis. Lastly, the outcomes between patients with accepted re-displacements and good alignments were compared using the independent samples t-test.

Ethics, data sharing, funding, and potential conflicts of interest

Our institutional review board approved this study, registered under protocol number NL41839.098.12. All authors declare no conflict of interest.

RESULTS

Participants, descriptive data

Between 2014 and 2016, 316 out of 410 participants (77%) were included, with a mean follow-up of 7.2 years (range 4.2 to 10.3). There were 149 diaphyseal fractures (46%) and 167 distal fractures (54%). The mean age at trauma was 8.1 years (range 0.9 to 16.5). There were no significant differences between the included patients and those lost to follow-up (Table 1).

Table 1. Representation of follow-up population

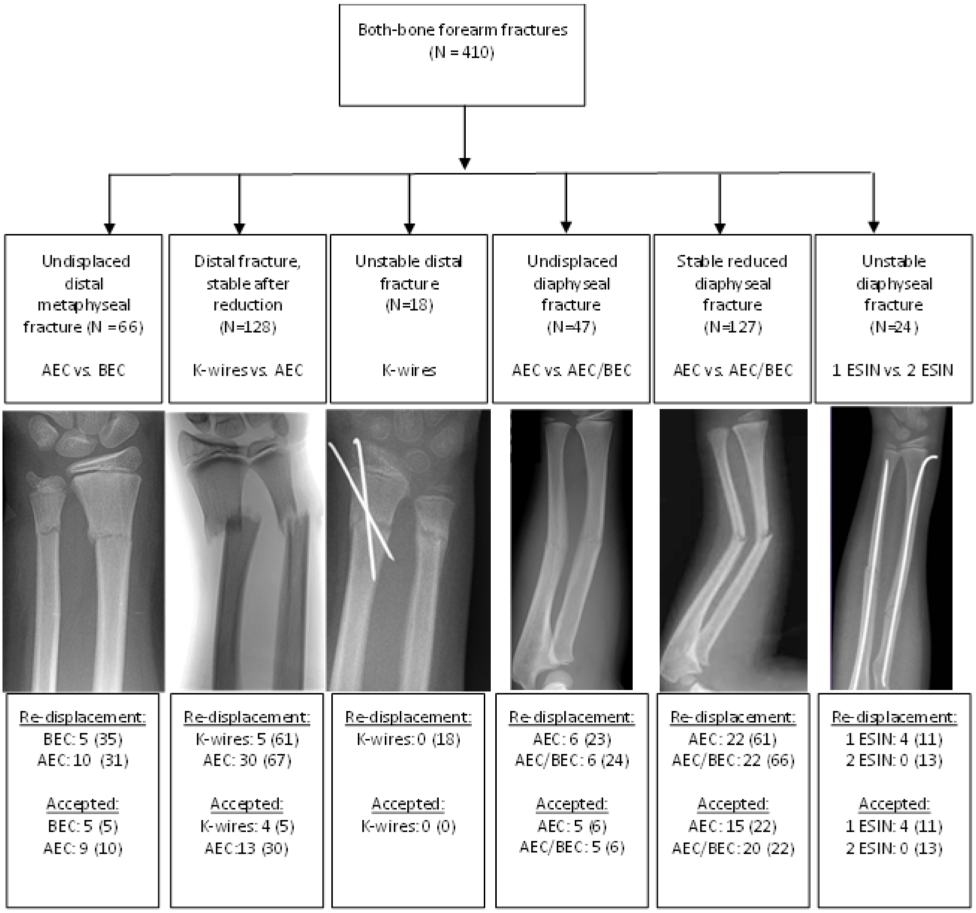
	Included for long-term FU (N = 316)	Lost to FU (N = 94)	Mean difference (95% CI)	P-value
Age at trauma	8.0 (±3.3)	8.4 (±3.6)	-0.4 (-1.2 to 0.4)	0.29
Male sex	60% (191)	70% (66)	-9.8% (-21 to 1)	0.08
Complete Radius Fracture	53% (167)	56% (53)	-3.2% (-15 to 9)	0.59
Complete Ulna Fracture	39% (122)	40% (37)	-1.0% (-13 to 10)	0.86
Re-displacement rate	27% (84)	27% (26)	0.0% (-10 to 10)	0.99
Accepted re-displacements	19% (59)	21% (11)	-2.6% (-12 to 7)	0.58
Loss in pro-sup at 6m FU	11.6° (±13.8)	13.9° (±15.1)	-2.3° (-6 to 1)	0.17
Complications	27% (86)	29% (27)	-1.2% (-12 to 9)	0.83

Data presented as % (n) or mean ±SD , unless noted otherwise

Fracture characteristics, re-displacements, and re-fractures

In Figure 1, we present an inclusion flowchart. Of the distal metaphyseal fractures: 66 out of 212 (31%) were minimally displaced. 146 out of 212 (69%) were displaced and underwent closed reduction, of which 128 (92%) were deemed stable and randomized between K-wire fixation versus casting, whereas 18 (8%) were unstable and received K-wires. In patients treated without stabilization, re-displacements occurred in 30 out of 67 (45%) reduced distal metaphyseal fractures, whereas 15 out of 66 (23%) of non/minimally displaced distal metaphyseal fractures re-displaced. Re-displacements were accepted in 31 out of 50 (62%) patients.

Figure 1. Inclusion Flowchart



N = number of children, AEC = above-elbow cast, BEC = below-elbow cast, K-wires = Kirschner wires, ESIN = elastic stable intramedullary nail. Re-displacements are presented as: number of re-displacements (total number of children). Accepted re-displacements are presented as: number of accepted re-displacements (total number of re-displacements).

Regarding the diaphyseal fractures: 47 out of 198 (24%) were minimally displaced. 151 out of 198 (76%) were displaced and treated by closed reduction. 127 out of 151 (84%) were deemed stable, whereas 24 out of 151 (16%) were unstable and received intramedullary nails. In patients without stabilization, re-displacements occurred in 44 out of 127 (35%) reduced diaphyseal fractures and 12 out of 47 (26%) non/minimally displaced diaphyseal fractures. Re-displacements were accepted in 39 out of 56 (70%).

Re-fractures occurred in 24 out of 149 diaphyseal fractures (16%) and 18 out of 167 distal metaphyseal fractures (11%). Fourteen diaphyseal re-fractures required re-operation, while only two distal re-fractures required re-operation

Which factors affect the limitation of pro-supination after both-bone forearm fractures in children?

Results of exploratory analysis for factors associated with pro-supination limitation at long-term follow-up are presented in Table 2. Multi-variate linear regression analysis revealed that predictors were: a complete ulnar fracture, older age at trauma and a diaphyseal fracture (Table 3).

Table 2.: Factors associated with limitation in pro-supination at long term FU

Factors		N	Limitation in ROM	Mean difference (95% CI)	P-value
Age at trauma	≤ 10 years	232	4.1° (±11)		
	> 10 years	84	6.7° (±13)	2.5° (-0.6 to 5.7)	0.09
Location Fracture	Distal	169	3.4° (± 8)		
	Diaphyseal	147	6.4° (±15)	-2.9° (-5.7 to-0.3)	0.03
Complete Radius	Yes	166	6.1° (±12)		
	No	148	2.6° (±7)	3.5° (1.3 to 5.7)	0.002
Complete Ulna	Yes	121	7.0° (±13)		
	No	193	2.9° (±8)	4.1° (1.5 to 6.7)	0.002
Distal Re-displacement	Yes	44	4.5° (±10)		
	No	119	2.9° (±8)	1.6° (-1.4 to 4.6)	0.29
Accepted Distal Re-displacement	Yes	28	3.5° (±10)		
	No	135	3.3° (±8)	0.2° (-3.3 to 3.7)	0.92
Diaphyseal Re-displacement	Yes	40	5.9° (±10)		
	No	107	6.6° (±16)	-0.7° (-6.0 to 4.7)	0.81
Accepted Diaphyseal Re-displacement	Yes	31	5.5° (±11)		
	No	116	6.6° (±15)	-1.1° (-6.9 to 4.8)	0.71
Re-fracture	Yes	41	8.0° (±21)		
	No	273	4.4° (±10)	3.6° (-3.0 to 10.2)	0.27
Total		316	4.8° (± 12)		

Table 3. Multi-variate Linear regression analysis: Loss in pro-sup at long term FU

	Unstandardized coefficients		
Model	B	Std. Error	Significance
Complete Ulna	3.4	1.2	0.004
Age at trauma	0.4	0.2	0.047
Diaphyseal location	2.3	1.1	0.048

Do accepted re-displacements in pediatric both-bone forearm fractures lead to inferior functional and radiographic long-term outcomes?

At long-term follow-up, there were no significant differences in outcomes between patients with accepted distal metaphyseal re-displacements versus good alignments (Table 4A and 4B). Patients with accepted diaphyseal re-displacements had greater residual sagittal angulation of the radius than patients with good alignments (p=0.007).

Table 4.A Radiographic long-term outcomes (Accepted re-displacements)

	Accepted re-displacement	Good alignment	Mean difference (95% CI)	P-value
Distal metaphyseal				
Radius - PA	4.9° (±3)	5.0° (±4)	-0.04° (0.8 to -1.6)	0.96
Radius - Lateral	4.2° (±3)	3.7° (±3)	0.4° (-0.9 to 1.8)	0.53
Ulna – PA	5.0° (±3)	4.8° (±3)	0.3° (-1.1 to 1.7)	0.70
Ulna - Lateral	3.1° (±3)	3.5° (±3)	-0.4° (-1.8 to 1.0)	0.58
Diaphyseal fractures				
Radius - PA	9.1° (±3)	9.2° (±3)	-0.1° (-1.3 to 1.1)	0.86
Radius - Lateral	5.6° (±4)	3.8° (±3)	1.8° (0.5 to 3.0)	0.007
Ulna – PA	5.3° (±3)	5.2° (±3)	0.1° (-1.2 to 1.4)	0.88
Ulna - Lateral	4.7° (±3)	4.5° (±3)	0.2° (-0.8 to 1.2)	070

Table 4.B Functional minimum 5-year outcomes (Accepted re-displacements)

	Accepted re-displacement	Good alignment	P-value
Distal metaphyseal fractures			
ABILHAND	41.4 (±1.4)	41.6 (±1.4)	0.59
QuickDASH	4.7 (±9)	4.0 (±8)	0.67
NRS cosmetics	8.7 (±1.5)	8.3 (±2.1)	0.20
Jamar ratio	100.5% (±18)	99% (±20)	0.67
Diaphyseal fractures			
ABILHAND	41.8 (±0.5)	40.5 (±5.3)	0.20
QuickDASH	3.7 (±4.8)	5.5 (±10)	0.35
NRS cosmetics	8.0 (±1.8)	8.4 (±2.0)	0.34
Jamar ratio	94.4% (±17)	98.0% (±16)	0.29

DISCUSSION

This study investigated the following questions: (1) Which factors are associated with a persisting pro-supination limitation after pediatric both-bone forearm fractures? (2) Do accepted re-displacements lead to inferior long-term outcomes?

Factors associated with limitation of pro-supination

Predictors for a pro-supination limitation at long-term follow-up were: a complete ulnar fracture, older age at trauma, and diaphyseal fracture.

In the literature, both-bone fractures are often considered highly unstable. Zamzam et al. stated that predictors for re-displacement of a distal metaphyseal fracture were: a both-bone fracture (odds ratio of 23) and complete displacement of the radius (odds ratio of 25) [20] . In our study, a complete ulnar fracture was very frequently accompanied by a complete radius fracture (86%). Thus, a complete both-bone forearm fracture is likely associated with a pro-supination limitation.

A diaphyseal fracture was associated with a persisting pro-supination limitation. Biomechanically, in a cadaveric study, diaphyseal angular deformities led to more severe pro-supination limitation than distal metaphyseal deformities [21]. Hereby, bony impingement causes a pronation limitation because the interosseous space is encroached during pronation due to dorsal angular deformity of the radius. A supination limitation is seen if there is a central band tightness due to valgus deformity of the ulna. [22] . Moreover, diaphyseal angular deformities located are less likely to remodel because, the nearer the fracture to the physis, the greater the potential for spontaneous correction [11].

Furthermore, older age at trauma was associated with inferior functional long-term outcomes. The capacity for spontaneous remodeling is related to the years of growth remaining. This remodeling potential differs in boys and girls because their physeal closure occurs at 14.5 and 12.9 years, respectively [23].

Distal metaphyseal re-displacements

Previously, one meta-analysis compared the functional outcomes after displaced distal radius fractures between children treated with closed reduction and casting versus K-wire fixation. They found a higher re-displacement rate in the casting group (46% versus 4%) but more complications after K-wiring (16% versus 4%), but no differences in functional outcomes at 3-23 months follow-up [6]. Delft et al. studied 200 consecutive children with displaced metaphyseal fractures: 70% were primarily treated in the emergency room (ER) and 30% in the operating room (OR), for instance, due to complete displacement [24]. Closed reduction was successful in 83% of patients treated in the ER, whereas 17% required subsequent treatment in the OR. Re-displacement occurred in 6% of patients treated in the ER. They advised that distal metaphyseal fractures can be successfully treated by closed reduction and casting in the ER. However, displaced metaphyseal fractures treated in the OR without stabilization resulted in unacceptable re-displacement rates (47%) and should therefore be fixed with K-wires. Unfortunately, this study did not determine the functional outcomes.

In our current study, patients with accepted distal metaphyseal re-displacements did not have inferior long-term outcomes compared to those with good alignments. This illustrates the exceptional potential for remodeling in distal metaphyseal fractures in children. Many previous studies support our findings. Zimmermann et al. studied 232 pediatric distal forearm fractures and found that large displacements (>20° angulation) in children aged <10 years did not influence the long-term outcomes [2]. In a previous study, re-manipulation of re-angulated distal forearm fractures in children <12 years did not improve outcomes at four-year follow-up compared to patients with accepted re-angulations [25]. Crawford et al. accepted distal radius fractures with 100% dorsal translation in 51 children aged <10 years and witnessed excellent outcomes in all children [26]. Orland et al. stated that 27% of closed reductions performed in children <10 years with distal radius fractures are potentially unnecessary [27]. In pediatric distal radius fractures, mean remodeling speeds of 2.4° of angulation per month have been observed [28,29]. In 2005, Wilkins and O'Brien suggested that dorsal angulations up to 30°–35° will remodel adequately in children with five growing years left [30]. Improved awareness of these acceptable deformities in young children may reduce the number of children requiring reduction with sedation [27]. We await the results of the AFIC and CRAFFT trials with great anticipation, in which children aged <11 years with severely displaced distal radius fractures are randomized between cast immobilization alone and closed reduction [31,32].

Diaphyseal re-displacements

Bowman et al. studied 321 children with both-bone diaphyseal fractures: 89% were treated by closed reduction and casting, and 11% underwent surgery, rates similar to ours [7]. In their study, re-displacements occurred in 51% versus 35% in our study. However, they only included fractures with complete cortical disruption.

Price et al. studied the outcomes of 39 children with malunions after severe diaphyseal both-bone forearm fractures with a mean follow-up of 6 years. Complete remodeling occurred in only 12 out of 39 patients, of which 11 were <10 years at trauma [33].

Zionts et al. studied the outcomes of 25 children with displaced diaphyseal forearm fractures treated by closed reduction at a mean age of 13. At one-year follow-up, residual angulations of the radius and ulna of 9° and 8° were seen. The limitation of forearm rotation was correlated with the maximum residual angulations [34]. In 1962 Gandhi et al. stated that mid-shaft angular deformity corrects poorly, resulting in pro-supination limitation [11]. Likewise, Kay et al. stated that midshaft both-bone forearm fractures in children >10 years results in functional deficit more often than is appreciated and therefore, >10° of malalignment in children >10 years old should not be accepted [35]. Jones et al. recommended performing closed reduction for midshaft forearm fractures in children aged ≤8 years with >10° of angulation and in children >9 years with >8° of angulation [36].

Limitations

Our main limitation is the long-term follow-up percentage of 77% of the primarily included children. Nevertheless, our analysis comparing the the included patients to those lost-to-follow-up revealed no significant differences. A second limitation is that the reduction criteria did not differentiate for fracture location or gender, and there were only two age groups (<10 or ≥10 years). A third limitation was that we did not correct for the natural bowing during the radiographic assessment of intramedullary angulation. Bowman et al. corrected for the coronal radial bowing by subtracting six degrees for apex radial measurements [7]. The radius has a mean coronal bowing of 6.0-9.3° and sagittal bowing of 4.7° [7,37].

CONCLUSIONS

Predictors for a persisting pro-supination limitation after a pediatric both-bone forearm fracture are a complete ulnar fracture, older age at trauma, and diaphyseal fracture. Excellent spontaneous remodeling of angular deformity and functional outcomes are seen in distal metaphyseal forearm fractures in children with remaining growth potential. On the contrary, our study reaffirmed the old adage by Hughstone et al. from 1962: *"In midshaft forearm fractures, growth will not correct angular deformity as it does in distal fractures"* [38].

Recommendations for acceptance of angulation

Based on our results and the literature [2,3,7,10,24-29,35,36,39], we recommend the following treatment guidelines:

<u>For distal forearm fractures in boys:</u>

- <9 years, accept ≤30° angulation.
- 9-11 years, accept ≤20° of angulation.
- 11-13 years, accept ≤15° of angulation.
- 13-15 years, accept ≤10° of angulation.
- ≥15 years, accept 5-10° angulation.

<u>For distal forearm fractures in girls:</u>

- <8 years, accept ≤30° angulation.
- 8-10 years, accept ≤20° of angulation.
- 10-12 years, accept ≤15° of angulation.
- 12-14 years, accept ≤10° of angulation.
- ≥14 years, accept 5-10° angulation

<u>For diaphyseal forearm fractures in all children:</u>

- <9 years, accept ≤10° angulation.
- ≥ 9 years, accept ≤8° of angulation.

Treatment of distal metaphyseal forearm fractures

Children with displaced metaphyseal both-bone forearm fractures can be treated by closed reduction and casting without K-wire fixation in the emergency room with excellent long-term outcomes.

If re-displacement occurs, the surgeon should perform family decision-making to discuss accepting re-displacement or performing a re-manipulation (with K-wire fixation).

Consider that tremendous remodeling can be seen in children aged <10 years.

If closed reduction is performed in the operation room due to complete initial displacement, K-wire fixation is recommended to prevent re-displacement.

Treatment of diaphyseal forearm fractures

Displaced diaphyseal forearm fractures, which appear stable after reduction, still re-displace in one-third of cases and show less remodeling potential. Therefore, we recommend performing closed reduction and intramedullary stabilization for diaphyseal complete both-bone fractures in children >10 years.

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PART III:
Malunited forearm fractures in children

Chapter 6

Factors determining outcome of corrective osteotomy for malunited pediatric forearm fractures: a systematic review and meta-analysis

J Hand Surg Eur Vol. 2017 Oct;42(8):810-816.

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ABSTRACT

The aim of this study was to identify predictors of a superior functional outcome after corrective osteotomy for pediatric malunited radius and both-bone forearm fractures. We performed a systematic review and meta-analysis of individual participant data, searching databases up to 1 October 2016. Our primary outcome was the gain in pro-supination seen after corrective osteotomy. Individual participant data of 11 cohort studies were included, concerning 71 participants with a median age of 11 years at trauma. Corrective osteotomy was performed after a median of 12 months after trauma, leading to a mean gain of 77° in pro-supination after a median follow-up of 29 months. Analysis of variance and multiple regression analysis revealed that predictors of superior functional outcome after corrective osteotomy are: an interval between trauma and corrective osteotomy of less than 1 year, an angular deformity of greater than 20° and the use of three-dimensional computer-assisted techniques.

Level of evidence: II.

INTRODUCTION

Displaced forearm fractures in children are commonly treated by closed reduction and cast immobilisation. This treatment carries the risk of re-displacement of the fracture in cast, resulting in malunion [1]. In general, young children with a malunion located close to the most active distal physis have the potential to remodel and have unrestricted function and a satisfactory cosmetic outcome. However, both-bone forearm fractures localized in the distal metaphysis have a high chance (60%) of developing a clinically relevant limitation of forearm rotation in case of more severe angular malalignment (greater than 16°), whereas children with diaphyseal both-bone forearm fractures had a moderate chance of limitation (13-33%) irrespective of the severity of the angular malalignment [2]. Unfortunately, severe malunions in older children have less potential for remodeling, which can result in disappointing clinical outcomes. Nevertheless, there is still no consensus on how much angular deformity is acceptable [3-5]. Although malunions of forearm fractures in children are relatively uncommon, they have a tendency to result in persistent functional impairment [6,7]. For these children, a corrective osteotomy may be considered, but few papers have been published on the outcome of corrective osteotomy for malunited forearm fractures in children. Previous studies have found that corrective osteotomies performed in patients older than 10 years and a time from injury until osteotomy of more than one year showed less favorable results [1,8,9]. Other studies have indicated that the location and type of fracture, the level of pre-operative disability and use of three-dimensional computer-assisted planning techniques may affect functional outcome after corrective osteotomy [7,8,10]. All previous studies have reported only small numbers of patients, limiting the reliability of the results. The aim of this study was to conduct a meta-analysis of individual participant data to provide the best available evidence on determinants of a superior functional outcome after corrective osteotomy for malunited radius or both-bone forearm fractures in children.

MATERIALS AND METHODS

We performed a meta-analysis of individual participant data (IPD), which we reported according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis of Individual Participant Data (PRISMA-IPD) statement [11]. Prior to starting the systematic search, we defined the research question, inclusion and exclusion criteria, treatment of interest and outcomes of interest. The protocol of this meta-analysis can be accessed on PROSPERO with trial registration number: PROSPERO CRD42015023964.

We included prospective and retrospective cohort studies containing data on functional outcomes (raw data published or supplied on request). Eligible participants were children with post-traumatic malunion of the radius or both forearm bones, who underwent a corrective osteotomy because of impairment in pronation and/or supination. Patients with an age at trauma of 16 years or younger; an age at corrective osteotomy of 18 years or younger; and an interval between trauma and corrective osteotomy of greater than 6 weeks, were included. We excluded participants with complex fractures (Monteggia, Galeazzi, intra-articular or open fractures) and those treated by callus osteoclasis. Our treatment of interest was corrective osteotomy, subdividing conventional corrective osteotomies using two-dimensional radiographic planning and three-dimensional computer-assisted corrective osteotomies. Our primary outcome of interest was the gain in forearm rotation measured at final follow-up after corrective osteotomy. Minimum follow-up required was six months after corrective osteotomy. Factors possibly influencing the gain in range of motion observed after corrective osteotomy were analyzed. Data were sought for the following variables: age at injury; age at osteotomy; time from trauma until osteotomy; level of malunion; single or both-bone fracture; degree of angular deformity; and the use of three-dimensional computer assisted techniques.

To identify all studies regarding the outcome after corrective osteotomy for post-traumatic malunions of the forearm in children, the following databases were searched: Medline, Embase, Web-of-Science, Scopus, Cinahl, Pubmed publisher, Cochrane and Google Scholar for articles published before March 21st, 2016. We repeated the search on the 1st of October 2016. The complete search strategy is described in Appendix 1. The search was limited to articles written in English, Dutch or German. Two reviewers (KCR and JWC) assessed the studies for relevance by initially reviewing the titles and abstracts and categorizing the papers in folders of relevancy within an EndNote library. All studies containing functional outcomes after corrective osteotomy of the radius or forearm were deemed potentially relevant. Next, the full manuscript was retrieved to determine appropriateness, by verifying if the studies met the inclusion and exclusion criteria. Any disagreements were resolved by consensus or consultation of a third reviewer. The references of the retrieved studies were scanned to identify additional relevant publications missed by the initial search.

