Older age and multi-joint external fixator are two risk factors of complications in ulnar lengthening in children with hereditary multiple exostosis

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Research article

Keywords: Hereditary multiple exostosis, Ulna lengthening, Unilateral external fixation, Forearm deformity

DOI: https://doi.org/10.21203/rs.3.rs-47412/v2

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Abstract

Objectives: Hereditary multiple exostosis (HME) often involves forearm deformities. The aim of this study was to present the clinical results of 37 children who underwent ulnar lengthening with two different types of unilateral external fixators, and to investigate the risk factors of complications. Methods: We evaluated 37 children with forearm deformities caused by HME treated in our hospital from January 2008 to July 2019. The surgical procedures included resection of exostosis, osteotomy of the ulna, and gradual lengthening of the ulna with a single arm external fixator. According to the type of external fixator they received, the children were divided into two groups: group A received monorail fixators, and group B received multi-joint fixators. Radiographic and functional parameters were assessed. Complications were recorded. Results: All patients were followed up for an average of 4.6 years (3.0 to 6.5). In both group A and group B, the ulna shortening (US), radial articular angle (RAA), carpal slip (CS), elbow flexion, forearm pronation, supination, and Mayo Elbow Performance Score (MEPS) values improved significantly from preoperatively to postoperatively (p < 0.05). However, ulnar deviation was observed in 4 cases in group B and no cases in group A. According to logistic regression, the difference was only related to age (p < 0.05) and the type of external fixator (p < 0.05). Conclusions: Ulnar lengthening with unilateral external fixation is a safe and effective procedure for the treatment of HME. Regarding complications, deviation of the ulna axis was more likely to occur in older children with multi-joint external fixators.

Introduction

Hereditary multiple exostosis (HME) is a type of autosomal dominant inheritance-induced skeletal dysplasia, with an incidence of approximately 1/50,000 [1]. Approximately 30–60% of cases involve the forearm, affecting the longitudinal growth of the metaphysis and resulting in slow or stagnant growth of the ulna or radius. Deformities such as ulnar shortening, radius curvature, wrist ulnar deviation and radial head dislocation gradually appear with age [2].

The optimal timing and surgical treatment for forearm deformity caused by HME remains controversial [3, 4, 5]. The most common treatment is proximal ulnar osteotomy. Different external fixators are used to gradually lengthen the ulna and correct forearm deformities [4, 6, 7, 8]. In particular, the unilateral external fixator is a reliable and effective method with advantages such as being simple to use in operations, leading to minimal surgical trauma, being easy to maintain post-operatively and yielding significant lengthening of the ulna; thus, it has been widely used [9, 10]. However, due to the curvature of the deformity of the ulna itself, the end of ulna osteotomy region may deviate from the axis during the lengthening process. Considering the age and nutritional status of the patients, reports about various complications, such as delayed union of the osteotomy end and malunion, are common [11, 12]. Launay F et al [13] inserted an axial Kirschner wire into the ulna to avoid axis deviation and guide the ulna during lengthening. Nevertheless, the relationship between the types of external fixators and complications remain unclear.

Thirty-seven children with forearm deformities caused by HME were enrolled in this study, and they underwent the surgical procedure of ulnar lengthening with two different types of single arm external fixators. The aim of this study was to retrospectively analyse and present the clinical results and compare the two types of external fixators in terms of the complications that occur postoperatively.

Methods

Patients
Thirty-seven patients (26 males and 11 females) with forearm deformities caused by HME were treated in our hospital from January 2008 to July 2019. All of the deformities were unilateral. The age at operation ranged from 4.5 to 12.5 years (average 7.4 years). The forearm deformities were classified according to the Masada Classification system[2,14], which is based on the morphological characteristics of the deformity on plain radiographs (Fig. 1).