The included studies were evaluated for their methodological quality by two authors (KCR and MMJW) independently. The Methodological Index for Non-Randomized Studies score (MINORS) was utilized for quality assessment and is provided in Appendix 2 [12]. Any disagreements were resolved by consensus or consultation of a third reviewer (JWC).

Individual participant data were extracted from the included studies. If data were unavailable, authors were contacted and raw data were requested. In additional data provided by authors, angular deformities were measured on original radiographs. These additional measurements were added to the data sheet. Intra-class correlation range was determined. Van Geenen et al. anonymously supplied radiographs of 19 eligible participants, in which we measured the angular deformities with an intra-class correlation range of 0.91-0.99 [1]. Walenkamp et al. also provided raw data, supplied in Appendix 3 [13]. Within the included studies, participants' raw data were screened and only participants meeting the inclusion criteria were included in our meta-analysis. Reasons for exclusion involved other indications for corrective osteotomy than deficit in range of motion; an age at trauma over 16 years of age and/or an age at osteotomy over 18 years of age. Data extraction was verified by the second reviewer. The available individual participant data were assembled and analyzed as if they were results from one study.

We performed one-way analysis of variance (ANOVA) with clinically relevant subgroups for each factor we investigated. Subgroups were created for: 1) Age at trauma (younger than 10 years versus 10 years and older); 2) Age at corrective osteotomy: (younger than 13 years versus 13 years and older); 3) Time from trauma until corrective osteotomy (within one year after trauma versus one year after trauma or more); 4) Level of malunion (in the proximal, middle or distal third); 5) Severity of angular deformity (under 20 degrees versus 20 degrees or more); 6) Type of corrective osteotomy (3-D computer assisted corrective osteotomy versus conventional corrective osteotomy using 2-D radiographic planning); and 7) Pre-operative complaint (predominant deficit in pronation versus predominant deficit in supination). Performing a corrective osteotomy within one year after trauma was defined as early management, whereas more than one year was defined as late management [9]. Subgroups dividing age at trauma were set at below or above 10 years in accordance with an earlier study [1].

We set the cut-off for age at osteotomy at below or above 13 years of age, due to a mean time from trauma until osteotomy of 3 years in a previous study [1]. Severity of angulation was subdivided at below or above 20 degrees, because in a cadaveric study, there was a statistically significant and functionally important loss of forearm rotation if angulation exceeded 20 degrees [14].

Next, multivariate regression analysis was performed to study the effect of the various factors on the gain in range of motion after corrective osteotomy, using a stepwise backward procedure. We reported medians and interquartile range (IQR) for non-parametric variables, and means and standard deviations (SD) for normally distributed variables. The 95% confidence intervals (CI) were calculated using the formula: $\chi \pm 1.96 (\sigma/\sqrt{n})$, with χ = mean; a confidence coefficient of 1.96 for a confidence level of 95%; σ = standard deviation of sample; (square root of) n = sample size. Statistical analyses were performed with Statistical Package for Social Sciences (SPSS). P values < 0.05 were considered statistically significant.

RESULTS

Our search resulted in 1423 potentially eligible studies, of which 22 full text articles were analyzed for eligibility. 12 studies met the inclusion criteria [1,7-9,13,15-21]. Two studies by Meier et al. contained duplicate participants [18,19]. Therefore 11 studies with individual participant data were included in the IPD meta-analysis, shown in the flow diagram in Figure 1. Assessment of methodological quality the included studies is provided in Table 1.

Table 1. MINORS methodological quality

Study	Clear aim	Inclusion Patients	Collection data	Appropriate end points	Assessment end points	Follow up period	Loss to follow-up	Calculation study size	Total
Trousdale	1	2	0	2	0	2	1	0	8
Meier	2	2	0	2	0	2	2	0	10
Price	2	2	0	2	0	2	2	0	10
Van Geenen	1	2	0	2	0	2	2	0	10
Murase	2	2	2	2	1	2	2	0	13
Nagy	2	0	0	2	1	2	2	0	9
Chia	2	2	0	2	0	2	2	0	10
Miyake	2	2	0	2	1	2	1	0	10
Kataoka	2	2	0	2	1	2	2	0	11
Boeckers	1	2	0	2	0	1	2	0	8
Walenkamp	2	2	0	2	1	1	0	0	8

† The items are scored 0 (not reported), 1 (reported but inadequate) or 2 (reported and adequate).

The included studies contained 158 participants who were treated for a symptomatic radius or both-bone forearm malunion by corrective osteotomy, of which 71 participants met the inclusion criteria. The participants fulfilling the eligibility criteria are reported in Table 2 with notes on the reasons for exclusion. The commonest reasons for exclusion were failure to match the inclusion criteria for age, or due to alternative indications for corrective osteotomy, such as a painful distal radio-ulnar joint, cosmetic appearance or a congenital deformity. Details on degree of radiographic angular deformity were provided in 49 out of 71 participants. Corrective osteotomies using three-dimensional computer-assisted techniques were performed in four out of 11 studies.

Table 2. Extraction of individual participant data

Year	Study	Eligible participants	Total Participants	Design	Excluded (participant number)	Reasons for exclusion**
1995	Trousdale	14	27	Retrospective	3,6,10,14,15,19,21-27	Age, Other
2003	Meier (GER)	6	14	Retrospective	All but 4,8-11,14	Other, Age, TUO,
2006	Price	9	9	Retrospective	None	-
2007	van Geenen	17	21	Retrospective	6,12,20,21	TUO, FU, Age
2008	Murase*	4	22	Prospective	All but 5,8,9,14	Age
2008	Nagy	7	17	Retrospective	2,6,7, 11-17	Age, Other
2011	Chia	1	6	Retrospective	All but 4	Age
2012	Miyake*	9	20	Retrospective	1,4-7,13,15-18,20	Age
2013	Kataoka*	1	9	Retrospective	All but 5	Age at trauma
2014	Boeckers (GER)	1	5	Retrospective	All but 4	FU, TUO
2015	Walenkamp*	2	8	Retrospective	All but 4,8	Age, Other
2016	Current study	71	158	Meta-analysis	-	-

* = 3-D computer assisted corrective osteotomy, GER = German.
**Age = age at trauma above 16 and/or osteotomy above 18 years, TUO = time until osteotomy < 6w, FU = follow-up < 6m

A summary of characteristics and outcomes of the individual studies is presented in Table 3, with medians for age at trauma, time until osteotomy and duration of follow-up and mean functional and radiographic measurements. A full overview of extracted individual participant data is supplemented in Table S2 on pages 112 and 113.

Table 3. Study characteristics

Year	Study	Age at trauma	Years until osteotomy	Months follow-up	Angulation	Pre-op ROM	ROM at FU	Gain in ROM	Complications
1995	Trousdale	11	3	61	NR	78°	132°	53°	5
2003	Meier	11	1	13	NR	76°	159°	83°	1
2006	Price	7	1	22	31°	63°	165°	102°	2
2007	van Geenen	9	2	26	30°	34°	120°	86°	1
2008	Murase*	11	4	22	18°	51°	144°	93°	1
2008	Nagy	12	4	41	18°	86°	137°	51°	0
2011	Chia	14	1	42	20°	130°	175°	45°	0
2012	Miyake*	11	4	30	22°	57°	146°	90°	0
2013	Kataoka*	4	7	22	35°	70°	130°	60°	0
2014	Boeckers	13	0,1	7	NR	90°	180°	90°	0
2015	Walenkamp*	1	4	18	14°	103°	158°	55°	0
2016	Current study	11	1,0	29	25°	63°	140°	77°	10

* = 3-D computer assisted corrective osteotomy, NR = Not Reported.

Characteristics of Individual Participant Data

The majority of participants were male (61%). Fractures of both forearm bones were seen in 45 out of 71 participants (63%). The malunions were located in the proximal third in 15 participants (21%), the middle third in 44 (62%) and the distal third in 12 (17%). Included participants had a median age at trauma of 11 years (IQR 8 to 13). Median age at corrective osteotomy was 13 years (IQR 11 to 16). Median time from trauma until osteotomy was 12 months (IQR 6 to 48). Functional outcome at final follow-up was measured at a median time of 29 months (IQR 16 to 37) after corrective osteotomy. As pre-operative complaint, 20 predominately had a deficit in pronation, 34 predominately had a deficit in supination and 17 had a similar deficit in both pro- and supination. Corrective osteotomies using three-dimensional computer-assisted techniques were performed in 16 participants, whereas 55 participants underwent conventional corrective osteotomy using two-dimensional pre-operative planning with standard radiographs. There was a complication rate of 14%, which primarily consisted of superficial infection or transient dysesthesia of the radial sensory nerve. There were no major complications.

Results of syntheses

Overall, there was a mean pre-operative forearm rotation of 63° (95% CI: 55° to 70°). At final follow-up, there was a mean forearm rotation of 140° (132° to 148°) indicating that corrective osteotomy provided a mean gain in forearm rotation of 77° (68° to 86°). Results of one-way analysis of variance (ANOVA) are presented in Table 4 showing comparisons of outcomes of clinically relevant subgroups with regards to our primary outcome, the gain in forearm rotation.

We found the following statistically significant differences during ANOVA: Children who underwent corrective osteotomy at an age younger than 13 years had a mean gain of 87° (74° to 101°) in forearm rotation, versus a mean gain of 68° (56° to 80°) in children aged 13 years and older ($p = 0.031$). Participants who underwent corrective osteotomy within one year after trauma gained 93° (80° to 106°) versus 61° (50° to 72°) in those who underwent osteotomy more than one year after trauma ($p < 0.001$). Participants who had an angular deformity of less than 20 degrees had a mean gain in forearm rotation after corrective osteotomy of 59° (45° to 74°) versus a mean gain of 97° (85° to 108°) in those with 20 degrees of angulation or more ($p < 0.001$).

ANOVA revealed that level of malunion was not statistically significantly associated with a higher gain in pro-supination. An additional Independent Sample's T-test was performed comparing malunions located in the middle third versus malunions located in the proximal and distal third, revealed a gain of respectively 84° (72° to 95°) vs. 66° (51° to 81°) in pro-supination ($p = 0.057$).

Multi-variate regression analysis revealed that a shorter time until osteotomy, a greater angular deformity and the use of three-dimensional computer assisted techniques were factors associated with a greater gain in forearm rotation (p-values are respectively 0.002; 0.044; and 0.042). The results of multiple regression analysis, including Beta values and standard errors, are presented in Table 5. There was an R square of 0.35.

Table 4. ANOVA: Effect of factors on gain in pro-supination.

Factor		N	Pre-op ROM (95% CI)	P =	ROM at FU (95% CI)	P =	Gain in ROM (95% CI)	P =
Age at trauma	<10 years	28	57° (46° to 69°)	0.23	132° (118° to 145°)	0.11	74° (58° to 90°)	0.64
	≥10 years	43	66° (57° to 77°)		145° (135° to 156°)		79° (67° to 90°)	
Age at osteotomy	< 13 years	33	53° (42° to 65°)	0.013	141° (128° to 154°)	0.87	87° (74° to 101°)	0.031
	≥13 years	38	71° (62° to 81°)		139° (128° to 150°)		68° (56° to 80°)	
Time until osteotomy	< 1 year	36	61° (50° to 73°)	0.69	154° (144° to 164°)	<0.001	93° (80° to 106°)	<0.001
	≥ 1 year	35	64° (55° to 74°)		125° (114° to 137°)		61° (50° to 72°)	
Location of malunion	Proximal	15	50° (32° to 68°)	0.08	113° (96° to 130°)	0.003	63° (43° to 84°)	0.16
	Middle	44	63° (54° to 73°)		147° (137° to 157°)		84° (73° to 95°)	
	Distal	12	63° (55° to 70°)		146° (126° to 166°)		69° (43° to 95°)	
Boned malunited	Single	26	67° (55° to 80°)	0.40	142° (129° to 155°)	0.66	75° (60° to 90°)	0.77
	Both-bone	45	60° (51° to 70°)		138° (128° to 149°)		78° (66° to 90°)	
Angulation	<20°	18	70° (54° to 86°)	0.030	129° (109° to 149°)	0.08	59° (45° to 74°)	<0.001
	≥20°	31	50° (38° to 61°)		146° (136° to 156°)		97° (85° to 108°)	
Technique	Conventional	55	63° (54° to 72°)	0.88	138° (128° to 148°)	0.43	75° (64° to 85°)	0.41
	3-D Assisted	16	62° (48° to 76°)		146° (129° to 162°)		84° (64° to 104°)	
Complaint	Pro- deficit	34	65° (54° to 76°)	0.18	136° (123° to 149°)	0.74	71° (58° to 83°)	0.42
	Sup- deficit	20	77° (64° to 90°)		139° (124° to 154°)		63° (45° to 80°)	
Total		71	63° (55° to 70°)		140° (132° to 148°)		77° (68° to 86°)	

ROM: range of motion; CI: confidence intervals; FU: follow-up; 3-D: three-dimensional.

Table 5. Multiple regression analysis

Model	B	Std. Error	Significance
	Unstandardized coefficients		
(Constant)	62.1	15.1	0.000
Months until osteotomy	-0.45	0.14	0.002
Angulation	0.95	0.46	0.044
3-D techniques	24.3	11.6	0.042

R square: 0.345, adjusted R square 0.302

DISCUSSION

Comparison to literature

In the literature, recommendations on indications for corrective osteotomy have been based on age and location of the malunion, severity of functional impairment and/or severity of angular deformity. Prommersberger et al. stated that in the case of functional disability, there is an indication for corrective osteotomy over the age over twelve in malunion of a fracture located in the distal third, and over the age of five in gross deformity of fractures to the midshaft of the forearm [22]. Others stated that an early corrective osteotomy is justified in patients with an established malunion with considerable functional impairment (pro-supination of less than 50-60% of normal)[1]. Price et al. recommended to perform corrective osteotomy in forearm shaft malunions with angulations of greater than 30 degrees as soon as possible; and to wait at least six months in malunions with angulations ranging from 20-30 degrees, because the greatest amount of remodeling occurs in the first six months [21].

Previous studies have generally suggested that children gain more in range of motion (ROM) if corrective osteotomy is performed at a younger age. It is suggested that this is due to the potential for residual bone deformities to improve with additional skeletal growth [1,7]. In our IPD meta-analysis, ANOVA revealed that both a younger age at osteotomy and a shorter time until osteotomy were associated with a better functional outcome. Logically, there was an overlap between these two groups, because participants with a shorter time until osteotomy often had a younger age at osteotomy than participants with a longer time until osteotomy. However, multiple regression analysis, which simultaneously studies the relationship between multiple factors, revealed that a shorter time until osteotomy is associated with a higher functional outcome, and this achieved statistical significance. This was not the case with a younger age at osteotomy.

Previous studies also found that a longer time from trauma until osteotomy compromised functional gain, which was thought to be the result of secondary joint changes and soft-tissue contractures [1,9].However, the presence of these soft-tissue contractures is yet to be proven. In a previous study, children who had a persisting deficit in pro-supination exceeding 40° at a follow-up beyond 6 months after fracture of both forearm bones underwent MRI analysis, which did not reveal contractures of the interosseous membrane [23]. The question remains whether the contractures did not exist, or whether they were not detectable on MRI. In our IPD meta-analysis, a shorter time until osteotomy was the most decisive factor in predicting a superior functional outcome, which does suggest a role of secondary joint changes and soft-tissue contractures.

One previous study analyzed the effect of location of the malunion and the outcome after corrective osteotomy and found no statistically significant effect [1]. In our IPD meta-analysis, we saw a moderate trend for the most favorable results after corrective osteotomies for malunions located in the middle third and the poorest results in proximal malunions; this did not achieve statistical significance (p = 0.057). Although a recent cadaveric study showed that dorsal tilt up to 30° did not lead to any significant restriction in forearm pro-supination [24], most studies have shown that angular deformity plays an important role in the limitation of forearm rotation [2,14,25-27]. In our IPD meta-analysis greater pre-operative angulation was associated with superior functional outcomes after corrective osteotomy. Moreover, a previous study advocated that improvement in ROM was greater in those who predominately had a supination deficit as pre-operative complaint [7]. This was not supported by our IPD meta-analysis.

In a previous study, computer-assisted 3D planning was found to improve functional results in patients with symptomatic radius malunions [13,28]. In our meta-analysis, the use of 3-D computer-assisted techniques also had a statistically significant effect on functional outcome.

Strengths and weaknesses

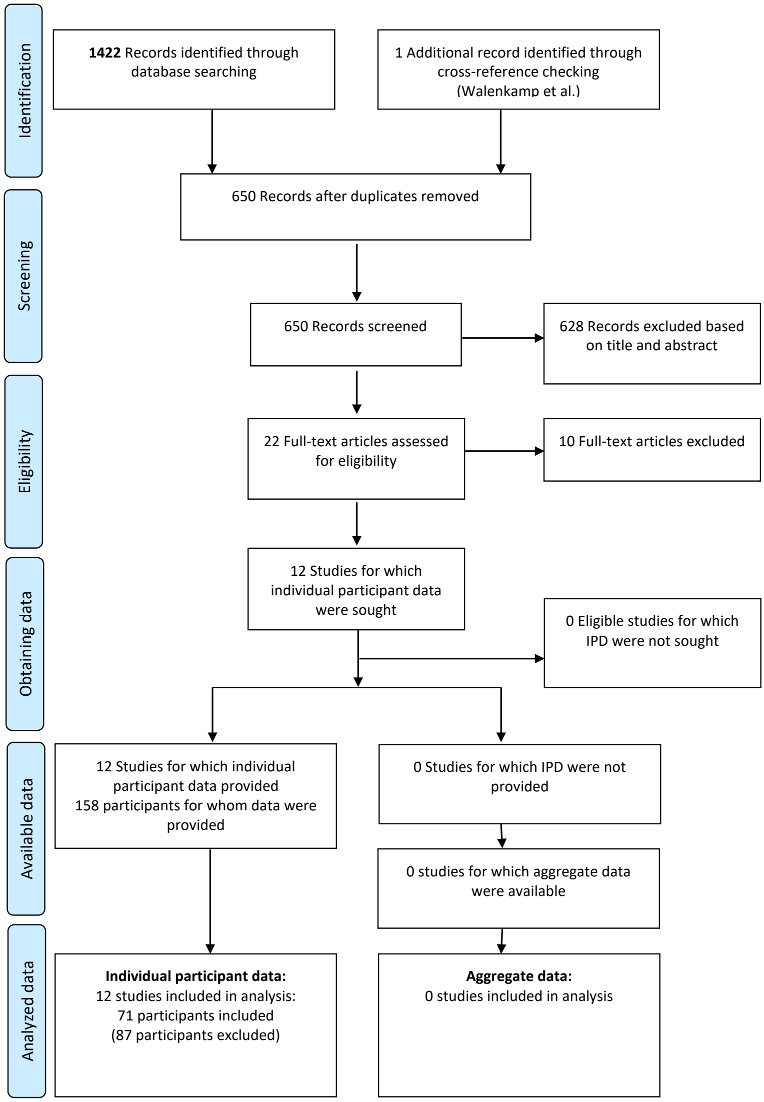
The main strength of this study is the access to individual participant data, which provided the opportunity to analyze a higher number of patients, resulting in several recommendations. A weakness of this meta-analysis is that the majority of the included studies were of retrospective nature. Furthermore, patient reported outcomes measures were not reported in the majority of included studies. Also, there were no control groups, so there is no possibility to compare functional outcomes with those who did not undergo a corrective osteotomy for their post-traumatic forearm malunion. Lastly, we included isolated radius fractures as well as fractures of both forearm bones in our IPD meta-analysis. However, we found no statistically significant difference in the gain of function after corrective osteotomy when comparing isolated radius and both-bone forearm fractures.

CONCLUSIONS

This meta-analysis of individual participant data provides recommendations which can facilitate decision making when considering corrective osteotomy for malunited pediatric fractures of the radius or both forearm bones. Based on this meta-analysis, predictors of a superior functional outcome are: an interval between trauma and corrective osteotomy of less than one year; an angular deformity greater than 20 degrees; and the use of three-dimensional computer assisted techniques.