According to the type of single arm external fixators applied in surgery, the 37 patients were divided into two groups: group A received monorail fixators, and group B received multi-joint fixators. The patients’ basic information is shown in Table 1.

| Subject                      | Group A          | Group B          | P*   |
|------------------------------|------------------|------------------|------|
| Number (n)                   | 27               | 10               | -    |
| Age (year)                   | 7.30 ± 1.80 (4.5–12.0) | 8.03 ± 2.42 (5.0–12.5) | 0.084 |
| Sex (n) Male to female       | 22:5             | 4:6              | 0.555|
| BMI (Kg/m²)                  | 15.8 ± 1.20 (13.6–18.9) | 16.72 ± 1.14 (14.8–18.8) | < 0.05 |
| Type (n) I:6; IIa:11; IIb:10; III:0 | I:3; IIa:4; IIb:3; III:0 | < 0.05 |
| Side (n) Left to right       | 15:12            | 4:6              | -    |
| Type of external fixator     | Monorail         | Multi-joint      | -    |
| Follow-up (year)             | 4.53 ± 1.42 (3.0-6.5) | 4.87 ± 1.33 (3.0-5.8) | 0.389 |
| US (mm)                      | 23.15 ± 5.55     | 23.40 ± 4.84     | 0.801|
| RAA (°)                      | 32.30 ± 4.07     | 31.30 ± 5.36     | 0.353|
| CS (%)                       | 70.37 ± 6.70     | 72.30 ± 4.35     | 0.408|
| MEPS                         | 74.07 ± 3.68     | 74.50 ± 3.69     | 0.775|
| Elbow Flexion (°)            | 104.26 ± 4.94    | 106.50 ± 4.74    | 0.242|
| Forearm Pronation(°)         | 71.11 ± 3.76     | 69.00 ± 3.94     | 0.180|
| Forearm supination(°)        | 83.33 ± 3.67     | 83.00 ± 4.22     | 0.749|

*By Mann-Whitney U test, p value < 0.05 Statistically significant

The inclusion criteria for surgery included worsening of the deformity or impairments in daily activities, such as ulnar shortening (US) by at least 15 mm or 8% of the length of the ulna[16], associated radial head dislocation, and dysfunction of the elbow, forearm, or wrist. For dysfunction, we evaluated movements of elbow flexion inferior to 110°, and pronation inferior to 70° or pronation function inferior to 40% compared with that of the contralateral side. Elbow extension and forearm supination were not included in the inclusion criteria because they have not been identified as obvious indicators of dysfunction in the clinical literature. The exclusion criteria were as follows: patients with good appearance and function of the upper limb, traumatic dislocation of the radial head, age of less than 3 years old, malnutrition, or any other condition considered a contraindication for operation and anaesthesia.

Surgical procedure
All surgical procedures were performed by the same senior surgeon. Under general anaesthesia, each patient was placed in the supine position. A longitudinal incision was made according to the location and size of the exostosis. The exostosis, as well as the surrounding periosteum, was bluntly separated and completely exposed. Adjacent vessels, nerves and epiphyses were protected. Four Schanz pins were placed on the ulna in parallel to each other and on the same plane, avoiding the plane of maximum curvature on the back of the ulnar arch. The preferred location for osteotomy was the proximal part of the ulna near the epiphysis, avoiding the middle segment of the ulna and leaving enough space for Schanz pins. Then, a monorail or multi-joint single arm external fixator (Orthopaedic External Fixator Systems, Via delle Nazioni 9, 37012 Bussolengo, Verona, Italy) was applied. In patients with dislocation of the radial head, no special procedures were performed for the radial head.

Radiological and clinical evaluation

According to Fogel et al [15], the radiological evaluation indicators of forearm deformities caused by HME mainly include ulna shortening (US), the radial articular angle (RAA), and carpal slip (CS) (Fig. 2). The above indexes were recorded preoperatively and at the final follow-up. Deviations in the ulna axis, poorly regenerated bone formation, redislocation of the radial head, the recurrence of exostosis, and other complications were also recorded. In the clinical evaluations performed preoperatively and at the final follow-up, elbow flexion and forearm rotation were recorded. The Mayo Elbow Performance Score (MEPS) [17] was used to evaluate elbow function.

Ulnar lengthening and follow-up

Many methods are available to estimate the lengthening length. To avoid the recurrence of deformity, most scholars suggest that the length of the ulna should be overextended by 4~10 mm more than the length of the contralateral ulna [6, 10, 15]. In this study, the length of the contralateral ulna + 4~10 mm was used as the target length. The ulna was lengthened 3~4 times a day on the 7th day after operation, and the lengthening knob was adjusted 90° each time, that is, 0.75-1 mm/day. The sensation, movement and circulation of the limbs were closely observed, as well as the increase in the ulna axis and degree of osteotylus at the osteotomy region. X-ray scans were taken every two weeks. After the predetermined length was reached, the extension process was stopped, and X-ray scans were taken every 1~2 months. When the ulnar osteotomy ends healed, the patients were readmitted to the hospital so that the lengthening device could be removed under anaesthesia.