SUPPLEMENTARY FIGURES AND TABLES

Figure S1. PRISMA flow diagram



Supplementary table S2: Overview of extracted individual participant data

Year	Study	N	Sex	Fracture location	Both-bone fracture	Angulation	Age trauma	Age osteotomy	Years until osteotomy	Follow-up (months)	Pre-op pronation	Pre-op supination	Pre-operative ROM	Pronation at FU	Supination at FU	Pro-sup at FU	Gain in ROM	Use of 3-D technique	Complication	Supplied on request
1995	Trousdale	1	F	Distal	Y	NR	13	13	0.7	35	72	30	102	85	85	170	68	N	N	N
1995	Trousdale	2	M	Distal	Y	NR	16	17	1	180	20	75	95	60	70	130	35	N	Y	N
1995	Trousdale	4	F	Proximal	N	NR	12	13	0.4	19	20	90	110	45	85	130	20	N	N	N
1995	Trousdale	5	M	Middle	Y	NR	9	10	0.4	84	10	10	20	90	90	180	160	N	Y	N
1995	Trousdale	7	F	Proximal	Y	NR	16	17	1	34	65	15	80	85	85	170	90	N	N	N
1995	Trousdale	8	M	Distal	N	NR	13	13	0.2	31	15	30	45	85	70	155	110	N	N	N
1995	Trousdale	9	F	Middle	N	NR	16	17	0.7	49	75	40	115	75	75	150	35	N	Y	N
1995	Trousdale	11	M	Distal	Y	NR	8	10	2	15	90	0	90	85	40	125	35	N	N	N
1995	Trousdale	12	M	Distal	Y	NR	7	18	11	65	80	5	85	70	5	75	-10	N	Y	N
1995	Trousdale	13	M	Middle	Y	NR	11	13	2	14	0	85	85	90	90	180	95	N	N	N
1995	Trousdale	16	M	Proximal	N	NR	11	17	6	120	20	50	70	50	-5	45	-25	N	Y	N
1995	Trousdale	17	M	Proximal	Y	NR	6	12	6	80	45	0	45	60	35	95	50	N	N	N
1995	Trousdale	18	M	Proximal	N	NR	4	9	6	90	85	15	100	75	45	125	25	N	N	N
1995	Trousdale	20	F	Proximal	Y	NR	12	16	4	36	10	45	55	85	30	115	60	N	N	N
2003	Meier	4	F	Distal	N	NR	14	14	0.4	19	70	30	100	100	90	190	90	N	N	N
2003	Meier	8	M	Middle	NR	NR	5	6	0.8	12	50	30	80	45	70	115	35	N	N	N
2003	Meier	9	F	Middle	NR	NR	11	14	3	8	40	30	70	80	80	160	90	N	Y	N
2003	Meier	10	F	Middle	NR	NR	10	10	0.2	17	20	20	40	70	90	160	120	N	N	N
2003	Meier	11	F	Middle	NR	NR	10	12	2	6	0	90	90	90	70	160	70	N	N	N
2003	Meier	14	M	Middle	NR	NR	15	16	0.3	16	45	30	75	90	80	170	95	N	N	N
2006	Price	1	M	Middle	Y	32	5	6	0.8	7	20	0	20	90	90	180	160	N	N	N
2006	Price	2	M	Middle	Y	45	6	7	0.6	16	45	25	70	50	90	140	70	N	N	N
2006	Price	3	M	Middle	Y	30	5	5	0.3	6	30	30	60	80	90	170	110	N	N	N
2006	Price	4	F	Middle	Y	35	10	10	0.6	9	20	20	40	70	90	160	120	N	N	N
2006	Price	5	M	Middle	N	34	5	6	0.8	20	20	20	40	90	90	180	140	N	N	N
2006	Price	6	F	Middle	N	30	5	5	0.3	60	45	45	90	70	90	160	70	N	N	N
2006	Price	7	F	Middle	Y	15	11	12	1	58	30	-10	20	45	90	135	115	N	N	N
2006	Price	8	M	Middle	N	25	8	9	0.3	12	45	90	135	90	90	180	45	N	Y	N
2006	Price	9	M	Middle	Y	30	5	5	0.3	13	0	90	90	90	90	180	90	N	Y	N
2007	van Geenen	1	F	Middle	Y	NR	9	11	2	59	20	10	30	45	45	90	60	N	N	Y
2007	van Geenen	2	M	Distal	N	40	6	7	0.3	9	40	0	40	70	80	150	110	N	N	Y
2007	van Geenen	3	F	Middle	Y	40	2	7	5	12	90	-60	30	45	80	125	95	N	N	Y
2007	van Geenen	4	F	Distal	N	23	4	4	0.2	12	0	35	35	70	80	150	115	N	N	Y
2007	van Geenen	5	M	Proximal	N	45	10	13	3	51	10	10	20	45	60	105	85	N	N	Y
2007	van Geenen	7	F	Proximal	N	13	5	12	7	33	60	-30	30	70	30	100	70	N	N	Y
2007	van Geenen	8	M	Middle	Y	10	13	16	3	51	20	70	90	60	90	150	60	N	N	Y
2007	van Geenen	9	M	Distal	N	35	15	15	0.2	17	20	40	60	70	80	150	90	N	N	Y
2007	van Geenen	10	F	Proximal	Y	30	10	14	5	21	10	0	10	50	30	80	70	N	Y	Y
2007	van Geenen	11	M	Middle	Y	33	11	11	0.2	23	5	5	10	70	80	150	140	N	N	Y
2007	van Geenen	13	M	Proximal	N	20	9	11	2	22	5	20	25	70	20	90	65	N	N	Y

Year	Study	N	Sex	Fracture location	Both-bone fracture	Angulation	Age trauma	Age osteotomy	Years until osteotomy	Follow-up (months)	Pre-op pronation	Pre-op supination	Pre-operative ROM	Pronation at FU	Supination at FU	Pro-sup at FU	Gain in ROM	Use of 3-D technique	Complication	Supplied on request
2007	van Geenen	14	F	Proximal	Y	45	8	8	0.3	26	20	20	40	60	80	140	100	N	N	Y
2007	van Geenen	15	F	Proximal	Y	23	8	8	0.7	22	20	10	30	60	80	140	110	N	N	Y
2007	van Geenen	16	M	Middle	Y	15	13	14	0.5	24	10	-10	0	10	10	20	20	N	N	Y
2007	van Geenen	17	M	Proximal	N	20	12	13	2	21	0	45	45	60	80	140	95	N	N	Y
2007	van Geenen	18	M	Distal	Y	20	14	15	2	19	45	40	85	85	80	165	80	N	N	Y
2007	van Geenen	19	F	Proximal	Y	27	11	12	1	15	45	-45	0	80	20	100	100	N	N	Y
2008	Nagy	1	F	Middle	Y	15	12	18	7	119	15	75	90	70	80	150	60	N	N	N
2008	Nagy	3	M	Middle	Y	20	14	16	2	12	10	75	85	60	80	140	55	N	N	N
2008	Nagy	4	F	Middle	Y	16	13	14	0.9	6	5	90	95	50	90	140	45	N	N	N
2008	Nagy	5	F	Distal	Y	16	15	16	2	37	40	60	100	65	50	115	15	N	N	N
2008	Nagy	8	M	Middle	Y	16	14	18	4	46	70	0	70	60	90	150	80	N	N	N
2008	Nagy	9	M	Middle	Y	30	11	13	2	18	90	10	100	80	90	170	70	N	N	N
2008	Nagy	10	M	Middle	Y	10	7	18	11	46	60	0	60	25	70	95	35	N	N	N
2008	Murase	5	F	Middle	Y	12	8	16	8	24	80	-30	50	80	10	90	40	Y	Y	N
2008	Murase	8	M	Middle	Y	33	12	12	0.5	22	10	15	25	95	80	175	150	Y	N	N
2008	Murase	9	M	Middle	N	22	13	14	0.8	23	60	-20	40	70	70	140	100	Y	N	N
2008	Murase	14	M	Middle	Y	6	11	18	7	14	80	10	90	90	80	170	80	Y	N	N
2011	Chia	4	M	Proximal	N	20	14	16	1.3	42	65	65	130	90	85	175	45	N	N	N
2012	Miyake	2	M	Middle	N	22	12	13	0.8	24	60	-20	40	70	70	140	100	Y	N	N
2012	Miyake	3	F	Middle	Y	12	7	15	8	24	80	-30	50	80	0	80	30	Y	N	N
2012	Miyake	8	M	Middle	Y	33	11	12	0.4	24	10	15	25	80	95	175	150	Y	N	N
2012	Miyake	9	M	Middle	Y	16	13	18	6	33	70	20	90	70	90	160	70	Y	N	N
2012	Miyake	10	M	Middle	N	14	16	16	0.7	48	60	10	70	70	80	150	80	Y	N	N
2012	Miyake	11	F	Middle	Y	13	11	16	5	37	60	0	60	90	90	180	120	Y	N	N
2012	Miyake	12	M	Middle	Y	23	13	17	4	24	0	45	45	70	90	160	115	Y	N	N
2012	Miyake	14	M	Middle	Y	27	10	11	0.8	32	60	0	60	80	80	160	100	Y	N	N
2012	Miyake	19	F	Middle	Y	35	3	11	8	28	90	-20	70	90	20	110	40	Y	N	N
2013	Kataoka	5	F	Middle	Y	35	4	11	7	22	90	-20	70	90	40	130	60	Y	N	N
2014	Boeckers	4	M	Distal	N	NR	13	13	0.1	7	90	0	90	90	90	180	90	N	N	N
2015	Walenkamp	4	M	Middle	Y	12	14	18	4	13	40	40	80	60	75	135	55	Y	N	Y
2015	Walenkamp	8	M	Middle	Y	16	12	13	0.8	23	90	35	125	90	90	180	55	Y	N	Y

NR = Not reported, M = Male, F = Female, Y = Yes, N = No.

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Chapter 7

Outcomes of 3D corrective osteotomies for pediatric malunited diaphyseal both-bone forearm fractures

J Hand Surg Eur Vol. 2022 Feb;47(2):164-171.

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ABSTRACT

Closed treatment of pediatric diaphyseal forearm fractures carries the risk of re-displacement, which can lead to symptomatic malunions, as growth will not correct angulation deformity as it does in metaphyseal fractures. The purpose of this prospective cohort study was to evaluate the outcomes after 3D-planned corrective osteotomy with patient-specific surgical guides for pediatric malunited forearm fractures causing impaired pro-supination. Our primary outcome measure was the gain in pro-supination at 12 months follow-up. Fifteen patients with a mean age at trauma of 9.6 years and time until osteotomy of 5.9 years were included. Pre-operatively, patients displayed a mean pro-supination of 67° corresponding to 44% of contralateral. At final follow-up, this improved to 128°, achieving 85% of contralateral. Multivariate linear regression analysis revealed that predictors of greater functional gain after 3D corrective osteotomy are severe pre-operative impairment in pro-supination, shorter interval until 3D corrective osteotomy and greater angulation of the radius.

Level of evidence: IV

INTRODUCTION

Diaphyseal forearm fractures account for 15% of pediatric fractures [1]. Closed reduction and cast immobilization continue to be a major treatment method, due to the great remodeling ability of pediatric fractures [2]. However, fracture re-displacement occurs in 34% of displaced diaphyseal both-bone forearm fractures in children [3], leading to malunion and decreased forearm rotation [4], which may need a corrective osteotomy [5]. Previously, a corrective osteotomy is indicated when pro-supination is less than 50–60% of the contralateral side [5]. Price and Knapp (2006) recommended performing corrective osteotomy in forearm shaft malunions with angulations greater than 30° as soon as possible, and to wait at least 6 months in malunions with angulations ranging from 20–30° [7].

A corrective osteotomy is challenging, due to angular deformity of both radius and ulna in coronal, sagittal and axial planes [8, 9]. 3D planned corrective osteotomy can aid in accurate correction of forearm malunions [10]. Using this method patient-specific drilling and cutting guides can be 3D printed to transfer the planned osteotomy plane to the patient's bony anatomy during surgery.

The purpose of this prospective cohort study was to evaluate the clinical outcomes after 3D-planned corrective osteotomy using patient-specific guides for malunited diaphyseal both-bone forearm fractures, sustained during childhood. Our main research questions were: what gain in forearm rotation can be achieved after 3D corrective osteotomy for pediatric malunited forearm fractures and which factors are associated with greater functional gain?

METHODS

This prospective cohort study was performed at a tertiary referral hospital (Erasmus University Medical Center, Rotterdam, the Netherlands). Ethical approval for this study was obtained from the Medical Ethical Testing Committee (reference 52987.078.15). Written informed consent was obtained from all participants and their parents before the study. Our research protocol was registered in the National Trial Register (reference number 6324). This study was reported according to the guidelines by the STROBE statement [11].

Participants

Inclusion criteria were a forearm malunion after a diaphyseal both-bone forearm fracture, sustained during childhood (<18 years), resulting in impaired pro-supination (pronation or supination of <50°), with unsatisfactory improvement after conservative treatment and a minimum age of ten years. Diaphysis was defined as the segment of the bone between 20% and 80% of its entire length [12] . Exclusion criteria were a traumatic osseous deformity of the contralateral forearm and a congenital or developmental deformity of the contralateral or affected forearm (such as radial or ulnar longitudinal deficiency, radioulnar synostosis, congenital radial head dislocation and Madelung deformity) [13]. Authors differentiated between traumatic and congenital deformity by inquiring about the manifestation and evolution of the forearm complaints, presence of trauma in previous medical history and studying clinical and radiographic appearance of both forearms. Patients' demographics were collected at baseline: age at trauma, age at osteotomy, sex, side of malunion, dominant arm, occurrence of a re-fracture in previous medical history, previous (operative) treatment of forearm malunion.

Pre-operative planning

In collaboration with Materialise (Materialise N.V., Leuven, Belgium) planning of the corrective osteotomy was performed according to the following steps: First, a CT scan of both forearms was obtained (0.7 mm slice thickness). Scans were performed with the patient prone with the shoulders in maximal abduction, elbows in maximal extension and forearms as close as possible to neutral (Superman position). A virtual model of the malunited forearm bones was superimposed on a mirrored version of the contralateral forearm bones. Next, the location and degree of deformity were determined. Virtual cutting planes to perform the osteotomy were selected to best match the contralateral side, whilst taking surgical approaches into account. Lastly, patient-specific drilling and cutting guides were 3D-printed and sterilized to be used during surgery (Figure 1). The drilling guides were designed with the rotational and angular correction built-in so that once the osteotomies are completed, the placement of screws determines the correction [14]. Also, real-sized models of the forearm bones of the pre-operative situation and planned correction were 3D-printed and used for orientation during surgery and, if needed, for bending of the plates (Figure 2).

Figure 1. 3D printed patient-specific drilling and cutting guides.

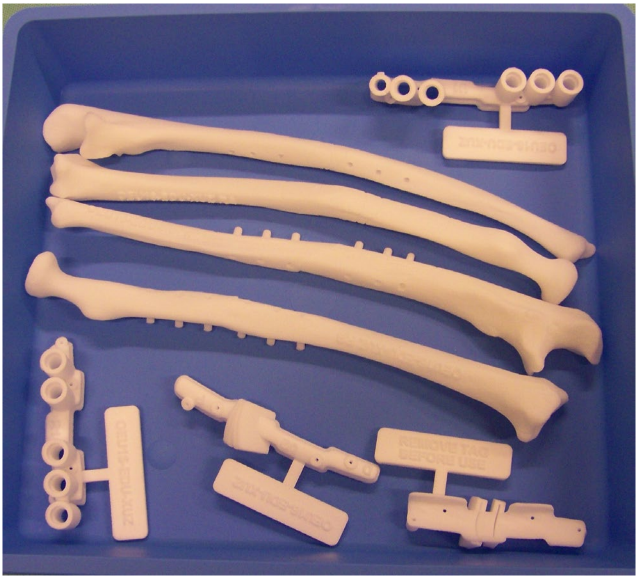
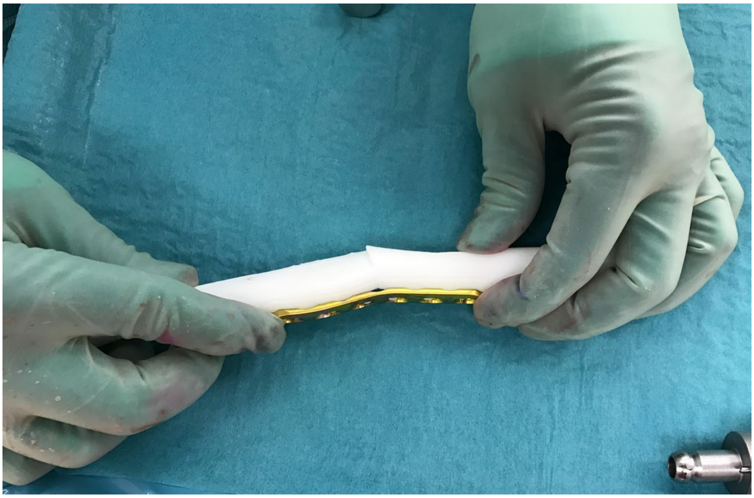


Figure 2. 3D printed real-sized model of the planned correction, for bending of the plates.



Surgical approach

The radius was exposed using a volar Henry approach. Precise positioning of the patient-specific guides was realized by obtaining a wide exposure, searching for recognizable bony landmarks, comparing the intra-operative guide fitting with the 3D-printed bone models as reference, and use of fluoroscopy. The drilling guide was fixed to the radial shaft with 1.25 mm K-wires to direct correct positioning of the screw holes. The osteotomy cutting guide was then positioned using the same K-wires and the osteotomy cut was made using an oscillating saw. Next, the ulna was approached in the interval between the flexor and extensor carpi ulnaris and the planned ulnar osteotomy was performed in a similar manner (Figure 3A-3F). Subsequently, the planned correction was performed by positioning the bone segments in a manner that the previously drilled screw holes align. Internal fixation was accomplished using a 3.5 mm 6-holes locking compression plate (DePuy Synthes Products, Inc., Warsaw, IN, USA). Lastly, the correction and the plate osteosynthesis of the radius were performed in a likewise manner. No patient-specific plates were used. No bone grafts were used. After completing the osteosynthesis, range of motion and distal radio-ulnar joint (DRUJ) stability were tested.

If there was unsatisfactory pro-supination after corrective osteotomy, a radio-ulnar osseous impingement was excluded and a further release of the interosseous membrane (IOM) could be performed. The IOM was routinely partially released at the level of the osteotomies: the extent of the partial release was the required release that allows the patient-specific drilling and cutting guides to fit around the radius and ulna. If there was persistent impairment in pronation or supination, respectively the dorsal or volar DRUJ capsule could be released. Kleinman et al. previously stated that a predictable loss of forearm supination will result from posttraumatic contracture of the oblique folds of the redundant volar capsule; pronation loss can result from similar pathology of the dorsal capsule [15].

Volar DRUJ release was performed by a ‘‘silhouette’’ resection of the volar DRUJ capsule to eliminate pathological thickened tissue that prevents normal forearm rotation [16].

This was performed using a volar approach: After identifying the position of the triangular fibrocartilage complex by fluoroscopy, the DRUJ was approached through an interval between the ulnar neurovascular bundle and the flexor carpi ulnaris tendon; the neurovascular bundle was retracted radially; the volar radio-ulnar ligament was identified and protected with great care; and then a volar "silhouette" resection of the DRUJ capsule was performed to completely excise the thickened elements of the capsule itself while protecting the articular surfaces of the distal ulna and distal radius sigmoid notch from injury, as described by Kleinman et al [15].

Figure 3A.
Real-sized model of
the pre-operative ulna
(for orientation)

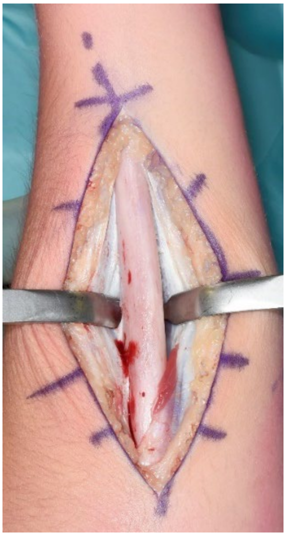


Figure 3B.
Surgical approach of the ulna

Figure 3C.
Positioning of the patient-
specific drilling guide
(for screw positioning).

Figure 3D.
Positioning of patient-specific
cutting guide
(for corrective osteotomy cut)

Figure 3E.
Corrective osteotomy of the
ulna.

Figure 3F.
Plate osteosynthesis of the ulna

Post-operative management

Post-operative management was patient-specific. If full pro-supination was achieved per-operatively, patients received a pressure bandage post-operatively. If there was a supination deficit post-operatively, patients received a cast in maximum supination for two weeks and vice versa for pronation deficits. Afterwards, treatment was functional (with restrictions for lifting until radiographic consolidation). Patients underwent physiotherapy and were referred to a Physical medicine and rehabilitation physician. If full pro-supination was not achieved, dynamic bracing in pro- or supination (depending on the deficit) was used (Figure 4).

Figure 4. Dynamic bracing in pro- or supination (depending on deficit).

Outcome measures

Our primary outcome was the gain in pro-supination at 12 months follow-up, measured with a universal goniometer, using the method as prescribed by the American Society of Hand Therapists [17]. We used a 180° protractor goniometer with two movable arms of 20 cm, constructed of clear, flexible plastic. One arm of the goniometer was lined up parallel to the upper arm of the patient, the other arm was placed parallel to the distal third of the forearm.

Our secondary outcomes were patient-reported outcome measures (PROMs): the QuickDASH questionnaire (11 items, range 0-100), patient-reported numerical rating scale (NRS) scores for pain and cosmetic appearance (range 0-10: higher score indicates more pain or poorer cosmetic appearance), maximal grip strength (best of three efforts) using a JAMAR-dynamometer (J.A. Preston Corporation, New York, NY, USA); and the occurrence of complications. Functional outcomes were measured by two non-blinded authors independently (E.E and J.C), two separate measurements (of pro-supination) were performed at each follow-up, and averages of both measurements were used. At final follow-up at 12 months after surgery, NRS score for treatment satisfaction was reported (range 1-5: higher score indicates more satisfaction). Outcomes were collected at baseline, 6 months and 12 months follow-up.

Radiographic analysis

One author (KR) measured angular deformity of the radius and ulna in reference to the contralateral forearm using radiographs in the same forearm position. By superposition of the outlines of the affected and contralateral forearm bones, we determined the location of maximal deformity and angular deformity in both planes [12]. The maximum deformity angle (MDA) was calculated from two measurements of angular deformity—one on the anteroposterior and 1 on the lateral radiograph—which represented the true angular deformity [18]. Hereby, deformity was reported as one calculated finding: the true angulation, which increases the comparability of fracture characteristics [12, 19]. Radiographic angulation was re-measured in all cases by a different author (EE) to assess reproducibility.

Statistics

Outcomes were tested for normality by using the Shapiro-Wilk normality test. We reported medians and interquartile range (IQR) for non-parametric variables and means and 95% confidence intervals (CI) for normally distributed variables. P-values <0.05 were considered statistically significant. Intra-class correlations were calculated to compare reliability of pro-supination and radiographic angulation measurements between observers. Differences between the pre-operative and the post-operative ranges of motion, patient-reported outcomes, grip strength and NRS pain and cosmetic appearance scores were determined using the related samples Wilcoxon signed-rank test, due to the small sample size of 15 patients.

ANOVA was performed to assess the relationship between the gain in pro-supination after corrective osteotomy and clinically relevant factors. Subgroups were created for: age at trauma (<10 years vs. ≥10 years); time from trauma until corrective osteotomy (<1 year vs. ≥1 year); severity of angular deformity (<20° vs ≥20°); severe vs. moderate pre-operatively impaired pro-supination (<69° vs. ≥69°). Severely impaired pro-supination was defined as an arc of less than 69° of pro-supination, which was based on the necessary arc of 103° (SD 34°), reported by Sardelli et al, (2011) and subtracting one standard deviation, equaling 69° [20]. Early corrective osteotomy was defined as corrective osteotomy performed within one year after trauma. Next, multivariate linear regression analysis was performed to assess the effect of each factor on a continuous scale, while correcting for baseline pro-supination, as we assumed that baseline pro-supination would definitely influence outcome.

RESULTS

15 participants with a malunited pediatric diaphyseal both-bone forearm fracture with symptomatic impairment in pro-supination were included between October 2016 and July 2018. All patients underwent 3D-planned corrective osteotomies of both radius and ulna. All surgeries were performed by two surgeons operating together (JC and FS). Patient demographics are presented in the supplementary Table S1.

Table 1. Association between factors and postoperative gain in pro-supination.

Factors	Number of patients	Gain in pro-supination (°)*	*p* -value
Age at trauma			
< 10 years	10	59 (48-70)	0.53
10 years or more	5	48 (26-108)	
Age at osteotomy			
< 13 years	6	71 (50-92)	0.17
13 years or more	9	55 (38-72)	
Time until osteotomy			
< 1 year	3	83 (13-152)	0.06
More than 1 year	12	56 (45-68)	
Angulation of Radius			
< 20°	8	50 (36-64)	0.02
20° or more	7	75 (57-93)	
Angulation of Ulna			
< 20°	13	63 (49-77)	0.57
20° or more	2	53 (15-91)	
Pre-op pro-supination			
< 69°	8	70 (50-89)	0.14
69° or more	7	53 (37-68)	

*Gain in pro-supination data presented as mean (95% confidence interval)

Mean age of these patients at trauma of 9.6 years (range 4-17.6) and a mean interval between trauma and corrective osteotomy of 5.9 years (range 0.4-12.4). There was a mean age at osteotomy of 15.5 years (range 10.2-23.3). There was a mean pre-operative true radial angulation of 20° (range 11-31) and true ulnar angulation of 15° (range 6-27).