Statistical evaluation

SPSS 26.0 (IBM, Armonk, NY, USA) software and the R programming language (R Core Team 2018. Vienna, Austria) were used to perform all the statistical analyses. For both group A and group B, variables that were measured preoperatively and at the final follow-up, such as the US, RAA, CS, MEPS, and ranges of elbow flexion, forearm pronation and supination, were assessed by the Wilcoxon test. The improvements in each variable listed above were assessed by Spearman correlation analysis adjusted for variables such as age, sex, type of HME, follow-up time, and increase in the length of the ulna. The differences in complications between the two groups were assessed by two logistic regression models using the R programming language.

Results
All 37 patients were followed up. According to the type of external fixator the patients received, they were divided into two groups, and the basic information is shown in Table 1.

In group A, as shown in Table 2, the US, RAA, CS, elbow flexion, forearm pronation, supination, and MEPS values improved from preoperatively to postoperatively. All the changes were found to be significant by the Wilcoxon test ($p < 0.05$). According to Spearman correlation analysis, although changes in RAA and CS were associated with improved length of the ulna (IL) ($p < 0.05$), all the other improvements were not associated with age, sex, the type of HME, follow-up time, or IL ($p > 0.05$). Pin site infections were found in 3 patients, and all patients recovered after oral antibiotics. No other complications were recorded. A typical case is shown in Fig. 3(A-E).
Table 2
Clinical results of group A

| Subject                  | Preoperative | FFU      | Wilcoxon test | P Value of Spearman correlation analysis |
|--------------------------|--------------|----------|---------------|----------------------------------------|
|                          |              |          | Z    | P         | Age | Gender | Follow-up | Type | IL* |
| US(mm)                   | 23.15 ± 5.55 | 0.70 ± 2.49 | -4.546 | < 0.05 | 0.555 | 0.855 | 0.239 | 0.097 | Na  |
| RAA (*)                  | 32.30 ± 4.07 | 22.22 ± 2.28 | -4.550 | < 0.05 | 0.807 | 0.193 | 0.184 | 0.440 | < 0.05 |
| CS (%)                   | 70.37 ± 6.70 | 26.70 ± 3.42 | -4.547 | < 0.05 | 0.863 | 0.951 | 0.828 | 0.558 | < 0.05 |
| MEPS                     | 74.07 ± 3.68 | 94.26 ± 6.46 | -4.541 | < 0.05 | 0.367 | 0.643 | 0.080 | 0.698 | 0.946 |
| Elbow Flexion (*)        | 104.26 ± 4.94 | 130.44 ± 5.18 | -4.569 | < 0.05 | 0.263 | 0.975 | 0.625 | 0.460 | 0.380 |
| Forearm Pronation(*)    | 71.11 ± 3.76 | 83.52 ± 3.34 | -4.592 | < 0.05 | 0.233 | 0.403 | 0.099 | 0.730 | 0.308 |
| Forearm supination(*)   | 83.33 ± 3.67 | 84.07 ± 3.68 | -2.000 | < 0.05 | 0.384 | 0.320 | 0.790 | 0.670 | 0.973 |

Complications (n)

- Deviation of ulna axis: 0 case
- Poor regenerate bone formation: 0 case
- Neurovascular complications: 0 case
- Fracture after fixator removal: 0 case
- Pin site infection: 3 cases
- Redislocation of radial head: 0 case
- Recurrence of exostosis: 0 case

Abbreviations: US, ulnar shortening; RAA, radial articular angle; CS, carpal slip; MEPS, Mayo Elbow Performance Score; FFU: Final Follow-up; IL, Improved length; Na, not available.