The interrater reproducibility of the radiological assessment showed an intra-class correlation of 0.78 (CI 0.34-0.93) for the radius and 0.90 (CI 0.71-0.97) for the ulna. Per-operatively, additional soft-tissue releases were performed in four out of 15 patients: there was persistent impairment in supination in patient 3, 4 and 15, who underwent an additional release of the volar DRUJ capsule; there was persistent impairment in pronation in patient 8, who underwent an additional further release of the interosseous membrane. There were no additional releases of the dorsal DRUJ capsule. We did not encounter instability of the distal radioulnar joint after corrective osteotomy requiring additional procedures.

Directly post-operatively, a cast in maximum pro- or supination was required in 10 patients: patient 2 and 8 received a cast in maximum pronation; patient 3, 4, 5, 9, 12, 13, 14 and 15 received a cast in maximum supination. After two weeks the cast was removed and patients received a dynamic removable split in maximum pro- or supination. From two to six weeks post-operatively, the dynamic splint was worn as much as possible and only removed for daily exercises from the physiotherapist. After six weeks the dynamic splint was used as a night splint, up to 3-6 months post-operatively, based on the function. In patients who did not receive a cast in maximum pro- or supination post-operatively (1, 6, 7 10 and 11), full pro-supination was not maintained and they too were treated by dynamic splinting. Patient 1 and 6 received a dynamic splint in maximum pronation, while patient 7, 10 and 11 received a dynamic splint which was alternatively used in maximum pro- and supination.

Primary outcome

Pre-operatively, there was a mean pro-supination of 67° (CI 55°-78°) of the affected side. Contralaterally, there was a mean pro-supination of 153° (CI 148°-158°). Thus, the affected side had a mean pro-supination of 44% (CI 36%-51%) of the contralateral side pre-operatively. At 6 months follow-up there was a mean pro-supination of 118° (CI 105°-130°) resulting in a mean gain in pro-supination of 51° (CI 38°-64°), achieving 78% (CI 71%-85%) of the contralateral side. At 12 months follow-up there was a mean pro-supination of 128° (CI 118°-139°), resulting in a mean total gain of 62° (CI 50°-74°). Contralaterally, there was a mean pro-supination of 150° (CI 144°-155°) at 12 months follow-up, thus patients achieved 85% (CI 80%-91%) of contralateral range of motion. The data of individual patients are in supplementary Table S2.

Predictors for greater functional gain

Multivariate linear regression analysis revealed that predictors of superior gain in pro-supination at 12 months follow-up after 3D corrective osteotomy were: severe pre-operative impairment in pro-supination ($p=0.006$), shorter time until corrective osteotomy ($p=0.03$) and substantial angular deformity of the radius ($p=0.04$) (Table 1).

Secondary outcomes

3D corrective osteotomy provided a statistically significant and clinically relevant improvement of the quickDASH score from 32 (15-38) at baseline to 2 (0-11) at final follow-up (p=0.01) [21]. Differences in grip strength were not statistically significant (p=0.90). Excellent scores for patient satisfaction were reported: 10 out of 15 patients were very satisfied, 4 patients were satisfied; 1 patient was neutral and 0 patients were unsatisfied/very unsatisfied. Secondary outcomes are presented in Table 2.

Table 2. Secondary outcomes before surgery and at 6- and 12-month follow up.

		Post operative (months)	
Outcome measures	Preoperative	6	12
QuickDASH score	32 (15-38)	14 (11-15)	2 (0-11)
Grip strength (%)*	93 (84-103)	82 (66-98)	93 (88-98)
NRS pain score	3 (0.5-6.5)	0 (0-6)	0 (0-3)
NRS cosmetic score	5 (2-6.5)	5 (2-7)	5 (4-5)
NRS satisfaction score	-	-	5 (4-5)

Data presented as score (interquartile range) in all except for grip strength;
grip strength data presented as percentage of the contralateral side (range).
QuickDASH: Disabilities of Arm, Shoulder and Hand score
NRS score (range 1-5: higher score indicates better cosmetics or more satisfaction)

Complications

Ulnar plate removal was performed in one case. One patient had a delayed union. There was a transient neuropraxia of the superficial radial nerve in one patient.

DISCUSSION

This study investigated the clinical outcomes after 3D-planned corrective osteotomy with patient-specific surgical guides for pediatric malunited forearm fractures causing impaired pro-supination. The results of this prospective study suggest that 3D corrective osteotomy for pediatric malunited forearm fractures is a good treatment option to achieve a satisfactory restoration of forearm rotation. Our patients in this study had a mean improvement in pro-supination from 67° (44% of contralateral) pre-operatively, to 128° (85% of contralateral) at 12 months follow-up. Hereby, a greater gain in pro-supination was seen if there was a shorter interval between trauma and corrective osteotomy, substantial angular deformity of the radius and severely impaired pro-supination. This confirmed the findings of a recent Individual Patient Data meta-analysis, which stated that the use of 3D computer-assisted techniques, a shorter interval between trauma until corrective osteotomy and severe angular deformity were factors associated with a greater gain in pro-supination after corrective osteotomy for pediatric forearm malunions [5]. Furthermore, in our series 3D corrective osteotomy provided a high patient satisfaction, a decrease in pain score and a clinically relevant improvement in the quickDASH, without the occurrence of any serious complication.

Previously, 3D corrective osteotomies for pediatric malunited both-bone forearm fractures due to symptomatic impairment in pro-supination have been described in seven studies with in total 34 [8,9,17,21-24]. Four out of seven studies were included in an individual participant data (IPD) meta-analysis [5] with a mean gain in pro-supination of 84° (from 62° to 146°) in 16 patients who underwent 3D corrective osteotomy. Afterwards, Byrne et al. (2017) reported a mean gain in pro-supination of 61°, from 115° to 176°, in five patients [24]. Bauer et al. (2017) performed 3D corrective osteotomies for post-traumatic pediatric forearm deformities with impaired pro-supination in 10 patients, leading to a gain in pro-supination of 53°, from 85° to 138° [8]. In the study by Oka et al. (2019) a mean gain in pro-supination of 47° was seen, from 115° to 162°, in three patients (patients 9-11) after 3D corrective osteotomy [17].

In the meta-analysis, a greater gain in pro-supination was found if corrective osteotomy was performed within one year after trauma: a mean gain of 93° vs. 61° [5]. In our prospective study only three out of 15 participants underwent corrective osteotomy within one year after trauma and a greater gain in pro-supination was realized (83° vs. 56°). Based on our experience, few patients undergo corrective osteotomy within one year after trauma, as preferred treatment starts with conservative management, awaiting the effect of remodeling and/or physiotherapy. Therefore, an interval until osteotomy of up to two years could be considered as an early corrective osteotomy.

Furthermore, severe pre-operative limitation in pro-supination was a predictor for greater functional gain in this study. In accordance with this, additional subgroup analysis of the 71 patients in the meta-analysis (including 16 3D corrective osteotomies and 55 conventional corrective osteotomies) revealed that 35 patients underwent corrective osteotomy with a pre-operative pro-supination <69° and displayed a mean gain of 97°, while 36 patients had a pre-operative pro-supination ≥69° and displayed a mean gain of 57° [5].

Also, our current study showed that severe angular deformity of the radius was associated with greater gain in pro-supination after 3D corrective osteotomy. A clear relationship between forearm shaft malunion and significant impairment in pro-supination has already been established [25]. Previously, in two cadaveric studies, it was demonstrated that angular deformities of 10° resulted in minimum limitation of pro-supination, whereas 20° of angulation caused an important loss of pro-supination, especially in middle-third deformities [26, 27].

The value of the conservative treatment of a pediatric forearm malunion is unclear. Until the effect and role of conservative treatment is clear, we recommend considering corrective osteotomy if there is unsatisfactory improvement after conservative treatment. The additional costs for the 3D planning, patient-specific cutting and drilling guides and 3D printed real-sized bones of the radius and ulna is approximately 4.000 euro per case.

This study has several limitations and strengths. There was a relatively small number of patients and the absence of a control group, which would ideally include patients in which a conventional corrective osteotomy using two-dimensional radiographic planning without patient-specific 3D printed surgical guides was performed. Also, investigators were not blinded for the side of the surgery during functional evaluation (due to visible scar). Another limitation of the study is that rotational deformity was not assessed. Nevertheless, this is the largest series of corrective osteotomies for pediatric malunited forearm fractures up to date.

SUPPLEMENTARY TABLES

Table S1. Individual participant data (Demographics)

Patient number	Age at trauma	Time until osteotomy	Sex	Side	Hand Dominance	Re-fracture	Previous treatment	Angulation of Radius	Angulation of Ulna
1	7.1	12.8	F	R	R	Y	C	17.0	19.8
2	9.7	9.8	F	R	R	Y	C	23.9	12.0
3	7.6	6.0	M	L	R	Y	O	14.3	6.0
4	5.0	9.6	F	L	R	Y	C	10.6	26.4
5	8.2	3.4	M	L	L	N	C	22.8	15.6
6	9.7	0.9	F	L	R	N	C	26.2	27.1
7	17.6	0.4	M	L	R	N	C	20.4	19.4
8	14.0	8.6	M	L	L	Y	O	18.2	13.4
9	10.7	9.3	F	L	L	N	C	31.1	12.0
10	13.7	0.7	M	L	R	Y	C	30.9	18.6
11	4.0	6.2	M	R	R	Y	C	22.2	11.4
12	7.4	4.7	M	L	L	Y	O	15.6	6.3
13	12.1	11.2	F	R	R	Y	CO	13.5	9.5
14	9.0	3.2	F	R	L	N	O	13.4	7.2
15	14.8	1.8	M	R	R	N	O	14.9	17.7
Average	9.6	5.9	-	-	-	-	-	19.7	14.8

R/L: Right/Left; F/M: Female/Male; Y/N: Yes/No, C/O: Conservative/Operative; CO: Corrective Osteotomy. Age at trauma and time until osteotomy data presented as year. Angulation of radius and angulation of ulna in degrees

Table S2. Individual participant data (Functional outcomes)

Pro-supination											
	Pre-operative			Postoperative follow-up (months)							
				6			12				
Pt	Pro/sup	Contra-lateral	% of contra	Pro/sup	% of contra	Gain	Pro/sup	Contra-lateral	% of contra	Gain
1	43/18	65/78	42%	25/65	67%	30°	30/70	58/75	75%	40°
2	30/18	63/90	31%	20/80	67%	53°	38/75	58/83	80%	65°
3	64/3	70/74	47%	48/48	63%	28°	55/53	65/78	75%	41°
4	53/-4	66/80	34%	33/45	51%	29°	50/55	68/80	71%	56°
5	62/10	74/83	46%	60/65	79%	54°	65/75	70/85	90%	68°
6	23/63	63/90	56%	48/80	80%	43°	53/83	58/85	95%	50°
7	5/35	75/85	25%	63/80	89%	103°	55/85	65/85	93%	100°
8	25/53	75/78	51%	38/73	72%	33°	38/80	75/75	78%	40°
9	45/43	63/90	57%	58/90	97%	60°	58/88	60/88	98%	58°
10	43/25	72/100	39%	60/90	97%	83°	68/98	73/103	94%	98°
11	43/5	80/88	28%	39/60	76%	69°	73/98	78/83	103%	88°
12	55/15	60/83	49%	39/80	76%	48°	70/78	63/85	90%	78°
13	43/65	78/83	67%	50/63	91%	33°	65/85	80/83	91%	28°
14	65/20	65/83	58%	46/68	79%	38°	78/70	68/83	92%	48°
15	50/-15	60/85	24%	47/53	84%	95°	68/75	58/85	91%	70°
Avg	43/24	68/85	44%	45/69	78%	51°	57/78	66/84	85%	62°

Pro-supination: pronation/supination

Pronation/supination and gain data presented as degrees

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Chapter 8

Accuracy of 3D corrective osteotomy for pediatric malunited both-bone forearm fractures

Children. 2023; 10(1):21

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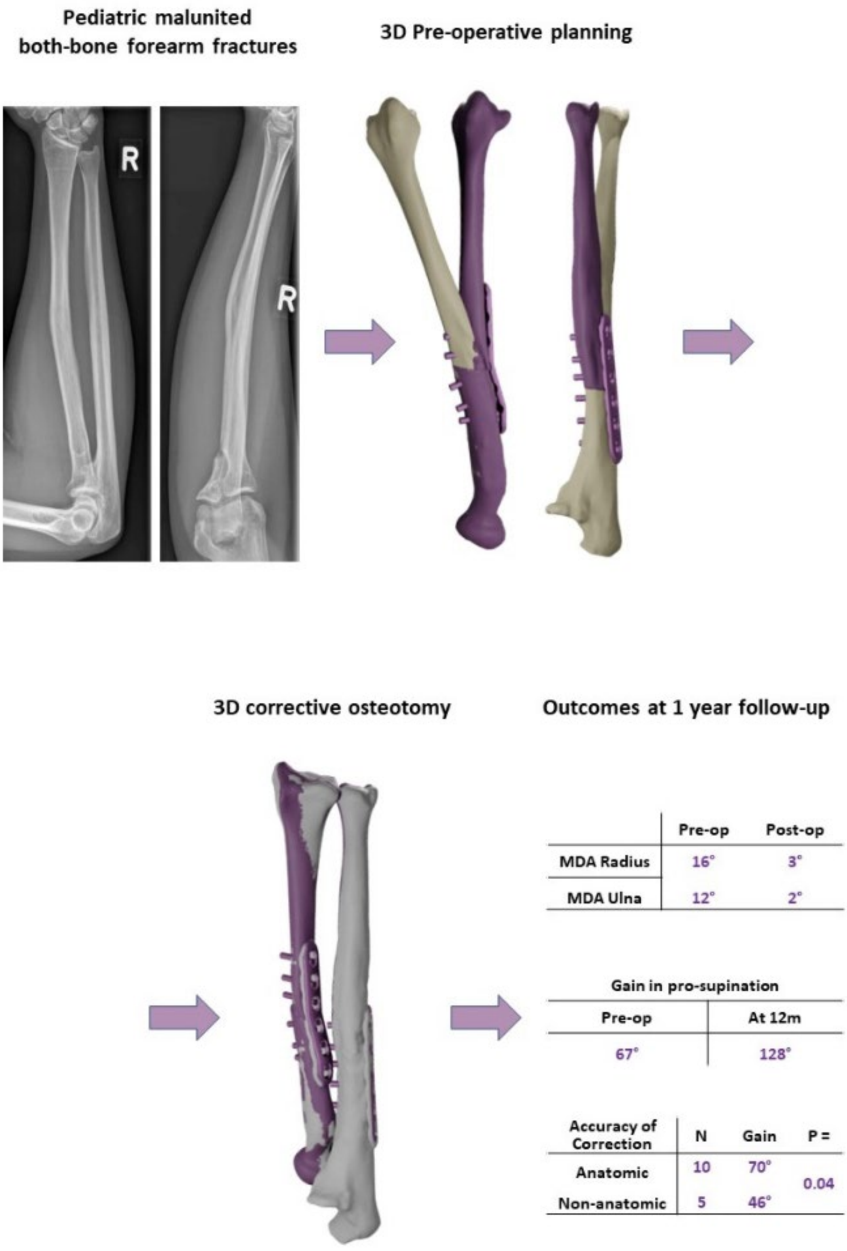
ABSTRACT

Re-displacement of a pediatric diaphyseal forearm fracture can lead to a malunion with symptomatic impairment in forearm rotation, which may require a corrective osteotomy. Corrective osteotomy with two-dimensional (2D) radiographic planning for malunited pediatric forearm fractures can be a complex procedure due to multiplanar deformities. Three-dimensional (3D) corrective osteotomy can aid the surgeon in planning and obtaining a more accurate correction and better forearm rotation. This prospective study aimed to assess the accuracy of correction after 3D corrective osteotomy for pediatric forearm malunion and if anatomic correction influences the functional outcome. Our primary outcome measures were the residual maximum deformity angle (MDA) and malrotation after 3D corrective osteotomy. Post-operative MDA > 5° or residual malrotation > 15° were defined as non-anatomic corrections. Our secondary outcome measure was the gain in pro-supination. Between 2016–2018, fifteen patients underwent 3D corrective osteotomies for pediatric malunited diaphyseal both-bone fractures. Three-dimensional corrective osteotomies provided anatomic correction in 10 out of 15 patients. Anatomic corrections resulted in a greater gain in pro-supination than non-anatomic corrections: 70° versus 46° ($p = 0.04$, ANOVA). Residual malrotation of the radius was associated with inferior gain in pro-supination ($p = 0.03$, multi-variate linear regression). Three-dimensional corrective osteotomy for pediatric forearm malunion reliably provided an accurate correction, which led to a close-to-normal forearm rotation. Non-anatomic correction, especially residual malrotation of the radius, leads to inferior functional outcomes.



	Pre-op	Post-op
MDA Radius	16°	3°
MDA Ulna	12°	2°

Gain in pro-supination	
Pre-op	At 12m
67°	128°

Accuracy of Correction	N	Gain	P =
Anatomic	10	70°	0.04
Non-anatomic	5	46°	

INTRODUCTION

In midshaft forearm fractures, growth will not remodel angular deformity as it does in distal fractures [1]. Impairment in forearm rotation is a critical problem associated with malunions of the forearm bones [2]. Malunited diaphyseal forearm fractures in children leading to a severe restriction in pro-supination may require corrective osteotomies [3]. A conventional corrective osteotomy can be technically demanding due to the multiplanar deformity of both forearm bones [4]. In a series by Miyake et al., one patient even had a rotational malunion of the radius of 136°, which is difficult to assess precisely using two-dimensional (2D) radiographic planning. Recent advancements in three-dimensional (3D) planning and 3D printing of patient-specific instruments (PSIs) can aid the surgeon in achieving a more accurate correction. Non-anatomic correction of the bony anatomy in malunions, especially of the upper extremity, may lead to inferior functional outcomes. Several authors have stated anatomically accurate correction during 3D corrective osteotomy is highly desirable to achieve a good outcome [5,6]. Few studies have tested this assumption nor have examined the effectiveness of 3D corrective osteotomy for pediatric malunited forearm fractures concerning the radiographic accuracy of the correction [3,7]. This prospective study aimed to assess the accuracy of correction after 3D corrective osteotomy for pediatric forearm malunion and if anatomic correction in-fluences the functional outcome.

MATERIAL AND METHODS

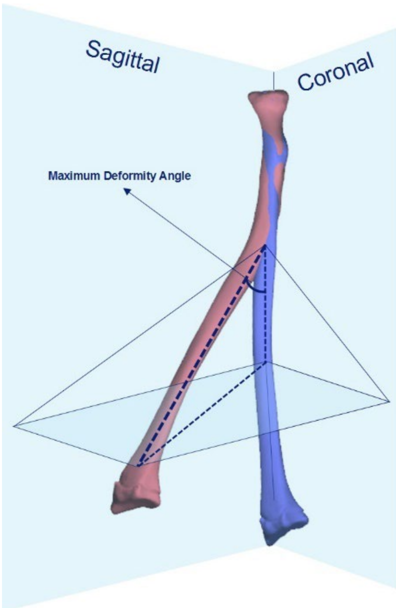
This study represents an additional analysis of the radiographic outcomes of a prospective cohort of patients whose clinical outcomes have been published previously [8]. Patients were eligible for enrollment if they met the following inclusion criteria: having a symptomatic forearm malunion after a diaphyseal both-bone forearm fracture sustained during childhood (<18 years), resulting in a limitation in pro-supination (pronation or supination of <50°), with unsatisfactory improvement after physiotherapy and a minimum age of 10 years at 3D corrective osteotomy. In addition, patients were excluded if they had an osseous deformity of the contralateral forearm. The pre-operative planning, surgical technique, and post-operative management of our 3D corrective osteotomies are described in our previous publication [8]. Planning of 3D corrective osteotomy and 3D printing of PSIs were performed at Materialise N.V., Leuven, Belgium in collaboration with our surgeons. An example of pre- and post-operative radiographs is provided in Figure 1.

Figure 1. Example of pre- and post-operative radiographs

Outcome Measures

Our primary outcome measure was the radiographic accuracy of the achieved correction after 3D corrective osteotomy. To assess the accuracy of correction, we compared the 3D pre-operative plan with the one-year post-operative computed tomography (CT). The residual maximum deformity angle (MDA) and malrotation after 3D corrective osteotomy were used to describe the accuracy of correction. The MDA is calculated by combining the angular deformity on both the coronal and sagittal plane derived from CT, as described by Nagy et al., illustrated in Figure 2 [3,9]. Similar to the study by Byrne et al., we assessed how often angular deformities could be corrected to within 5° of contralateral by 3D corrective osteotomy. Residual MDA ≥ 5° was defined as a non-anatomic correction. Unlike for the lower extremity, which most authors recommend to correct a torsional deformity of ≥15° [10]], there are still no uniform cut-off values as to when a correction is indicated in post-traumatic rotational deformity of the forearm [11]. In the current study, malrotation of the radius or ulna ≥ 15° was defined as a non-anatomic correction.

Figure 2. Maximum deformity angle



The maximal deformity angle (MDA) was calculated by combining the measurements of angular deformity in the coronal and sagittal plane according to the following formula:

$$MDA = \sqrt{tan^2(Coronal) + tan^2(Sagittal)}$$

Our secondary outcome measures were: functional gain in pro-supination and patient-reported outcome measures (PROMs): the QuickDASH questionnaire (11 items, range 0–100), numerical rating scale (NRS) scores for pain and appearance (range 0–10), and maximal grip strength using a JAMAR hand dynamometer (J.A. Preston Corporation, New York, NY, USA). Pro-supination was measured with a universal goniometer utilizing the method of the American Society of Hand Therapists [12]. Functional outcome was measured by two authors independently (E.E. and J.C.).

3D Radiographic Assessment

Radiographic evaluation of the accuracy of the performed correction was per-formed by analyzing the 3D models of the pre- and post-operative forearm bones according to the following steps: using Mimics software (Mimics Research 25.0), seg-mentation is performed using a threshold-connected region growing algorithm that collects voxels that belong to the affected bone. Then, the forearm bones are extracted as separate 3D objects.

Next, 3-Matic software (3-Matic Research 17.0) was used to compare 3D models of the pre-operative situation, planned correction, and post-operative result. First, analytic cylinders of the proximal and distal shafts of the radius and ulna are created to establish the axis of the proximal and distal parts of both bones in all three situations. Next, using a closest fit algorithm, the proximal ends of the radius and ulna of all three situations are aligned proximally. The axes of the proximal shaft proximal to the planned correction were used for the coordinate system, as this axis was alike in all three situations. Finally, the deviation between the distal segments in all three situations was measured to assess the degree of angular and rotational malalignment in the coronal, sagittal, and axial planes. The coordinate system of the radius was established as described by the International Society of Biomechanics (ISB) 2005 recommendations [13]. The maximum deformity angle (MDA) was calculated by combining the measurements of angular deformity in the coronal and sagittal planes, according to the Pythagorean theorem. MDA was calculated from the coronal and sagittal planes derived from CT instead of plain radiographs to increase the accuracy of the measurement be-cause the reliability of measurements from 2D images is hampered by over-projection [14]. Two authors measured radiographic outcomes independently (K.R. and E.E). Mean values of both assessors are presented.

2.3. Statistical Analysis

p-values < 0.05 were considered statistically significant. The intraclass correlation coefficient (ICC) was measured to assess the inter-observer reliability of the radio-graphic measurements. One-way analysis of variance (ANOVA) was performed to study the relationship between an anatomic correction and functional outcomes (gain in pro-supination and PROMs). Subsequently, multi-variate linear regression analysis was performed to investigate the relationship between the accuracy of correction (re-sidual MDA and malrotation of radius and ulna) and gain in pro-supination, both on a continuous scale.

Table 1. Radiographic outcomes: pre- and post-operative malalignment (°).

	Radius								Ulna							
	Pre-Operative				Final Follow-Up				Pre-Operative				Final Follow-Up			
Pt	Cor	Sag	MDA	Ax	Cor	Sag	MDA	RM	Cor	Sag	MDA	Ax	Cor	Sag	MDA	RM
1	3	14	14	−6	0	0	0	6	6	−5	7	32	−5	5	7	−12
2	9	22	23	24	0	0	1	−2	16	1	16	−1	1	0	1	15
3	−4	8	8	−11	−3	0	3	−9	8	−6	11	−4	1	0	1	−16
4	0	17	17	−26	11	7	13	−6	8	−20	21	11	1	−1	2	2
5	−2	22	23	−8	−1	0	1	14	9	−7	11	0	0	0	0	4
6	7	27	27	−31	0	−3	3	−3	8	−18	19	6	1	0	1	3
7	−11	0	11	−13	1	−1	1	−4	−7	19	20	3	0	1	1	−5
8	5	18	19	18	−1	−1	1	−8	11	2	11	−1	−1	0	1	3
9	0	16	16	1	−1	−1	2	10	6	−5	8	−7	0	−1	1	0
10	17	24	29	−4	0	−2	2	−5	1	−13	13	13	−1	−1	2	9
11	6	19	20	−12	3	3	5	0	3	−15	15	−4	0	0	0	0
12	1	11	11	49	−1	0	1	−1	5	−3	6	−3	−1	−4	4	5
13	9	4	10	15	−2	4	5	17	7	2	7	−20	0	2	2	0
14	−2	6	6	−5	1	1	1	−10	13	−1	13	−7	0	1	1	−1
15	−7	3	7	−17	2	0	2	−3	4	3	5	5	−2	−1	3	−6
Mean	8,1	14,0	16,1	15,9	1,8	1,7	2,6	6,6	7,4	8,0	12,2	7,7	1,0	1,2	1,7	5,5
SD	9,4	8,4	7,2	12,4	2,7	2,0	3,1	4,8	3,7	6,9	5,2	8,7	1,2	1,4	1,7	5,3

Cor = coronal; Sag = sagittal; Ax = axial. MDA = maximum deformity angle; RM = residual malrotation Dorsal angulation = positive; volar = negative; radial = positive; ulnar = negative; axial malrotation in pronation = positive; axial malrotation in supination = negative. Means are calculated based on absolute values

RESULTS

Between October 2016 and July 2018, 3D corrective osteotomies of both the radius and ulna were performed in fifteen patients due to pediatric malunited both-bone di-aphyseal forearm fractures. Patients had a mean age at trauma of 9.6 years, a mean time until 3D corrective osteotomy of 5.9 years, and a mean age at osteotomy of 15.5 years. There was a mean operating time of 138 min (SD 35) for the 3D corrective oste-otomies of the radius and ulna. In addition, four out of fifteen patients underwent an additional soft-tissue release. There were three minor complications: ulnar plate re-moval, delayed union, and transient neuropraxia of the superficial radial nerve. There were three minor complications: ulnar plate removal, delayed union, and transient neuropraxia of the superficial radial nerve.

Primary Outcomes

The pre- and post-operative malalignments of the radius and ulna are provided in Table 1. Anatomic correction was achieved in 10 out of 15 patients (25 out of 30 fore-arm bones) after 3D corrective osteotomy. Examples of an anatomic and a non-anatomic correction of the radius are supplied in Figures 3 and 4 (Case 1 and 4). Likewise, an example of residual malrotation of the radius is provided in Figure 5 (Case 13).

Figure 3. Example of an anatomic correction of the radius.

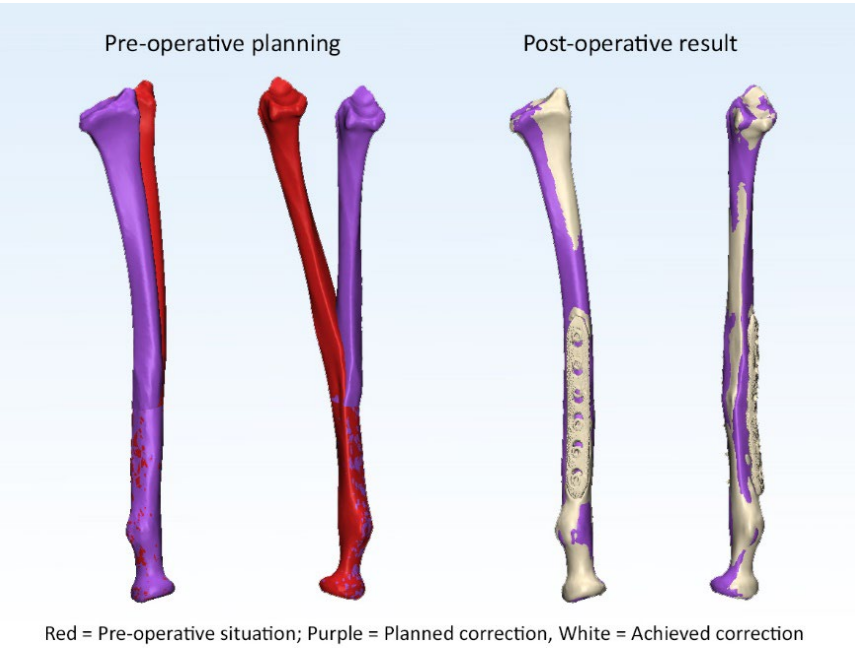


Figure 4. Example of a non-anatomic correction of the radius.

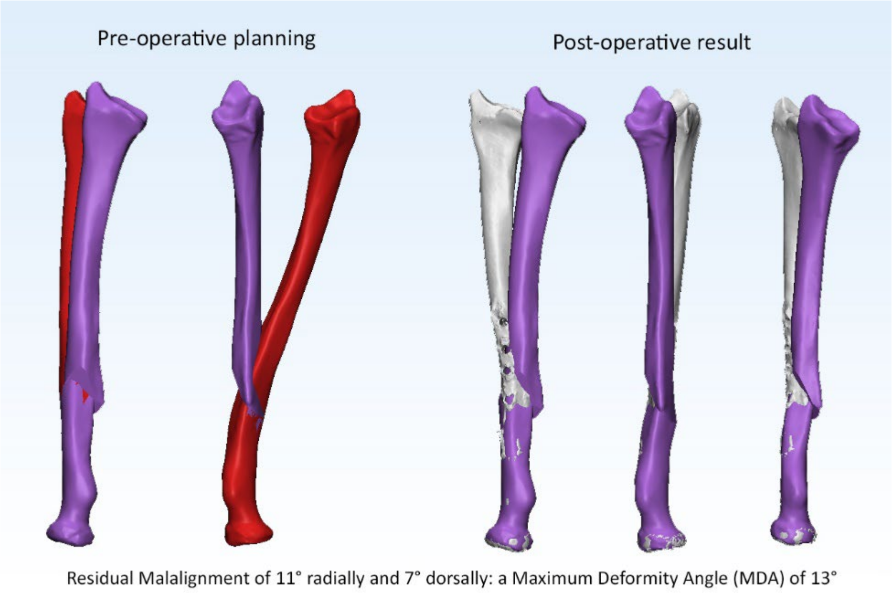
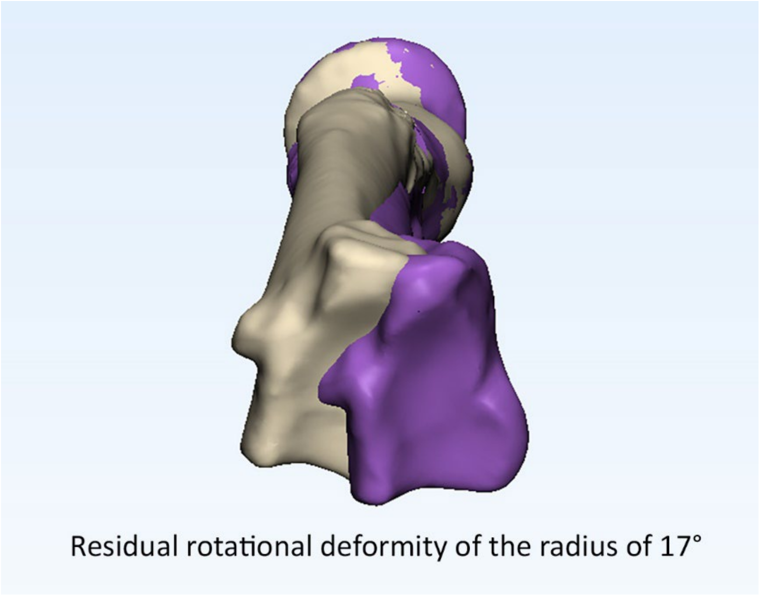


Figure 5. Example of residual malrotation of the radius.



Is Anatomic Correction Associated with Greater Functional Outcomes?

Three dimensional corrective osteotomy provided a mean gain in pro-supination from 67° (44% of contralateral) pre-operatively to 128° (85% of contralateral), thus a mean total gain of 62°. The results of ANOVA are presented in Table 2. ANOVA revealed ten patients who achieved anatomic correction after 3D corrective osteotomies had significantly greater gains in pro-supination than those with non-anatomic corrections: 70° (95% CI: 55–85°) versus 46° (95% CI: 28–64°). Patient-reported outcome measures or grip strength measurements between anatomic and non-anatomic corrections showed no significant differences. Multi-variate linear regression analysis revealed residual malrotation of the radius was associated with inferior pro-supination (p = 0.026); the model is provided in Table 3.

In our radiographic assessment, the interrater reproducibility showed intra-class correlations of 0.996 (95% CI: 0.991–0.998) and 0.992 (0.984–0.996) for measurement of the MDA of the radius and ulna; 0.990 (0.979–0.995) and 0.971 (0.938–0.986) for rotational assessment of the radius and ulna.

Table 2. ANOVA

	Anatomic Correction (n = 10)	Non-anatomic Correction (n = 5)	p =
Pre-op pro-supination	67° (53–80°)	66° (36–97°)	0.97
Pro-supination at FU	136° (125–148°)	112° (95–129°)	0.01
Gain in pro-supination	70° (55–85°)	46° (28–85°)	0.04
Pre-op QUICKDASH	22 (13–30)	31 (19–43)	0.16
QUICKDASH at FU	13 (10–16)	17 (14–20)	0.07
Δ QUICKDASH	8 (0–17)	14 (2–26)	0.38
NRS pain score	1.1 (−0.5–2.7)	3.0 (−0.4–6.4)	0.18
NRS cosmetics	2.3 (0.5–4.2)	4.6 (1.3–8.0)	0.13
Grip strength (%)	94 (88–98)	90 (78–102)	0.41

Confidence interval of 95%presented as: (95%CI); FU: follow-up; NRS: numeric rating scale.

Table 3. Multi-variate linear regression.

	Unstandardized Coefficients		
Model	B	Std. Error	Significance
(Constant)	79.0	8.4	<0.001
Residual malrotation Radius	−2.6	1.1	0.026

Dependent variable: gain in pro-supination.

DISCUSSION

This prospective study aimed to assess the accuracy of correction after 3D corrective osteotomy for pediatric forearm malunion and if anatomic correction influences the functional outcome. In this study, 3D-planned corrective osteotomies for pediatric malunited both-bone forearm fractures resulted in anatomic corrections in 10 out of 15 patients (25 out of 30 operated forearm bones). Patients with anatomic corrections had statistically significantly greater gains in pro-supination after 3D corrective osteotomies than non-anatomic corrections (70° versus 46°). Residual malrotation of the radius after 3D corrective osteotomy was associated with an inferior gain in forearm rotation.

Understanding the complex 3D deformities of both forearm bones in a malunited forearm fracture remains challenging. Therefore, a 3D corrective osteotomy is a promising technique. Recurrent patterns in forearm malunion are often seen. The supinator, pronator teres, and pronator quadratus muscles exert a pulling force upon fracture fragments, which can lead to angular deformity, malrotation, or narrowing of the in-terosseous space. In fractures located proximal to the pronator teres insertion, the proximal fragment supinates and flexes due to unopposed forces of the supinator and biceps brachii, whereas the distal fragment pronates due to the pronator quadratus and pronator teres. In contrast, in fractures located distal to the pronator teres insertion, the proximal fragment will not rotate as the supinator opposes the forces of the pronator teres and biceps brachii. The distal fragment will pronate and deviate towards the ulna due to the pronator quad-ratus [4]. Angular deformities of the radius and ulna lead to bony impingement or in-creased interosseous membrane (IOM) tension, which causes impairment in forearm rotation [15]. In a cadaveric study, a dorsal angular deformity of 20° caused a limitation in pronation. Correspondingly, a volar angular deformity of 20° led to supination limitation. Lastly, angular deformity narrowing the interosseous space limited both pro- and supination [16]]. In 2018, Abe et al. stated a pronation limitation was found if there was bony impingement due to dorsal angulation of the radius (>8°) because the interosseous space is encroached during pronation [17]. A supination limitation was found if there was a tightness of the transverse central band (CB) due to valgus deformity of the ulna (>6°), which increases the interosseous space during supination.

Unfortunately, there is no published literature with CT-based accuracy assessment of conventional 2D planned corrective osteotomies with which to compare.

In 2008, Murase and colleagues reported the accuracy of 3D corrective osteotomy for malunited forearm fractures in 10 patients. The mean angle of deformity improved from 16° pre-operatively to 1° after surgery. The mean pro-supination improved from 79° to 155° post-operatively.

In 2012, Miyake et al. published the outcomes of 3D corrective osteotomies for malunited forearm fractures in 20 patients. The average radiographic deformity im-proved from 21° pre-operatively to 1° post-operatively. In addition, their forearm motion improved from 76° pre-operatively to 152° post-operatively.

In 2013, Kataoka et al. published the results of 3D corrective osteotomies with PSIs for malunited forearm fractures in four patients. They used standard plates, which were pre-bent to fit around 3D-printed, real-sized plastic bone models of the radius and ulna. They achieved an accuracy of correction with a mean error in all directions of <2° for both the radius and the ulna. Mean errors were greater in growing children, as longitudinal forearm growth was not considered. They achieved a mean gain in pro-supination from 106° pre-operatively to 158° post-operatively [18].

In 2015, Bauer et al. performed 19 3D corrective osteotomies due to forearm deformity in children of which 15 were post-traumatic. In their study, maximum deformity angulation of the radius and ulna improved from 23° and 23° to 9° and 8°, respectively. Ten patients were operated on due to limited pro-supination, and a gain in pro-supination was seen from 85° to 138°.

In 2017, Byrne et al. published the outcomes of five patients who underwent 3D corrective osteotomies for malunited diaphyseal forearm fractures. Besides 3D-printed PSIs, they also used patient-specific plates. They found a mean error in the correction of 1.4° for the radius and 1.8° for the ulna. They aimed to correct angular deformities within 5° of the contralateral side and succeeded in 80% of cases. In addition, 3D corrective osteotomy improved mean pro-supination from 115° to 176°.

In 2019, Oka et al. performed 16 3D corrective osteotomies for malunited fractures of the upper extremity. They also used patient-matched plates. They achieved a correction to within 5° of contralateral in 15 of 16 patients after 3D corrective osteotomies. In their study, the mean difference between the planned correction and the achieved result was <1° in all three planes. In patients who were operated on due to limited pro-supination, a gain in pro-supination was seen from 115° to 162°.

In our series, the 3D osteotomy to correct a pediatric forearm malunion provided a highly accurate correction comparable to the studies mentioned above. Anatomic corrections were associated with greater gains in pro-supination. Thus, a lesser gain in forearm rotation was seen if a greater residual angular or rotational deformity persisted after 3D corrective osteotomy. Besides the highly accurate correction and excel-lent functional outcomes, another potential advantage of 3D modeling and 3D printing is to improve the patient–doctor relationship by giving them insights into the deformity’s complexity and the surgical procedure’s goal [19].

In our study, residual malrotation of the radius was associated with inferior pro-supination. Not restoring the natural radial bow may lead to bony impingement or too tight soft-tissue, which hinders the radius from swiveling around the ulna. In 1984, Tarr et al. claimed any torsional deformity of the radius leads to a loss of forearm rotation equal to the magnitude of the rotational malalignment but in the opposite direction [16]. However, in a cadaveric study by Kasten et al., a rotational malalignment of the radius of 30° in pronation resulted in a supination deficit of only 14°. Similarly, a rotational malalignment of 30° in supination resulted in a pronation deficit of only 11° [20]. Malrotation of the ulna is well tolerated since the ulna is a relatively straight bone. Thus, this leads to less restriction in forearm rotation than malrotation of the radius [11,21]. A study by Tynan et al. created malrotations of the ulna of 30°, which led to a decrease in forearm rotation of less than 20° [21].

In our study, there were a few cases with considerable residual malalignment or malrotation (Cases 1, 2, 3, 4, and 13). Although all patients were operated on by two experienced orthopedic hand surgeons operating together, four out of five non-anatomic corrections occurred in the first four operated patients. This suggests a considerable learning curve exists for 3D corrective osteotomy for diaphyseal both-bone forearm malunion. Therefore, a larger series is needed to detect if the surgical experience is a source of bias in the accuracy of a 3D corrective osteotomy. Oka et al. stated, "The simple surgical procedure is another advantage of the use of PMIs" [3]. However, we advocate there are still many possible challenges during surgery. For example, the absence of bony landmarks on the forearm bones and additional soft-tissue hindrance may impede the optimal guide position, which may result in un-der- or over-correction, as suggested by Jeuken et al. [22].

We did not expect residual malalignment or malrotation. The drilling guides dictate screw placement proximal and distal of the planned osteotomy. They are designed with the correct amount of rotational and angular correction built in so once the osteotomies are completed, the placement of screws should provide the desired correction [23].

Therefore, we investigated our outliers in more detail. There were no manufacturing issues. Three out of five non-anatomic corrections were malunions in the proximal diaphysis, suggesting a relation with a more complex surgical approach and more soft tissue hindering snug fit positioning of the surgical guides. Furthermore, the pre-operative plan for 3D corrective osteotomy does not consider the soft-tissue issues seen in post-traumatic forearm malunion. If there is a long interval between trauma and osteotomy in a growing child, soft-tissue contractures of the IOM, proximal and distal radioulnar joint capsule (DRUJ) can be seen [5].

Previously, persisting deficits in pro-supination after corrective osteotomy in longstanding forearm malunions have been seen, regardless of full geometric restoration of bony anatomy [2,24]. The IPD meta-analysis results supported soft tissue contracture's role in a longstanding malunion [25]. A long interval between trauma and corrective osteotomy compromised the functional gain in pro-supination, which was confirmed in our previous publication [8].

Limitations

This study has some limitations. First, there was no control group that underwent conventional corrective osteotomy using 2D radiographic planning without patient-specific 3D printed surgical guides. However, we find using only 2D radiographic planning for the correction of a 3D deformity unethical, as inferior results can unequivocally be expected. A previous meta-analysis showed the use of 3D computer-assisted techniques is a predictor of superior functional outcome after corrective osteotomy for a malunited pediatric forearm fracture [25].

Additionally, we included a relatively small number of patients. However, severe limitation in forearm rotation due to a pediatric malunited both-bone forearm fracture fortunately occurs seldomly. Therefore, a corrective osteotomy is rarely indicated.

Another limitation is if 3D corrective osteotomy did not provide full pro-supination, additional IOM or DRUJ release was performed during surgery. Thus, post-operative outcomes were not solely determined by correcting the bony anatomy. In the previous studies, no additional soft-tissue releases were performed [2,3,5,6,18,23]. Yet, this surgical plan does reflect our clinical approach to treating a post-traumatic forearm rotation: correct the bony deformity first, then solve the soft-tissue problems.

Furthermore, the post-operative CT scan was obtained one year after surgery. Thus, in children with remaining growing potential, additional remodeling could occur. Eight out of fifteen patients were aged <15 years at the time of 3D corrective osteotomy.

Lastly, there were only a few outliers to investigate due to the overall high accuracy of the correction and excellent functional outcome after 3D corrective osteotomy. Therefore, perhaps there are other unknown predictors for an inferior outcome we have yet to identify. Larger series are needed.

CONCLUSIONS

Three-dimensional corrective osteotomy using patient-specific instruments results in an accurate correction of pediatric malunited forearm fractures. A close to normal pro-supination was obtained in the majority of patients. Patients with an anatomic correction of the radius had better forearm rotation than non-anatomic corrections. Residual malrotation of the radius after a 3D corrective osteotomy is associated with an inferior outcome. Although PSIs simplify the operative procedure, a considerable learning curve still exists for 3D corrective osteotomy.

Desirable future research is a randomized controlled trial (RCT) comparing the outcomes after 3D-planned corrective osteotomy with or without PSIs because cost in-creases are substantially due to the 3D printing of PSIs. Future studies on 3D corrective osteotomy should provide patient-reported outcomes measures, functional outcomes, as well as radiographic outcomes on the accuracy of the achieved correction.

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PART IV:
General discussion and summary

Chapter 9

General Discussion

PART I: Distal metaphyseal forearm fractures in children

What to do in case of re-displacement?

Treatment of displaced distal metaphyseal forearm fractures in children by closed reduction is not always successful. Re-displacement occurs in 46% of displaced distal radius fractures in children [1]. In the case of re-displacement, the clinician is often confronted with a treatment dilemma: whether to perform a re-manipulation (with or without K-wire fixation) or to accept re-displacement and trust in the correction by growth.

One of the factors affecting the decision whether or not to re-manipulate is the degree of angulation. However, the clinician only assesses the angular deformity of a fracture in two dimensions (based on the posteroanterior (PA) and lateral radiographs), while the deformity is 3D. Therefore, we recommend using the method Nagy et al. suggested, combining the measurements from the PA and lateral radiographs to calculate the true angular deformity [2].

Not only the severity of angulation and remaining growth years but also surgeons', parents and child's preferences are taken into account in the decision-making process regarding whether to manipulate or not. Also, an early or late start to puberty can lead to considerable differences in skeletal maturity between a 12-year-old girl and a 12-year-old boy, but even so between two 12- year olds of the same gender. This matter should be considered during family decision-making to determine the best child-specific treatment.

Surprisingly, it does matter who treats the fracture because hand surgeons are 2.9 times more likely to recommend an operation on the same child with a distal radius fracture than pediatric orthopedic surgeons [3].

In our studies a secondary intervention was not performed often despite protocols suggesting re-manipulating all fractures that failed to maintain the reduction parameters. This is because the treating surgeon may expect a correction of the malunion by growth, may be reluctant to burden the child again, or may find it difficult to accept the failure of the initial treatment. We have also seen that there is a higher threshold for the clinician to perform a secondary intervention than after initial injury in the following studies: in a study by Jordan et al., there was a re-displacement rate of 27%, although a second surgical intervention was only performed in 4.7% [4]. In the study by Colaris et al., a re-manipulation was performed in 19 out of 35 re-displacements (54%), with additional K-wiring in 7 (20%) [5]. In a study by Mazzini et al., there was a loss of reduction in 57 out of 161 (35%), but a re-manipulation was performed in only 29 patients (51%), of which 22 had additional K-wiring (39%) [6]. In an RCT by McLaughlan et al., 14 out of 33 children with displaced distal radius fractures had re-displacement after closed reduction and casting. Still, only 7 out of 14 (50%) underwent a secondary procedure [7].