*IL|=|US(Preoperative)-US(FFU)|

During the process of gradual extension of the ulna, the reduction of the radial head was satisfying. In group B, as shown in Table 3, the US, RAA, CS, elbow flexion, forearm pronation, supination, and MEPS values improved as well. According to the Wilcoxon test, all the improvements were found to be significant (p < 0.05), except for that of supination (p > 0.05). The improvements were not associated with age, sex, the type of HME, follow-up time, or IL (p > 0.05), according to Spearman correlation analysis. The reduction of the radial head was satisfying in the process of ulna lengthening, and no redislocation was reported. One patient presented with pin site infection and recovered after oral antibiotics. Deviation of the ulna axis (deviated more than one time of ulna diameter, or more than 15° of ulna axis) and poor bone formation were found in 4 patients, two typical cases are shown in Fig. 4(A-F). These cases were
resolved by removing the external fixator, inserting a bone block harvested from the autologous iliac crest, and inserting a locking compression plate (LCP) plate.

Table 3
Clinical results of group B

| Subject            | Preoperative | FFU     | Wilcoxon test | P Value of Spearman correlation analysis | Age | Gender | Follow-up | Type | IL* |
|--------------------|--------------|---------|---------------|----------------------------------------|-----|--------|-----------|------|-----|
| US (mm)            | 23.40 ± 4.84 | 0.70 ± 3.09 | -2.805 < 0.05 | 0.724 | 0.554 | 0.475 | 0.619 | Na |
| RAA (*)            | 31.30 ± 5.36 | 21.70 ± 3.40 | -2.805 < 0.05 | 0.337 | 0.921 | 0.772 | 0.525 | 0.242 |
| CS (%)             | 72.30 ± 4.35 | 28.30 ± 3.43 | -2.807 < 0.05 | 0.148 | 0.171 | 0.148 | 0.333 | 0.358 |
| MEPS               | 74.50 ± 3.69 | 96.00 ± 3.944 | -2.919 < 0.05 | 0.080 | 0.486 | 0.365 | 1.000 | 0.554 |
| Elbow Flexion (*)  | 106.50 ± 4.74 | 127.00 ± 2.58 | -2.850 < 0.05 | 0.581 | 0.076 | 0.462 | 0.949 | 0.361 |
| Forearm Pronation(*) | 69.00 ± 3.94 | 84.00 ± 3.16 | -2.836 < 0.05 | 0.064 | 0.606 | 0.868 | 0.251 | 0.598 |
| Forearm supination(*) | 83.00 ± 4.22 | 83.50 ± 4.12 | -1.000 | 0.317 | 0.122 | 0.447 | 0.174 | 0.214 | 0.122 |

Complications (n)

- Deviation of ulna axis: 4 case
- Poor regenerate bone formation: 4 case
- Neurovascular complications: 0 case
- Fracture after fixator removal: 0 case
- Pin site infection: 1 case
- Redislocation of radial head: 0 case
- Recurrence of exostosis: 0 case

Abbreviations: US, ulnar shortening; RAA, radial articular angle; CS, carpal slip; MEPS, Mayo Elbow Performance Score; FFU: Final Follow-up; IL, improved length; Na, not available.

*IL = |US(Preoperative) - US(FFU)|

P value < 0.05 Statistically significant

Differences in complications between the two groups were assessed by two logistic regression models using the R programming language (Table 4). For the deviation of the ulna axis, the difference between the two groups was only related to age (p < 0.05) and the type of external fixator (p < 0.05) but not to sex (p > 0.05), the follow-up time (p > 0.05),...
or the type of HME ($p > 0.05$). The positive estimate values suggested that ulna axis deviation was more likely to occur in older children with multi-joint external fixators.

### Table 4

| Subject                  | Logistic regression model 1* | Logistic regression model 2** |
|--------------------------|-----------------------------|-------------------------------|
|                          | Estimate value | $t$   | $P$   | Estimate value | $t$   | $P$   |
| Age                      | 0.044 ± 0.021 | 2.110 | < 0.05 | 0.048 ± 0.020 | 2.376 | < 0.05 |
| Gender                   | 0.083 ± 0.099 | 0.838 | 0.409  | -             | -     | -     |
| Follow-up                | -0.015 ± 0.031 | -0.483 | 0.632  | -             | -     | -     |
| Typella                  | -0.032 ± 0.106 | -0.305 | 0.763  | -             | -     | -     |
| Typellb                  | 0.179 ± 0.103 | 1.737 | 0.093  | -             | -     | -     |
| Type of external fixator | 0.415 ± 0.099 | 4.197 | < 0.05 | 0.360 ± 0.093 | 3.884 | < 0.05 |

Assignment: Type I, Type IIa, Type IIb: 0, 0; 1, 0; 0, 1; Gender: male: 1; female: 0; Type of external fixator:, Monorail: 0; Multi joint: 1.