In *Chapter 2*, we observed similar outcomes after re-angulated pediatric distal forearm fractures in patients who did or did not undergo re-manipulation. Therefore, we recommend to "*Think twice before re-manipulating pediatric distal forearm fractures."*

What degree of angulation is tolerable?

In 2006 Ploegmakers et al. published a relevant manuscript titled "Acceptance of angulation in the non-operative treatment of pediatric forearm fractures." They stated that although angulated pediatric forearm fractures are famous for their tendency for spontaneous correction by remodeling, they are still considered unpredictable by many [8].

For distal metaphyseal forearm fractures, in 2005, Wilkins and O'Brien advised that dorsal angulations up to even 30°–35° will remodel satisfactorily in children who have at least five growing years left [9]. In 2008 Bae et al. recommended that 20° to 25° of dorso-volar angulation will remodel by growth in younger children [10]. In 2008 Hove et al. proposed the treatment guidelines that in children below 9 years, accept up to 20° of dorsal angulation, up to 10°–15° in children aged 9–13 years, and up to 5°–10° in children aged 13–15 [11]. In 2019, the Dutch "Children's Fractures" Guideline was published, which recommends performing a closed reduction in children aged 0-5 years if angulation exceeds 25°, in children aged 5-10 years if angulation exceeds 15°, and in children older than 10 years if angulation exceeds 10-15° [12]. Thus, recently treatment recommendations have become more strict.

In *Chapter 2*, we witnessed the excellent capacity of children to correct angular deformities during their growing years and deemed that current treatment guidelines are too strict. Tremendous remodeling is especially seen in children younger than 10 years old with distal forearm fractures close to the growth plate [9].

Crawford et al. accepted overriding distal radius fractures (with 100% dorsal translation) in 51 children aged <10 years and saw excellent functional and radiographic outcomes in 100% of cases [13]. In addition, the children's parents were very satisfied and indicated they would select the same treatment if given a chance to choose again.

Nonetheless, the parents of a young child can be very skeptical when a surgeon first proposes to accept a visible deformity of the forearm [14]. Family decision-making is critical, as parents feel responsible for the outcome of their child's injury. They worry about the consequences of making the wrong decision. Some parents initially prefer surgery to guarantee their child's arm would heal and be straight. For instance, Phelps et al. surveyed parents caring for a child with a displaced distal radius fracture. The mother of an 8-year-old child stated: "*Having to potentially bring him back in at some stage to re-break and reset the bones or for him to have to go potentially for the rest of his life with a big bend in his forearm. I didn't feel I could make that decision for him because I don't know what impact that could have on him for the rest of his life*." [15].

For other parents, it makes sense that a child's bone would be able to heal itself. Therefore, it is essential to take the time to explain the remodeling of children's fractures and what kind of patient journey can be expected following either a conservative or an operative treatment strategy. More and more research is being performed to explore the parent's and child' experience of a fracture and their thoughts about the uncertainty regarding the optimal treatment [15, 16].

Parents will also ask, *"How long will it persist?"* if there is a clinical deformity after cast removal [17]. Jeroense et al. attempted to answer this question. They studied 33 malunions of distal radius fractures in children aged under 14 years with angulations over 15°. They found a mean remodeling speed of 2.5° per month [18]. Likewise, in 2020 Lynch et al. studied the remodeling potential of pediatric distal radius fractures in the coronal plane and found a mean remodeling speed of 2.3° per month [19]. They observed an impressive improvement in coronal angulation from 17° to 3° in 6 months in 36 patients. They even stated that: re-manipulation is not indicated in children under 12 years of age where the maximum coronal angulation is <24 degrees [19]. Thus far greater potential for remodeling can also be seen in coronal angulations than previously thought [19].

Zimmerman et al. studied the long-term outcomes of pediatric distal radius fractures. They found that in children <10 years, large displacement at the time of fracture consolidation did not influence the 10-year functional outcome [20]. Zimmerman et al. also showed that the remodeling of volar angulation occurs just as well as in dorsal angulation [21]. Per contra, malrotations will not remodel [10].

Finally, a recent publication in the JAMA by Orland et al. stated that distal radius fractures with 100% displacement and less than 20° angulation may still be treated without closed reduction in children younger than 10 years [14]. In a cross-sectional study of 258 children, 55% of all children aged <10 years who presented with a distal radius fracture underwent closed reduction with procedural sedation, of which 27% were considered potentially unnecessary. Children who underwent closed reduction spent significantly more time in the emergency department than those who did not: 4.2 hours versus 2.2 hours. Cost analysis revealed that closed reduction and manipulation using procedural sedation in the emergency department was eight times more expensive than casting alone [14]. Sedation of a child for a closed reduction of a forearm fracture is not without risk. Approximately 14% of these children experience side effects such as over-sedation, hallucinations, or vomiting [22].

Thus, healing of a distal forearm fracture with some degree of displacement can thus be safely accepted in the expectance that remodeling will occur [20].

Based on our results and the literature mentioned above, we would like to suggest the following recommendations for the acceptance of angulation in pediatric distal forearm fractures:

For distal forearm fractures in boys:

- <9 years, accept up to 30° angulation.
- 9-11 years, accept up to 20° angulation.
- 11-13 years, accept up to 15° angulation.
- 13-15 years, accept up to 10° angulation.
- ≥15 years, accept up to 5-10° angulation.

For distal forearm fractures in girls:

- <8 years, accept up to 30° angulation.
- 8-10 years, accept up to 20° angulation.
- 10-12 years, accept up to 15° angulation.
- 12-14 years, accept up to 10° angulation.
- ≥14 years, accept up to 5-10° angulation.

Take into account that maximum angular deformity may occur in a plane other than the PA or lateral and can therefore be underestimated. To illustrate, a dorsal angulation of 20° with a radial angulation of 15° leads to a true angular deformity of 25°.

To K-wire or not to K-wire?

In 2013 Colaris et al. published an RCT which randomized children with a displaced distal metaphyseal both-bone forearm fracture to closed reduction with or without K-wire fixation [23]. This study recommended performing K-wire fixation since children treated without K-wire fixation had more re-displacements (45% vs. 8%) and more limitation in forearm rotation (14° vs. 7°) at short-term follow-up.

Afterward, in 2018 a meta-analysis was published on the same subject [1]. This meta-analysis included six studies with 382 participants [7, 23-27]. 76% of included children had a both-bone forearm fracture. K-wire fixation resulted in significantly better maintenance of the alignment of the fracture. There was a re-displacement rate of 46% in patients who did not receive K-wires versus 4% in patients with K-wire fixation. Functionally, a greater limitation of pro-supination was seen if treated without additional K-wires in one study [23]. In contrast, in two studies, there were no differences in functional outcomes between those treated with or without K-wires [26, 27]. Sengab et al. concluded that K-wires are suitable for preventing re-displacement but do not result in a better functional outcome than cast immobilization alone and are associated with more complications (15.7 versus 3.6%). They stated that more research is desired to identify those patients who will benefit most from K-wire fixation.

Earlier, Zamzam et al. tried to answer the question: *Who will benefit most from K-wire fixation?* They analyzed 183 children with a displaced distal radius fracture, where re-displacement occurred in 25% [28]. Fractures with complete displacement had an odds ratio of 25 of a re-displacement occurring. A perfect reduction did not prevent the re-displacement of fractures with complete displacement. They explained that the lack of a periosteal hinge might affect stability. Therefore, they recommended that pediatric fractures of the distal radius with complete displacement should be reduced under anesthesia and fixed by K-wires even when a satisfactory closed reduction has been achieved. Furthermore, both-bone forearm fractures were 23 times more likely to re-displace than isolated distal radius fractures in their series.

A recent series by van Delft et al. studied 200 consecutive patients with displaced distal metaphyseal forearm fractures. They stated that most metaphyseal forearm fractures could be treated with a very high success rate by closed reduction and casting alone in the emergency room [29]. However, complete initial displacement was again highly predictive of unsuccessful reduction. Thus, they recommended performing closed reduction with additional K-wire fixation in the operating room in children with unsuccessful reduction in the emergency room or complete initial displacement.

Thus, the goal of operative treatment is to prevent re-displacement. But is re-displacement a problem in a young child? Recently, there has been an increase in the operative management of fractures in children, although no long-term outcome studies show superior results after operative treatment [30].

Therefore, we studied the long-term outcomes of children with a displaced metaphyseal both-bone forearm fracture, who were randomized to closed reduction with or without K-wire fixation in ***Chapter 3***. Regarding the need for a reduction, the following treatment algorithm was used: a closed reduction was performed in pediatric both-bone forearm fractures with >15° of angulation in children aged <10 years or >10° of angulation in children aged 10-16 years. The criteria for re-manipulation were the same as those used for decision-making at the initial presentation. Although the RCT protocol stated that re-manipulation should be performed for re-displacement, 13 of 30 re-displacements were left untreated. At long-term follow-up, overall excellent long-term outcomes were seen in all patients, irrespective of the use of additional K-wire fixation or the occurrence of a re-displacement. This indicates that the criteria for the reduction of distal metaphyseal fractures were too strict. Therefore, more angulation can be accepted, especially in young children.

Regarding the key question, *To K-wire or not to K-wire?,* we concluded that most children with displaced metaphyseal both-bone forearm fractures can be treated by a closed reduction in the emergency room without additional K-wire fixation under procedural sedation or local anesthesia. However, if closed reduction is performed in the operating room, for instance, due to complete initial displacement, we recommend performing additional K-wire fixation. If closed reduction without K-wires is performed in the emergency room, the clinician should inform parents and the patient about the risk of fracture re-displacement. This can result in a malunion with functional impairment. Weekly radiographic monitoring is recommended to detect re-displacement. If re-displacement occurs and insufficient remaining remodeling potential is expected, a re-manipulation with additional K-wire fixation should be considered.

Future research

In future research, we do not recommend using an above-elbow cast to treat distal metaphyseal forearm fractures. Below-elbow cast is sufficient for the treatment of distal forearm fractures in children [31, 32].

A model which can predict the degree of remodeling based on clinical factors (age, gender), and radiographic factors (location of fracture, degree and direction of angular deformity, translation, rotation) would be ideal for aiding in clinical decision-making when treating a child with a displaced forearm fracture in the emergency department.

The desirable trials we have all been waiting for are already being performed. The Angulated Fractures in Children (AFIC) trial by Adrian et al. [33] randomizes children younger than 11 years of age with displaced distal forearm fractures with up to 30° angulation between cast immobilization without any reduction versus closed reduction with additional K-wire fixation. The Children's Radius Acute Fracture Fixation (CRAFFT) trial includes children aged 4-10 years with severely displaced wrist fractures and randomizes between surgical reduction and non-surgical casting. Likewise, Garcia-Rueda et al. are performing a similar trial [34].

Lastly, if K-wire fixation is performed, we wonder if fixation by 1 K-wire leads to similar outcomes as fixation by 2 K-wires.

PART II: Diaphyseal forearm fractures in children

Because midshaft fractures, especially in the adolescent population, do not have the same potential for remodeling as distal metaphyseal fractures, the criteria for reduction are more strict than for distal fractures. For diaphyseal forearm fractures, the Dutch national guidelines recommend accepting up to 15° for children younger than 10 years and up to 10° for children 10 years or older. Bowman et al. accepted shaft angles up to 20° in the distal third, 15° of angulation in the middle third and 10° in the proximal third in girls under 9 and boys under 11. For girls older than 8 years and boys older than 10 years, they accepted 10° of angulation at all shaft levels [35]. Jones et al. performed reduction for any patient with a midshaft forearm fracture aged ≤8 years with >10° of angulation. In children aged 9–17, a reduction was performed for any fracture with >8 ° of angulation [36].

Early conversion to a below-elbow cast for diaphyseal both-bone forearm fractures

Previously, three RCTs showed that a below-the-elbow cast (BEC) performs as well as an above-the-elbow cast (AEC) in maintaining the reduction of fractures in the distal third of the forearm in children and interferes less with daily activities [31, 32, 37].

In *Chapter 4,* we concluded that for diaphyseal both-bone forearm fractures, early conversion to BEC after three weeks does not lead to inferior functional outcomes compared to six weeks in an AEC. Thus, early conversion is safe for stable midshaft both-bone forearm fractures, also at long-term follow-up. Likewise, for diaphyseal forearm fractures in children, it is time to change practice and avoid the discomfort and morbidity of unnecessary elbow immobilization for six weeks.

Predictors for a limitation of pro-supination after pediatric both-bone forearm fractures

In *Chapter 5*, we aimed to answer the following questions:

1) *Which factors are associated with a limitation of pro-supination after pediatric both-bone forearm fractures at long-term follow-up?*
2) *Do accepted re-displacements of pediatric both-bone forearm fractures lead to inferior outcomes at long-term follow-up?*

In our prospective cohort study, factors associated with a limitation in pro-supination after a pediatric both-bone forearm fracture at minimum 4-year follow-up were: a complete fracture of the ulna, an older age at trauma, and a diaphyseal fracture location.

In 1962 Gandhi et al. stated that angular deformity of the mid-shaft of the forearm bones, thus diaphyseal fracture location, corrects relatively poorly and results in limitation of pro-supination [17]. In addition, Kay et al. stated that midshaft both-bone forearm fractures in children >10 years old result in residual functional deficit more often than is commonly appreciated. Therefore, >10° of malalignment in children >10 years old should not be accepted since it will result in significant loss of forearm rotation [38].

Franklin and Sinikumpu et al. found that conversion to operative treatment for both-bone forearm shaft fractures occurred more often in children aged > 10 years [39, 40]. It is well-known that *"The younger the child and the nearer the fracture is to the metaphysis, the greater are the potentialities for spontaneous correction"* [8, 17, 41].

In line with these studies, Zionts et al. prospectively found that malalignment in a child after a displaced diaphyseal both-bone forearm fracture causes loss of forearm rotation [42].

Do accepted re-displacements lead to inferior outcomes?

Many studies have been performed to identify predictors for re-displacement. For distal radius fractures, Zamzam et al. stated that predictors for re-displacement were: the presence of an associated fracture of the distal ulna and complete initial displacement of the radius [28]. For diaphyseal forearm fractures, Yang et al. stated that complete fractures were 10 times more likely to re-displace than greenstick fractures [43]. They suggested that greater care should be given in the treatment of complete fractures, especially those with malreduction, to avoid malalignment.

In *Chapter 5*, in distal metaphyseal forearm fractures, re-displacement occurred in 45% of the initially displaced fractures, which underwent closed reduction. In diaphyseal fractures, re-displacement occurred in 35% of displaced fractures which underwent closed reduction without additional stabilization. Re-displacement was accepted in 62% of distal metaphyseal re-displacements and 80% of diaphyseal forearm fractures.

Functionally, patients with an accepted re-displacement (diaphyseal and distal metaphyseal re-displacements) had less pro-supination than patients with a good alignment at 7-months follow-up. Yet, an accepted re-displacement did not lead to inferior pro-supination at 7-year follow-up.

Radiographically, patients with accepted re-displacements in the distal metaphyseal forearm had similar radiographic angulations compared to those with good alignment at consolidation at 7-year follow-up. This illustrates the exceptional potential for remodeling the distal metaphyseal forearm. However, less remodeling was seen of diaphyseal re-displacements at 7-year follow-up.

For diaphyseal forearm fracture, the Dutch national guidelines recommend accepting up to 15° for children younger than 10 years and up to 10° for children 10 years or older.

Based on the lesser potential for remodeling of diaphyseal forearm fractures we observed in our study and the literature [8, 35, 36, 44], we suggest the following recommendations for the acceptance of angulation in pediatric diaphyseal forearm fractures:

For diaphyseal forearm fractures in all children:

- <9 years, accept up to 10° angulation.
- ≥ 9 years, accept up to 8° angulation.

Future research:

Ideal future research would identify which children with diaphyseal both-bone forearm fractures are unstable and require elastic intramedullary nailing and which are stable after reduction and can be treated in a cast. This is because the per-operative test we used, pro-supinating the forearm after reduction [45, 46], does not seem reliable, as there is still a high re-displacement rate of fractures deemed stable during the first few weeks after reduction.

PART III: Malunited forearm fractures in children

When is a corrective osteotomy indicated?

Trousdale et al. stated that there is disagreement on how much of deformity will lead to functional loss in a skeletally immature patient [47]. Prommersberger et al. advised that in the case of functional disability, there is an indication for corrective osteotomy over the age of 12 in a malunion located in the distal third and over the age of 5 in gross deformity of the midshaft of the forearm [48]. Price et al. suggested performing corrective osteotomy in forearm shaft malunions as soon as possible for angulations greater than 30° and waiting at least six months for malunions with 20–30° [49]. Van Geenen et al. recommended that a corrective osteotomy was indicated in forearm malunions when pro-supination was less than 50–60% of the contralateral side [50].

Predictors for greater gain in forearm rotation after corrective osteotomy?

In *Chapter 6,* we provided the results from our IPD meta-analysis, in which we concluded that predictors of a superior functional outcome after corrective osteotomy due to pediatric malunited forearm fracture were: a shorter interval between trauma and corrective osteotomy, more severe angular deformity and the use of 3D computer-assisted techniques.

In 1995 Trousdale et al. published an impressive series of 27 corrective osteotomies for malunited pediatric forearm fractures performed at the Mayo Clinic between 1976 and 1991 [47]. They stated that a shorter time between trauma and corrective osteotomy provided a superior functional gain. In their study, the patients who had been managed early (corrective osteotomy <1 year after trauma) regained more than twice the amount of rotation compared to those who had been managed late: a gain of 79° if managed early versus a gain of 30° if managed late. In the patients who were managed late, soft-tissue scarring might have developed in the interosseous membrane, distal, or proximal radio-ulnar joint. Although the malunion was corrected anatomically, soft-tissue constraints partly compromised the result.

In 2006 van Geenen et al. published a series of 20 corrective osteotomies for malunited forearm fractures sustained during childhood and confirmed this previous finding: if corrective osteotomy was performed <1 year after trauma, a significantly greater gain in forearm rotation was seen than those operated ≥1 year: a gain of 98° vs. 76° [50]. Also, they stated that a younger age at osteotomy might be associated with a greater functional outcome. However, the latter was not a significant factor in our IPD meta-analysis.

Another predictor for a superior gain in function after corrective osteotomy was an angular deformity greater than 20°, which was in line with previous research. Two cadaveric studies demonstrated that angular deformities of 10° resulted in minimum limitation of pro-supination, whereas 20° of angulation caused an important loss of pro-supination, especially in middle-third deformities [51, 52].

Lastly, using 3D computer-assisted techniques during corrective osteotomy was a predictor for a superior outcome. A series of publications on 3D corrective osteotomies for malunited upper extremity fractures has emerged from the Osaka group [53-56]. Restricted forearm rotation is the key problem associated with malunions of the forearm bones. Correct coronal, sagittal, and axial alignment of both bones and restoration of normal length are necessary to obtain a good range of forearm rotation. In corrective surgery, the challenge is to reduce two linked rotating long bones while maintaining the congruity of the adjacent joints. The proposed advantages of a 3D osteotomy are that by calculating the degree and direction of 3D deformity, the osteotomy template can navigate the surgical procedure to realize the pre-operative simulation [54]. Another benefit is that although simple angular deformity can be assessed using radiography, rotational malalignment is difficult to detect on 2D radiographs [53].

What gain in forearm rotation can 3D osteotomy provide?

In *Chapter 7,* we aimed to determine what gain in forearm rotation can be achieved after a 3D osteotomy for a malunited pediatric diaphyseal both-bone forearm fracture and to assess which factors are associated with superior outcomes after a 3D osteotomy.

In the literature, nine studies provided individual participant data on thirty-two patients who underwent 3D corrective osteotomy due to a pediatric malunited diaphyseal forearm fracture with rotational impairment. A gain in pro-supination from an average of 72° pre-operatively to 149° post-operatively was seen, leading to a gain of 78° in pro-supination [53-61]. In our prospective study, 3D-planned corrective osteotomy resulted in a mean improvement in pro-supination from 67° (44% of contralateral) pre-operatively to 128° (85% of contralateral) at one-year follow-up, deeming our functional outcomes comparable.

As a discussion point, the mean time between trauma and 3D corrective osteotomy in our series of 15 patients was 5.9 years. In our series, 4 out of 15 patients achieved unsatisfactory intra-operative pro-supination after the osseous correction by 3D corrective osteotomy, and an additional soft-tissue release was performed. This confirms Trousdale et al.'s statement that contractures of the soft tissues may have developed in patients with a long interval between trauma and osteotomy. Therefore, we recommend the following clinical approach to treat a post-traumatic forearm rotation: correct the bony deformity first, then solve the soft-tissue problems. In previous studies in the literature, no additional soft-tissue releases were performed [53-56, 58, 61].

A greater functional gain could have been achieved after 3D corrective osteotomy if there is a lesser delay between trauma and corrective osteotomy or if an additional soft-tissue release is performed in case of a longstanding malunion.

Based on our experience, few patients undergo corrective osteotomy within one year after trauma because the preferred treatment starts with conservative management and awaits the effect of remodeling and physiotherapy. Many patients are referred late. Therefore, an interval until osteotomy of up to 2 years may be considered an early corrective osteotomy.

What accuracy of correction can 3D osteotomy provide?

A new, relatively expensive technology such as 3D corrective osteotomy should yield a shorter operation time, a more accurate correction, and superior functional outcomes compared to conventional 2D planned corrective osteotomy.

In a study by Bauer et al., 3D computer-assisted corrective osteotomy for malunions of the radius has a significantly shorter operating time than conventional corrective osteotomy: 108 minutes vs. 140 minutes [62].

Byrne et al. performed 3D corrective osteotomies for diaphyseal forearm malunions, aiming to correct angular deformities within 5° of the contralateral side, and succeeded in four out of five cases [59]. Oka et al. achieved a correction to within 5° of contralateral in 15 out of 16 patients after 3D corrective osteotomies for upper extremity malunions, though not all cases were diaphyseal forearm malunions [56].

In *Chapter 8,* 3D-planned corrective osteotomy for symptomatic malunited pediatric both-bone forearm fractures resulted in a correction of angular deformities within 5° of the contralateral side in 10 out of 15 patients (25 out of 30 forearm bones). Functionally, patients with anatomic corrections had a statistically significant greater gain in pro-supination than those with non-anatomic corrections (70° versus 46°).

Unfortunately, there is no published literature with CT-based accuracy assessment for conventional 2D planned corrective osteotomies to compare with [60]. However, we would not advocate the initiation of a trial comparing 3D versus 2D planned corrective osteotomy as we find using only 2D radiographic planning for the correction of a 3D deformity unethical, as inferior results can be expected.

The relationship between 3D osseous deformity and rotational impairment

Based on the available literature and our results, we have attempted to summarize our concepts of the relationship between 3D osseous deformity and rotational impairment of the forearm.

A malunion in the proximal forearm often results in greater loss of function than a comparable malunion in the distal region because proximally, the radius and ulna are more vulnerable to impingement due to the narrower interosseous space proximally [63]. Restricted pronation is most often found if there is bony impingement due to extension deformity of the radius (>8°), which narrows the distance between the radius and ulna during pronation [1]. Limited supination is often found if there is tightness of the transverse central band (CB) due to valgus deformity of the ulna (>6°), which leads to an increased distance between the radius and ulna during supination [63].

Axial malalignment should be suspected in a patient with a distal diaphyseal radius malunion, who presents with severe restriction of forearm supination, despite a mild-to-moderate angulation deformity on plain X-rays, which alone does not sufficiently explain the supination limitation [61]. A radiologic sign described by Naimark et al. may uncover rotational fracture deformity: in the absence of comminution, whenever the diameter of a long bone changes abruptly across a fracture line, a significant rotational deformity must be considered [64]. Kataoka et al. suggested that axial malalignment in pronation may have occurred due to contraction of the pronator quadratus muscle, as all deformities were located in the distal part of the malunited radius.

In our study, non-anatomic correction of the forearm bones after 3D corrective osteotomy for pediatric forearm malunion led to inferior functional outcomes. We expect that angular and rotational malalignment of the radius maily influences pro-supination, as the radius has a natural bowing, while the ulna is relatively straight. This can induce soft tissue problems in the interosseus membrane and an incorrect reconstruction of the radial bow can cause bone impingement issues when the radius swivels around the ulna. On the other hand, a rotational deformity of the ulna is well tolerated since the ulna is a relatively straight bone.

Regarding malrotation, previously, it was thought that any torsional deformity of the radius results in a loss of motion equal to the magnitude of the torsion deformity but in the opposite direction [51]. However, in a cadaveric study by Karsten et al., a torsional deformity of the radial shaft of 30° in pronation resulted in a supination deficit of only 14° and a torsional deformity of the radial shaft of 30° in supination, resulted in a pronation deficit of only 11°, which disclaims that magnitude of deformity equals the magnitude of the loss of motion [65]. Furthermore, another cadaveric study showed that malrotation of the ulna resulted in less forearm rotation restriction than malrotation of the radius [66].

Future research:

Desirable future research is a randomized controlled trial comparing outcomes after 3D-planned corrective osteotomy with or without PSIs. Conventional 2D planning is unethical due to the complexity of multiplanar deformity in both bone forearm malunions, and cost increases are mainly due to the use of PSIs.

We are also very interested in kinematic models which can predict functional deficit in forearm rotation based on osseous malunion and which functional outcome can be expected if 3D corrective osteotomy is performed. Additionally, it would be interesting to predict the added value of a soft-tissue release (interosseous membrane or capsule of the DRUJ).

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Chapter 10

Summary

PART I: Distal metaphyseal forearm fractures in children

In 2006 Ploegmakers and Verheyen published a very relevant manuscript titled "Acceptance of angulation in the non-operative treatment of pediatric forearm fractures." They stated that although angulated pediatric forearm fractures are renowned for their tendency to correct over time, many still consider them unpredictable. Internationally, there is still no consensus on the degree to which a deformity is acceptable.

You are an orthopedic surgeon. A 9-year-old child presents at the emergency department with a distal radius fracture with a severe angulation of 30°. Under procedural sedation, a closed reduction is performed, and a cast is applied. The X-ray reveals an anatomical reduction. One week later, the patient presents at your fracture clinic for cast replacement and radiographic follow-up. Unfortunately, the fracture has re-displaced with an angular deformity of 20°, a so-called re-angulation. What is the optimal treatment strategy?

In ***Chapter 2***, we report the findings from our study performed in Adelaide, Australia, which aimed to determine whether re-manipulation of re-angulated fractures in children leads to an improved long-term outcome or if re-angulations can be accepted, deeming that current treatment guidelines may be too strict.

Retrospectively, 66 children with a re-angulated distal forearm fracture were included, of which 24 underwent re-manipulation. In children aged <12 years, long-term outcomes of patients who underwent re-manipulation, did not differ from patients in which a re-angulation was accepted, deeming these re-manipulations unnecessary. Children aged ≥12 years with fractures that were not re-manipulated achieved satisfactory outcomes despite re-angulations exceeding the reduction criteria. Therefore, we titled our manuscript: *"Think twice before re-manipulating distal forearm fractures in children."*

Based on our results in combination with the literature, we suggest the following treatment guidelines on the acceptance of angulation in pediatric distal radius fractures:

<u>For distal forearm fractures in boys</u>:

- <9 years, accept up to 30° angulation.
- 9-11 years, accept up to 20° angulation.
- 11-13 years, accept up to 15° angulation.
- 13-15 years, accept up to 10° angulation.
- ≥15 years, accept up to 5-10° angulation.

<u>For distal forearm fractures in girls</u>:

- <8 years, accept up to 30° angulation.
- 8-10 years, accept up to 20° angulation.
- 10-12 years, accept up to 15° angulation.
- 12-14 years, accept up to 10° angulation.
- ≥14 years, accept up to 5-10° angulation.

Recently, there has been a trend toward increasingly more operative management, which takes away the opportunity for spontaneous correction of angulation by remodeling.

In *Chapter 3*, our study aimed to evaluate if K-wire fixation is essential for displaced distal metaphyseal forearm fractures to prevent long-term sequelae or if nature is forgiving. We asked: *Do We Need to Stabilize All Reduced Metaphyseal Forearm Fractures in Children with K-wires?*

We reported the extended follow-up of an RCT in which children with a displaced metaphyseal both-bone forearm fracture were randomized to closed reduction with or without K-wire fixation. This RCT recommended performing K-wire fixation, as children treated without K-wires had more re-displacements and more limitation in forearm rotation at short-term follow-up. Recently, there has been a trend toward increasingly more operative management. Therefore, we evaluated the long-term follow-up to check if this should be justified or if the well-known spontaneous correction of angulation, seen in pediatric fractures, might change treatment. 105 patients were included, of which 51 underwent K-wire fixation. At least five years after injury, there were no differences in radiographic or functional outcomes between children who did or did not receive K-wire fixation. However, malunion after re-displacement in a child who did not receive K-wires was a risk factor for developing a clinically relevant limitation in forearm rotation.

We concluded that children with displaced metaphyseal both-bone forearm fractures can be treated by closed reduction with or without K-wire fixation, as long-term outcomes did not differ.

We recommend that:

- In children with a displaced metaphyseal both-bone forearm exceeding the tolerable degree of angulation, a closed reduction can be performed without K-wire fixation in the emergency department. In this case, the clinician should inform parents and patients about the risk of fracture re-displacement and limited forearm rotation if left untreated. Weekly radiographic monitoring is recommended to detect re-displacement.
- If closed reduction is performed in the operating room, for instance, due to complete initial displacement, additional K-wire fixation should be performed. In this case, the risks of complications due to K-wire fixation should be discussed.

PART II: Diaphyseal forearm fractures in children

For distal forearm fractures in children, it has been shown that treatment in a below-elbow cast (BEC) instead of an above-elbow cast (AEC) is safe and more comfortable.

In ***Chapter 4,*** we investigated if early conversion to BEC after three weeks is safe for displaced diaphyseal both-bone forearm fractures which underwent closed reduction, even at long-term follow-up. We performed the extended follow-up of an RCT, in which 127 children were randomized to six weeks of AEC or early conversion to BEC at three weeks. At minimum 5-year follow-up, there were no significant differences in outcomes between both treatment groups. Therefore, it is time to change practice for diaphyseal forearm fractures and avoid the discomfort of unnecessarily immobilizing the elbow in an AEC for an extra three weeks.

Our recommendations:

- Early conversion to a below-elbow cast is the favored treatment for pediatric midshaft forearm fractures.

In ***Chapter 5,*** we analyzed the minimum 5-year follow-up outcomes of a cohort of 316 children with diaphyseal or distal metaphyseal both-bone forearm fractures. We asked:

1) *Which factors are associated with a limitation of pro-supination after pediatric both-bone forearm fractures at long-term follow-up?*
2) *Do accepted re-displacements of pediatric both-bone forearm fractures lead to inferior outcomes at long-term follow-up?*

316 participants with 149 diaphyseal and 167 distal metaphyseal fractures were included, with a mean follow-up of 7.2 years. Re-displacements occurred in 48% of conservatively treated displaced distal fractures and 35% of displaced diaphyseal fractures.

Predictors for a persisting impairment in forearm rotation after a both-bone forearm fracture in a child were a complete fracture of the ulna, an older age at trauma, and a diaphyseal fracture location. Excellent spontaneous remodeling of angular deformity by growth was seen in distal metaphyseal forearm fractures in children with remaining growth potential. Our study reaffirmed the old adage by Hughstone et al. from 1962: *"In midshaft forearm fractures, growth will not correct angular deformity as it does in distal fractures" (26).*

Based on our results in combination with the literature, we recommend the following criteria on the acceptance of angulation in pediatric diaphyseal forearm fractures:

For diaphyseal forearm fractures in all children:

- <9 years, accept up to 10° angulation.
- ≥ 9 years, accept up to 8° angulation.

Our treatment recommendations:

- For diaphyseal complete fractures of both forearm bones, especially in older children, we recommend performing closed reduction and additional stabilization in the operating room, even if they appear stable after reduction.
- All both-bone forearm fractures, which are treated without additional stabilization, should be monitored after 1 and 2 weeks to detect re-displacement which occur frequently.

PART III: Malunited forearm fractures in children

For patients with a malunited forearm fracture with symptomatic restriction in forearm rotation, a corrective osteotomy can be considered to restore normal bone alignment and thereby restore function. However, few articles have been published on the outcomes after corrective osteotomy. Therefore, we asked: *Who are the winners after a corrective osteotomy for a pediatric malunited forearm fracture?*

In ***Chapter 6,*** we presented our meta-analysis of individual participant data (IPD) on predictors of a superior functional outcome after corrective osteotomy for malunited radius or both-bone forearm fractures in children. Individual participant data from 11 cohort studies were included, concerning 71 participants, of which 55 underwent conventional corrective osteotomy, and 16 underwent 3D corrective osteotomy.

Predictors of a superior functional outcome after corrective osteotomy:

- An interval between trauma and corrective osteotomy of less than one year;
- An angular deformity greater than 20°
- The use of 3D computer-assisted techniques.

A corrective osteotomy is often challenging due to angular deformities of both the radius and the ulna involving three dimensions. Three-dimensional (3D) planning of the osteotomy and 3D printing of patient-specific instruments (PSIs) can simplify the surgical procedure. However, few studies examined the outcomes after 3D corrective osteotomy for pediatric forearm malunion. We asked: *What gain in forearm rotation can be achieved after 3D corrective osteotomy, and which factors are associated with a superior outcome?*

In ***Chapter 7,*** we described the results of our prospective study on the functional outcomes after 3D corrective osteotomies for pediatric malunited both-bone forearm fractures causing impaired pro-supination. Fifteen patients with a mean age at trauma of 10 years and time until osteotomy of 6 years were included. Our primary outcome measure was the gain in forearm rotation (pro-supination). Patients improved from 67° pro-supination pre-operatively to 118° at six months and 128° at 12 months follow-up. Supporting most findings of our IPD meta-analysis, this study revealed that predictors of greater functional gain after 3D corrective osteotomy are severe pre-operative impairment in pro-supination, shorter intervals until osteotomy and greater angulation of the radius.

In resume, 3D corrective osteotomy provided:

- A gain in pro-supination from 44% of the contralateral side pre-operatively to 77% at six months and 85% at 12 months follow-up.
- A greater pro-supination if there is a shorter interval between trauma and osteotomy and severe pre-operative impairment in pro-supination.

An anatomically accurate correction of a pediatric malunited forearm fracture is highly desirable to achieve the best functional outcome. A 3D corrective osteotomy can aid the surgeon in achieving a more accurate correction. Few studies have reported the effectiveness of 3D corrective osteotomy for pediatric malunited forearm fractures, with regard to the accuracy of the correction and gain in forearm rotation.

Therefore, in ***Chapter 8,*** we aimed to assess what accuracy of correction can be achieved after 3D corrective osteotomy and whether or not anatomic correction is associated with a greater functional gain. Our primary outcome measure was the residual maximum deformity angle (MDA) and malrotation after 3D corrective osteotomy. Post-operative MDA >5° or residual malrotation >15° was defined as non-anatomic corrections.

Our results:

- 3D corrective osteotomy provided an anatomic correction in 25 out of 30 operated forearm bones (10 out of 15 patients).
- Anatomic corrections resulted in greater final pro-supination than non-anatomic corrections: 136° versus 112° and a greater gain in pro-supination: 70° vs. 46°.
- Residual malrotation of the radius was associated with less gain in forearm rotation after 3D corrective osteotomy.

Our recommendations:

- We recommend to consider performing a 3D corrective osteotomy for patients with a diaphyseal forearm malunion with an obvious 3D bony deformity, a limitation of pro-supination ≥50°, preferably within two years after trauma.
- Pre-operative counseling is essential: *The rehabilitation must not be underestimated.*

PRACTICE-CHANGING ADVICE:

Angular deformity of pediatric forearm fractures

The true angular deformity may be underestimated: a dorsal angulation of 20° with a radial angulation of 15° leads to a true angulation of 25°.
Most distal metaphyseal forearm fractures exceeding the tolerable degree of angulation can be treated by a closed reduction in the emergency department without K-wires.
If closed reduction is performed in the operating room, for instance, due to complete initial displacement, additional K-wire fixation should be performed.
Excellent remodeling can be expected in distal metaphyseal forearm fractures in children with remaining growth. In contrast, in midshaft forearm fractures, growth will not correct angular deformity as it does in distal fractures.
Early conversion to a below-elbow cast after three weeks is recommended for pediatric minimally-displaced diaphyseal forearm fractures.
We recommend performing closed reduction and intramedullary pinning in the operating room for diaphyseal forearm fractures with complete fractures of both forearm bones in children older than 10.

Tips & Tricks for 3D corrective osteotomy:

Indication for 3D corrective osteotomy:	For diaphyseal forearm malunion with obvious 3D bony deformity, a limitation of pro-supination ≥50°, consider performing 3D corrective osteotomy. Preferably within two years after trauma.
Counseling:	Post-operative rehabilitation must not be underestimated.
Recommended order of osteotomy & fixation:	1. Volar Henry approach to the radius. 2. Standard ulnar approach (between ECU and FCU). 3. Osteotomy of the ulna. 4. Osteotomy of the radius. 5. Fixation of the radius. 6. Fixation of the ulna.
If there is unsatisfactory pro-supination after 3D corrective osteotomy:	1. Look for impingement of bone spikes created by the osteotomy & look for potential impingement of the plates. 2. Further release the interosseous membrane. 3. Release the dorsal DRUJ capsule for impairment in pronation. 4. Release the volar DRUJ capsule for impairment in supination. 5. Post-op casting in maximum pro- or supination for two weeks. 6. Dynamic bracing from 2-6 weeks and refer to a physiotherapist. 7. Night bracing in maximum pro- or supination for three months.
What can be expected after 3D corrective osteotomy?	• Gain in pro-supination from 44% to 85% of contralateral. • Greater functional can be achieved if there is: a shorter time until osteotomy, severe angulation, and severe pre-op limitation. • Minor complications occur in 20% (neuropraxia, plate removal). • Anatomic corrections are achieved in 83% of forearm bones. • Anatomic corrections result in a greater functional gain than non-anatomic corrections: 70◦ versus 46◦.

Chapter 11

Nederlandse Samenvatting

Deel I: Polsbreuken bij kinderen

In 2006 publiceerden Ploegmakers en Verheyen een zeer relevant manuscript getiteld "*Het accepteren van angulaties in de niet-operatieve behandeling van polsbreuken bij kinderen*". Zij stelden dat hoewel verplaatste polsbreuken bij kinderen vaak erg vergevingsgezind zijn vanwege de spontane neiging tot correctie door remodelering, zij door velen nog steeds als onvoorspelbaar worden beschouwd. Er is internationaal nog geen consensus over welke mate van angulatie (knikstand) geaccepteerd mag worden bij polsbreuken van kinderen in verschillende leeftijdsgroepen, zonder dat dit leidt tot consequenties op de lange termijn.

Stelt u zich voor: *U bent orthopedisch chirurg. Op de spoedeisende hulp komt een 9-jarig kind bij u, die zijn/haar pols heeft gebroken met een behoorlijke angulatie van circa 30°. U geeft het kind verdoving, de breuk wordt gezet en vervolgens ingegipst. Er wordt ter controle een röntgenfoto gemaakt, waarop de breuk weer mooi recht staat. Eén week later komt hij/zij terug bij u op de gipskamer voor een gipswissel en wordt er weer een röntgenfoto gemaakt. Helaas, de breuk toont opnieuw een scheefstand met zo'n 20° angulatie. Dit noemen we een re-dislocatie. Wat kunt u nu het beste doen?*

In ***Hoofdstuk 2*** bespreken wij een studie, die wij in Adelaide (Australië) hebben uitgevoerd naar de klinische lange-termijn uitkomsten bij kinderen na een polsbreuk. Specifiek hebben wij gekeken naar kinderen die theoretisch de slechtste uitgangssituatie hadden, namelijk de kinderen waarbij er een zogenaamde "re-dislocatie" is opgetreden. Dit zal ik toelichten met een voorbeeld:

Het doel van deze studie was om te achterhalen of het opnieuw zetten van de breuk leidt tot betere langetermijnuitkomsten dan het accepteren van deze scheefstand in afwachting van remodelering door de groei. Retrospectief hebben wij 66 kinderen met een re-dislocatie van een polsbreuk geïncludeerd, waarvan bij 24 kinderen de breuk opnieuw gezet was (re-manipulatie) en 42 kinderen waarbij dit niet gebeurd was. Vier jaar later verschilden de radiologische en functionele uitkomsten van patiënten die een re-manipulatie hadden gehad niet significant van patiënten bij wie een re-dislocatie was geaccepteerd. Hierdoor concludeerden wij dat deze re-manipulaties onnodig waren.

Op basis van deze studie en de beschikbare literatuur doen wij de volgende aanbevelingen voor het accepteren van scheefstand bij polsbreuken van kinderen:

Voor polsbreuken bij jongens:

- < 9 jaar, accepteer tot 30° angulatie.
- 9-11 jaar, accepteer tot 20° angulatie.
- 11-13 jaar, accepteer tot 15° angulatie.
- 13-15 jaar, accepteer tot 10° angulatie.
- ≥15 jaar, accepteer tot 5-10° angulatie.

Voor polsbreuken bij meisjes:

- < 8 jaar, accepteer tot 30° angulatie.
- 8-10 jaar, accepteer tot 20° angulatie.
- 10-12 jaar, accepteer tot 15° angulatie.
- 12-14 jaar, accepteer tot 10° angulatie.
- ≥14 jaar, accepteer tot 5-10° angulatie.

In ***Hoofdstuk 3*** beschrijven wij de langetermijn follow-up van een onderzoek naar de uitkomsten van kinderen met distale metafysaire antebrachium fracturen met dislocatie. De antebrachium fractuur is een botbreuk van zowel de radius (spaakbeen) als de ulna (ellepijp) in de onderarm en wordt in het Latijn de *fractura antebrachii* genoemd. Een distale metafysaire breuk is een breuk dichtbij de groeischrijf van de pols. In dit onderzoek werden kinderen gerandomiseerd naar repositie met of zonder stabilisatie met twee K-draden (ijzeren pennetjes). Kinderen die zonder K-draden werden behandeld hadden meer re-dislocaties en meer beperking in pro-supinatie (rotatie van de onderarm) bij follow-up op de korte termijn, waardoor er werd aanbevolen K-draadfixatie toe te passen. De laatste jaren is er een trend naar steeds meer operatieve behandeling zonder dat er sterk bewijs voor is ten aanzien van de lange termijn uitkomsten. Wij evalueerden de langetermijn follow-up om na te gaan of dit te rechtvaardigen is. Wij hebben 105 patiënten geïncludeerd, waarvan 51 een K-draadfixatie ondergingen. Bij de lange-termijn follow-up waren er geen verschillen in radiologische of functionele resultaten tussen kinderen die wel of geen K-draadfixatie kregen.

Op basis van deze studie zouden we volgende aanbevelingen willen doen:

- Bij kinderen met een distale antebrachium fractuur kan er gekozen worden om de breuk recht te zetten zonder aanvullende fixatie met K-draden. In dat geval dient de behandelend arts met het kind en de ouders te bespreken wat de risico’s zijn van een re-dislocatie en adviseren wij om de eerste 2 weken wekelijks röntgenfoto’s te maken om geen re-dislocaties te missen.

- Indien op de operatiekamer een repositie wordt uitgevoerd, bijvoorbeeld als gevolg van een volledige initiële verplaatsing, adviseren wij om direct K-draadfixatie toe te passen. In dit geval moeten de risico's van complicaties als gevolg van K-draadfixatie worden besproken.

Deel II: Breuken in het midden van de onderarm

In *Hoofdstuk 4* hebben wij een studie gedaan naar de uitkomsten van breuken in het midden van de onderarm, de zogenaamde diafysaire antebrachium fracturen. Wij hebben onderzocht of deze breuken na repositie gedurende zes weken in een bovenarmgips behandeld moeten worden, of dat er na drie weken gewisseld kan worden naar een onderarmgips. Eerder lieten de korte termijn uitkomsten van een onderzoek geen nadelige gevolgen zien van deze gipswissel na drie weken. De wissel naar het onderarmgips werd als een stuk comfortabeler ervaren door de kinderen. Met onze huidige studie hebben we gekeken naar de langetermijnuitkomsten van dit onderzoek en waren er opnieuw geen nadelige gevolgen van het wisselen van een bovenarmgips naar een onderarmgips na drie weken.

We adviseren dat:

- Conversie naar een onderarmgips na drie weken is de aanbevolen nabehandeling voor diafysaire antebrachium fracturen.

In *Hoofdstuk 5* hebben wij de langetermijnuitkomsten van een cohort van 316 kinderen met een distale metafysaire of diafysaire antebrachium fractuur bestudeerd. Hierbij hadden wij als vraagstellingen:

1. Welke factoren zijn geassocieerd met een beperking in pro-supinatie na een antebrachium fractuur bij lange-termijn follow-up?
2. Zorgen geaccepteerde re-dislocaties van een antebrachium fractuur voor slechtere uitkomsten op de lange termijn?

316 kinderen met 149 diafysaire en 167 distale antebrachium fracturen werden geïncludeerd met een gemiddelde follow-up van 7.2 jaar. Re-dislocaties kwamen voor bij 48% van de conservatief behandelde gedisloceerde distale fracturen en bij 35% van de gedisloceerde diafysaire fracturen.

Voorspellers voor een blijvende functiebeperking na een antebrachium fractuur waren: een complete breuk van de ellepijp, een oudere leeftijd en een diafysaire fractuur locatie.

Scheefstand van distale metafysaire antebrachium fracturen bij kinderen bleek erg vergevingsgezind; door de groei van het kind herstelden deze breuken in een goede stand. Dit was niet helemaal het geval voor diafysaire antebrachium fracturen, hierbij werd minder remodelering gezien. Hiermee werd een oud adagium bevestigd: *"Bij breuken midden in de onderarm bij kinderen zal groei de scheefstand niet herstellen zoals in polsbreuken" – Hughstone et al, 1962.*

Wij doen de volgende aanbevelingen omtrent het accepteren van een scheefstand van een onderarmbreuk bij kinderen:

Voor diafysaire onderarm breuken bij kinderen:

- < 9 jaar, accepteer tot 10° angulatie.
- ≥ 9 jaar, accepteer tot 8° angulatie.

Onze aanbevelingen aan de hand van deze studie zijn:

- Bij kinderen kunnen de meeste antebrachium fracturen gelokaliseerd dichtbij de pols (al of niet na het zetten) behandeld worden in gips, aangezien we op lange termijn zeer goede functionele uitkomsten zien.
- Als er sprake is van een antebrachium fractuur gelokaliseerd midden in de onderarm, waarbij de botten volledig doorgebroken zijn, adviseren wij om bij kinderen ouder dan 10 jaar op de operatiekamer de breuk recht te zetten en te stabiliseren met flexibele pennetjes.
- Alle onderarmbreuken, die behandeld worden zonder aanvullende stabilisatie, dienen na 1 en 2 weken een röntgenfoto te krijgen om geen re-dislocaties te missen.

Deel III: Verkeerd vastgegroeide botbreuken van de onderarm

Helaas ontstaat er soms een “malunion”, dat wil zeggen: de botten op de plaats van de breuk zijn in een verkeerde stand aan elkaar vastgegroeid. Dit kan gepaard gaan met pijn en functiebeperking. Een dergelijke malunion kan worden gecorrigeerd door middel van een correctie osteotomie. Tijdens deze operatie worden de verkeerd vastgegroeide botten van de onderarm doorgezaagd en vervolgens in de juiste stand vastgezet met platen en schroeven.

In ***Hoofdstuk 6*** presenteren wij onze meta-analyse van individuele patiënten data naar voorspellers voor een goed functioneel resultaat na een correctie osteotomie wegens een post-traumatische onderarm malunion ontstaan tijdens kinderleeftijd. Hiervoor hebben wij de data van 71 patiënten uit 11 verschillende studies samengevoegd. Van deze patiënten ondergingen er 55 een conventionele correctie osteotomie en bij 16 patiënten werd er gebruik gemaakt van 3D computer-geassisteerde technieken, de zogenaamde 3D correctie osteotomie.

Doorslaggevende factoren voor goede resultaten na een correctie osteotomie voor een malunion van een onderarm breuk op kinderleeftijd waren:

- Een korter tijdsinterval tussen de breuk en de correctie osteotomie.
- Een angulatie van de radius van 20° of meer.
- Het gebruik van de 3D computer geassisteerde techniek, de zogenaamde 3D correctie osteotomie.

In ***Hoofdstuk 7*** presenteren wij onze prospectieve studie naar de uitkomsten na 3D correctie osteotomieën wegens een malunion na een antebrachium fractuur. Vóór de ontwikkeling van deze 3D techniek, werd de correctie altijd verricht op het “timmermansoog”. De resultaten hiervan waren regelmatig teleurstellend, waardoor veel orthopedisch chirurgen terughoudend waren met het uitvoeren van deze operatie. Om de ingreep minder complex en beter voorspelbaar te maken zijn wij gestart met de 3D correctie osteotomie studie. In deze studie planden we met behulp van computermodellen de correctie osteotomie al vóór de operatie. Hiervoor maken wij een CT-scan van beide onderarmen. Door de gezonde arm nu te vergelijken met de aangedane arm kunnen we al voor de operatie berekenen wat de beste plek is om de botten door te zagen. Met een 3D printer werden er mallen gemaakt, waarmee de orthopedisch chirurg nauwkeurig kon opereren. Vijftien patiënten ondergingen 3D correctie osteotomie met een gemiddelde traumaleeftijd van 10 jaar en een gemiddelde tijd tot osteotomie van 6 jaar.

3D correctie osteotomie zorgde voor:

- Een verbetering van de pro-supinatie van 44% van de contralaterale zijde pre-operatief tot 85% van contralateraal bij de 1-jaars follow-up.
- Meer winst in functie indien er sprake was van een korter interval tussen trauma en 3D correctie ostetomie en/of ernstige pre-operatieve functiebeperking.

Conventionele correctie osteotomie met 2D planning voor malunions van zowel de radius als de ulna kan een zeer ingewikkelde procedure zijn, wegens deformiteiten in drie verschillende dimensies. Een 3D correctie osteotomie kan de orthopedisch chirurg mogelijk helpen bij het bereiken van een anatomische correctie. Tot op heden hebben weinig studies de effectiviteit van 3D correctie osteotomieën voor malunions na antebrachium fracturen bij kinderen, met betrekking tot de accuratesse van de correctie en daarmee gepaard gaande winst in pro-supinatie onderzocht.

In *Hoofdstuk 8* hebben wij gekeken naar de mate van nauwkeurigheid van de 3D correctie osteotomie en of een anatomische correctie geassocieerd is met een betere winst in pro-supinatie. Onze primaire uitkomstmaat was de maximale deformiteit angulatie (MDA). Hierbij worden de angulaties in verschillende richtingen volgens de stelling van Pythagoras bij elkaar opgeteld. Tevens hebben we gekeken naar de mate van malrotatie. Een post-operatieve MDA >5° of malrotatie >15° werden als een niet-anatomische correctie geduid.

Onze resultaten:

- 3D correctie osteotomie zorgde voor een anatomische correctie in 25 van de 30 geopereerde onderarm botten, in 10 van de 15 geopereerde patiënten.
- Anatomische correctie na een 3D correctie osteotomie zorgde voor een betere winst in draaifunctie van de onderarm dan een niet-anatomische correctie (70° vs. 46°).
- Persisterende malrotatie van de radius na 3D correctie osteotomie was geassocieerd met een beperking in pro-supinatie.

Onze aanbevelingen:

- Wij adviseren het uitvoeren van een 3D correctie osteotomie te overwegen bij patiënten met een diafysaire malunion na een antebrachium fractuur met: een duidelijke 3D deformiteit, een beperking van de pro-supinatie ≥50°, bij voorkeur binnen 2 jaar na het trauma.
- Pre-operatieve voorlichting is zeer belangrijk: de revalidatie mag niet worden onderschat.

AANBEVELINGEN:

Fractura antebrachii bij kinderen

De werkelijke angulatie kan worden onderschat: een dorsale angulatie van 20° met een radiale angulatie van 15° leidt tot een werkelijke angulatie van 25°.
De meeste distale metafysaire onderarmfracturen die de acceptabele mate van angulatie overschrijden, kunnen worden behandeld door een gesloten reductie op de spoedeisende hulp zonder K-draadfixatie.
Indien er op de operatiekamer een gesloten repositie wordt verricht, bijvoorbeeld wegens een volledig verplaatste breuk, adviseren wij om K-draadfixatie uit te voeren.
Bij distale metafysaire onderarmfracturen bij kinderen met resterende groei kan een uitstekende mate van remodellering worden verwacht. Bij mid-schacht onderarmfracturen daarentegen zal de groei de malunion niet corrigeren zoals bij distale fracturen.
Voor minimaal verplaatste diaphysaire onderarmfracturen bij kinderen wordt drie weken bovenarmgips, gevolgd door drie weken onderarmgips aanbevolen.
Wij adviseren intramedullaire penfixatie voor diafysaire onderarmfracturen met volledige fracturen van beide onderarmbotten bij kinderen ouder dan 10 jaar.

Tips & Tricks voor een 3D correctie osteotomie:

Indicaties voor een 3D correctie osteotomie:	Overweeg om bij diaphysaire malunions van de onderarm met duidelijke 3D ossale afwijking en een beperking van de pro-supinatie ≥50° een 3D correctie osteotomie uit te voeren. Bij voorkeur binnen twee jaar na het trauma.
Voorlichting:	De post-operatieve revalidatie mag niet worden onderschat.
Aanbevolen volgorde van osteotomie en fixatie:	1. Benadering van de radius. 2. Benadering van de ulna. 3. Osteotomie van de ulna. 4. Osteotomie van de radius. 5. Fixatie van de radius. 6. Fixatie van de ulna.
Indien er onvoldoende winst in pro-supinatie is na 3D correctie osteotomie:	1. Beoordeel of de door de osteotomie ontstane botpieken en de platen leiden tot impingement. 2. Maak het ligamentum interosseum los. 3. Maak het dorsale distale radio-ulnaire gewricht (DRUJ) kapsel los indien er pronatie beperking is. 4. Maak volaire DRUJ-kapsel los indien er supinatie beperking is. 5. Leg post-operatief een bovenarmgips aan gedurende 2 weken bovenarmgips in maximale pro- of supinatie. 6. Start dynamische bracing in maximale pro- of supinatie gedurende 2-6 weken. 7. Start nacht bracing in maximale pro- of supinatie.
Wat kan er verwacht worden na een 3D correctie osteotomie?	• Winst in pro-supinatie van 44% tot 85% van contralateraal. • Meer winst indien: een korter interval tot de osteotomie, meer angulatie en ernstige preoperatieve beperking. • Milde complicaties treden op in 20% (neuropraxie, klachten van het osteosynthesemateriaal). • Anatomische correcties worden bereikt in 83%. • Anatomische correcties resulteren in een betere winst in functie dan niet-anatomische correcties: 70◦ versus 46◦.

Chapter 12

PhD portfolio / Curriculum Vitae

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06-2022 **FESSH/IFSSH, London:** Radiographic and functional outcomes of 3D corrective osteotomies for pediatric malunited both-bone forearm fractures

09-2020 **FESSH, Basel:** 3D corrective osteotomies for pediatric malunited diaphyseal forearm fractures: Is anatomic correction associated with a greater functional gain?

01-2020 **NOV 's Hertogenbosch:** 3-D corrective osteotomies for pediatric forearm malunions: what gain in forearm rotation can be achieved?

11-2019 **Traumadays (NVT),** Amsterdam: Accurate correction and good function after 3D corrective osteotomies for pediatric diaphyseal both-bone forearm malunions

06-2019 **IFSSH/FESSH, Berlin:** 3D corrective osteotomies for pediatric malunited diaphyseal forearm fractures: do we achieve the desired functional gain and the planned correction?

07-2018 **EFORT, Barcelona:** Which factors affect limitation of pro-supination after both-bone forearm fractures in children?

07-2018 **EFORT, Barcelona:** Corrective osteotomies for pediatric malunited forearm fractures: preliminary results

07-2018 **FESSH, Copenhagen:** Which factors affect limitation of pro-supination after both-bone forearm fractures in children?

07-2018 **FESSH, Copenhagen:** Three-dimensional virtual planning of corrective osteotomies for pediatric malunited forearm fractures

07-2018 **FESSH, Copenhagen:** Factors determining outcome of corrective osteotomy for malunited pediatric forearm fractures: a systematic review

03-2018 **3rd Trauma Platform, Davos, Zwitersland:** Which factors affect limitation of pro-supination after both-bone forearm fractures in children?

01-2018 **NOV 's Hertogenbosch:** "Long-term outcomes after both-bone forearm fractures in children."

11-2017 **Amphia Science day, Breda:** "Corrective osteotomies for pediatric forearm malunions: who are the winners?"

11-2016 **International Conference NVvH:** "Corrective osteotomies for forearm malunions in children: who are winners?"

11-2016 **Traumadays (NVT),** Amsterdam: "Corrective osteotomies for forearm malunions in children: who are winners?"

06-2016 **FESSH, Santander:** “Corrective osteotomies for forearm malunions in children: who are winners?”

06-2016 **FESSH, Santander:** “Think twice before re-manipulating distal metaphyseal forearm fractures in children”

01-2016 **NOV, ’s Hertogenbosch:** “Corrective osteotomies for forearm malunions in children: who are winners?”

11-2015 **Traumadays (NVT), Amsterdam:** “Think twice before re-manipulating distal metaphyseal forearm fractures in children”

01-2015 **Residents symposium NVT, Soest:** “Think twice before re-manipulating distal metaphyseal forearm fractures in children”

01-2014 **NOV, Rotterdam: “**Think twice before re-manipulating distal metaphyseal forearm fractures in children”

POSTER PRESENTATIONS

05-2023 **FESSH, Rimini:** Predictors for a limitation of pro-supination after pediatric both-bone forearm fractures: the 7-year follow-up of 316 patients

01-2018 **Brussels Hand Symposium:** Distraction osteogenesis of the distal radius and ulna in young patients to obtain a normal DRUJ anatomy

01-2018 **Brussels Hand Symposium:** Corrective osteotomies for pediatric malunited forearm fractures: IPD meta-analysis & preliminary results of a prospective study

11-2017 **Amphia Science day, Breda:** “Think twice before re-manipulating distal metaphyseal forearm fractures in children”

PRIZES

11-2017 **Pieter Stijnen Prize**: Best poster presentation at Amphia Science Day

01-2015 **Dr. G. J. Heijmans Prize** Best presentation at Residents symposium

ORGANIZATIONAL

2016	Organisation of Summerclass Orthopedics, Delft
2015-2018	Editor bij 4Bone. (http://4abstracts.nl/4b)
2018 - now	Board member of the NOV workgroup Hand & Wrist

COURSES/FELLOWSHIPS

2016	Advanced Trauma Life Support
2018	Good Clinical Practice
2019	AO Basic Principles of Fracture Management, Leeds, UK
2020	ATLS Refresher
2021	Scientific Integrity Course, Erasmus MC
2022	Travelling fellowship Hand Surgery Zürich
2022	FESSH Academy - Budapest
2022	OTC 2 & 3 course
2022	Good Clinical Practice Refresher

Chapter 13

Dankwoord / Acknowledgements

Dankwoord

De leden van de beoordelingscommissie: **prof. dr. M. H. J. Verhofstad**, **prof. dr. E. H. G. Oei, prof. dr. I. B. Schipper** en **prof. dr. J. N. Doornberg**. Hartelijk dank voor de tijd en moeite die u heeft genomen om mijn proefschrift te lezen en de bereidheid zitting te nemen in de beoordelingscommissie.

Mijn promotor **prof. Eygendaal**, beste Denise, wat ontzettend leuk dat je mijn promotor bent geworden. Ik kan mij nog goed de dag herinneren dat wij elkaar ontmoetten in 2017 tijdens een lunch-sollicitatiegesprek voor een ANIOS plek in het Amphia. Je begon vrijwel direct met de vraag: "*Hoe gaan wij jou in opleiding krijgen?"* Eenmaal gestart als ANIOS gaf ik aan dat ik interesse had in hand- en polschirurgie, waarna ik direct mijn eigen hand-pols-elleboog poli kreeg. Voor mij heel inspirerend hoe je mensen enthousiasmeert en concreet met ze aan de slag gaat. Ik ga mijn best doen om deze eigenschap van jou over te nemen.

Mijn promotor **prof. Stockmans**, beste Filip, bedankt voor alle uren die wij samen hebben doorgebracht met het verrichten van de 3D metingen op uw laptop in het operatiecomplex van het AZ Groeninge, te Kortrijk. Alle tijd en moeite die u in dit 3D correctie osteotomie project hebt gestopt is indrukwekkend. Ik heb erg veel gehad aan uw technische blik en kritische discussies.

Mijn copromotor **dr. Colaris**, beste Joost, je bent oprecht de beste copromotor die ik mij had kunnen wensen! Al direct vanaf het begin (2013) bezig met het einddoel (dit proefschrift) in gedachten. Continu bezig met alles eruit halen: via Whatsapp berichtjes als *"Kasper, stuur jij nog abstracts in voor de FESSH? Deadline is bijna!"* Je hebt mij hierdoor letterlijk meegenomen in de wereld van de handchirurgie. Het is mijn *spark* geweest om mij te willen specialiseren in de hand- en traumachirurgie.

Eline van Es, jij bent de afgelopen jaren mijn *go-to person* geweest als ik er zelf niet helemaal uit kwam. Erg mooi om te zien hoe je van junior research coördinator uitgegroeid bent tot 3D planning specialist van de afdeling.

Maria Osorio, thank you so much for all the Skype sessions in which you helped me out with all the problems I ran into while performing our 3D analyses.

Leon Diederix, wat een hoop tijd en energie heb jij in dit project gestopt. Ik hoop dat **Linde Musters** en ik samen een mooi einde kunnen maken aan het onderzoek waar jij mee gestart bent.

Mijn opleiders **dr. Bos**, **dr. Kraan** en **dr. van der Linde**, beste Koen, Gerald en Just, bedankt voor jullie geduld, als ik telkens opnieuw kom met een verzoek voor extra congres- of cursusdagen, PhD tijd of een fellowship in het buitenland. Ik heb van jullie beiden zeer regelmatig te horen gekregen: *"het doel van de opleiding orthopedie is om jou orthopedisch chirurg te maken en niet promovendus*, *handchirurg of dokter in het buitenland.*" Ik ben erg dankbaar dat ik jullie als opleiders heb.

Mijn paranimfen, **Alex Roth** en **Bianca Rijken**.

Beste **Alex,** in 2017 mocht ik jouw paranimf zijn en ik zou graag exact dezelfde woorden willen terugkaatsen, die je toen over mij zei: *"Bro, fantastisch al die reizen die we samen hebben gemaakt de afgelopen jaren! Ik ben er trots op dat je mijn paranimf wilt zijn en heb niemand liever naast m'n zijde staan!"*

Beste **Bianca,** ik hoor het je nog zeggen: "Kasper*, ben je nou nog niet klaar met je PhD?!"* Mega leuk dat ik jou in 2015 op een congres heb leren kennen. We grapten toen dat je mijn paranimf zou worden en je staat er straks op 9 september 2023 daadwerkelijk. Oh, en nogmaals excuses dat ik je loepbril heb gesloopt op mijn huisfeestje.

Prof. Dr. Jaarsma, beste Ruurd, ontzettend bedankt dat ik mijn master research kon doen bij jou in Adelaide, Australië in 2013. Hieraan heb ik mijn eerste publicatie te danken, samen met jou en **Katharina Denk**. Daarnaast heb ik zeer mooie herinneringen overgehouden aan mijn tijd in Adelaide met als highlight: het behalen van de Australian Football Finals met het Flinders University FC team. Ik kijk er erg naar uit om in 2026 terug te komen voor een trauma fellowship.

Uit mijn Amphia tijd wil ik verder nog bedanken: **Elsa Spaans**, wat waren wij een gezellig ANIOS orthopedie duo. Thanks voor de Mimosa foto tijdens de EFORT in Barcelona toen ik dacht aangenomen te zijn voor de opleiding orthopedie! Dat bleek enkele weken later pas echt waar te zijn. **Annechien Beumer**, jij hebt mij laten zien dat je het toch ver kan schoppen zonder ooit je mailbox te openen. **Bertram The**, jij hebt mij altijd geprikkeld om meer te willen leren, bijvoorbeeld door mij door allerlei ingewikkelde ASSH examen vragen te laten ploeteren voor een wekelijkse AIOS hand quiz ter voorbereiding op hun CCOC examen. **Christaan van Bergen**, jij bent voor mij een voorbeeld van dé orthopeed, die ik later wil worden: oprecht geïnteresseerd, operatief zeer kundig en daarnaast een relaxte vent, waar je na het kitesurfen in Tarifa gezellig een biertje mee kan drinken. **Geert Buijze**, jou zie ik ook als een goed voorbeeld: als je iets wilt, ga je er vol voor. Zo heb jij een mooie plek bemachtigd in Frankrijk als bovenste extremiteit- en trauma specialist.

De FESSH party crew met o.a. **Jaime Ruas**, **Jantine Posthuma, Jorien Werkman, Veronique van de Lucht, Louise de Haas, Bas Derksen** en **Phillip D'Ailly** Ik vind het echt heel leuk dat wij in London en Rimini zo'n gezellig FESSH borrel clubje hebben gecreëerd. Dat elk jaar steeds wat groter wordt. De FESSH volgend jaar in Rotterdam wordt mooi!

Mijn goede vrienden, **Thomas Coebergh**, **Niels van Peer** en **Victor van Oorschot**. Inmiddels hebben wij alle vier Rotterdam verlaten. Ik kijk terug op ontzettend veel mooie herinneringen met als hoogtepunt onze reis door Zuid-Amerika en kijk uit naar de volgende.

Mijn huisgenoot **Ralph Pijls**. Mede dankzij jou heb ik mij na mijn overstap naar Amsterdam direct op mijn plek gevoeld. Ook heb jij mij geïntroduceerd aan de term “Lil Friday” samen met **Maxim Bours**, **Bas Wolff, Murk de Roos** en **Salto Sam.**

Mijn wielrenmaatjes van de “Fietsen A’dam” whatsapp groep met o.a. **Koen van Maasakkers, Jorn Heeringa**, **Luuk Schellens**, **Job Baken** en **Robin Botman**. Het zinnetje *“Vandaag gaan we echt rustig aan doen”* ben ik erg vaak ingetrapt. Met jullie heb ik mooie tripjes gemaakt naar le Col de la Croix-de-Fer, de Doppio Stelvio en El Teide. Met **Koen** de Mont Ventoux beklimmen op 15 jarige leeftijd stond aan het begin.

Mijn kitesurfmaatjes, met o.a. **Axel**, **Barry**, **Wiebe**, **Joost** en **Erik**. Onze kitesurf reizen naar Jericoacoara of Tarifa waren telkens een hele goede afleiding om erna volledig uitgerust terug te komen om weer aan mijn PhD te gaan werken.

Mijn vriendjes-van-vroeger met o.a. **Rens Kruisbrink, Joost Löring, Thomas Eggen, Bob Klaassen** en mijn *brother from another mother*, **Jelmer Vriesema**. Ik zie jullie niet zo vaak als ik zou willen, maar toch blijft het elke keer als vanouds.

Mijn oma **Annie**, dank u voor al uw bijna-wekelijkse Whatsappjes (ondanks uw 94 jaar) waarin u mij succes wenste bij het afronden van mijn proefschrift. Telkens zei u: “Ik hoop dat ik je verdediging nog mee kan maken”.

Mijn broer, **Ivan Roth**, jij bent een rolmodel voor mij geweest en zette mij steeds aan om overal het maximale uit te halen. Heel leuk dat ik je mij aan het kitesurfen hebt gekregen. Nu doe ik hetzelfde en smeer ik al mijn vrienden een hoop kitesurf lesjes aan.

Mijn broer, **Alex Roth**, respect voor hoe jij jouw orthopedische start-up “Avalanche Medical” aan het uitrollen bent met een nieuw implantaat tegen kraakbeendefecten. Al vanaf 2014 zijn wij regelmatig samen te vinden op de NOV jaarcongressen, maar ook op de fiets in de Limburgse heuvels.

Mijn zusje, **Kristel Roth**, ik kan jou altijd bellen. Daarom doe ik dat ook. Je betekent heel veel voor mij, *my lil sister*.

Mijn vader, **Ruud Roth**, ik kan mij voorstellen dat het niet altijd even makkelijk is geweest, maar ik ben je dankbaar voor jouw support. Ook voor de hulp bij de opmaak van dit proefschrift.

Lieve mam, **Ciska Roth-Baljeu,** je bent er echt echt echt altijd voor mij en daar ben ik je ontzettend dankbaar voor!

ISBN :
xxxxxxxxx

Printing:
Ipskamp Printing, Enschede

Lenticular cover printing:
Bergman Media, Gorinchem

Cover Design:
Rudy Roth, Stijn Hoekstra & Kasper Roth