*Logistic regression model with multiple variables included.

**Logical regression model after eliminating irrelevant variables on the basis of model 1.

### Discussion

Most forearm deformities in patients with HME are caused by ulna shortening\[^{2,18,19}\]. The distal ulna is affected by exostoses, which lead to dysplasia and shortening. Moreover, it tethers the ulnar side of the distal radius, hinders the growth of the distal radius, increases the RAA and CS, and weakens the support provided by the ulna to the wrist joint. On the other hand, the pressure of the radius increases, and with increasing age, the radius bends gradually, resulting in dislocation of the radial head\[^{20}\]. Therefore, treatments of forearm deformity caused by HME should primarily involve the early correction of ulnar shortening.

The aim of this study was to present the clinical results of 37 children who underwent surgical ulnar lengthening. In this study, two types of unilateral external fixations were selected: the monorail (Group A) and multi-joint fixators (Group B). Table 2 and Table 3 show that the US, RAA, CS, elbow flexion, forearm pronation, and MEPS values significantly improved in both group A and group B ($p < 0.05$). Only supination in group B did not significantly change, which might be attributed to the small sample size. The appearance and function of the upper limb significantly improved in the two groups, and the effect of unilateral external fixation on ulnar lengthening was obvious.

Some other methods of ulnar lengthening have been used. Some scholars \[^{2,25}\] have used bone grafting and steel plate fixation to treat ulnar shortening by 2 cm or less with satisfactory results. However, due to the bending of the radius and the influence of exostosis, the target length cannot be obtained in a single operation. Another common method is to use the Ilizarov ring\[^{4,6}\]. However, the circular external fixator has a complex structure and requires multi-needle and multi-plane fixation, which increases the risk of needle infection and neurovascular injury. In addition, in the process of lengthening, many parts of the Ilizarov ring need to be adjusted, which is not convenient for the parents of the patients. The Ilizarov ring is bulky, which affects daily activities and joint function exercise. In this study, two types of unilateral...
external fixations were selected. The operation was simple, the devices were light and comfortable to wear, and the potential of lengthening met the clinical needs.

The optimal timing of surgical intervention is controversial. One view $^{[2,9,21,22]}$ is that early surgery can slow or prevent the progression of deformities, especially the dislocation of the radial head, while for patients with dislocation, early surgery often leads to self-reduction. Another view $^{[11,23,24]}$ is that surgery should be postponed until the patient is 10 years old or at the age of epiphyseal closure. We believe that the timing of surgery should be determined on the basis of not only age but also the actual condition of the patients. For patients with obvious forearm and wrist deformities, a US value larger than 1.5 cm $^{[22]}$, radial head dislocation, enlargement of an exostosis, dysfunction, or chronic pain, the operation should be performed early. When the radial head has been dislocated for a long time, the morphology of the humerus and radius joint, annular ligament and other soft tissue structures may change, the failure rate of surgical reduction is high, and the function of the forearm may be poor. As shown in Fig. 3 (A-E), at the age of 3.5 years, a girl had good forearm appearance and function, without dislocation of the radial head. At the age of 6.5 years, the radial head was dislocated, the upper limb force line was obviously skewed, elbow flexion and forearm rotation were limited, and the operation was performed. At the three-year follow-up, the reduction of the radial head was satisfying, and the ulnar shortening deformity was corrected.

For complications, no fractures, neurovascular problems, or recurrences of exostoses were observed in the two groups. Pin site infections (5 cases in total) easily recovered. For radial head dislocation in patients with type IIa and type IIb deformities, no special procedures were performed during the operation, all cases self-reduced during ulna lengthening, and no cases of redislocation were found. This result suggests that it may be best not to treat radial head dislocation. The interosseous membrane can transmit the forces leading to lengthening, and the reduction of the radial head can be reached gradually$^{[26]}$.

Ulna deviations and poorly regenerated bone formation were more likely to occur in group B than in group A. The occurrence of these complications may be related to the position of the osteotomy and direction of the nail. It has been reported that$^{[12]}$ the diameter of the osteotomy site is negatively related to the time of bone healing. Some scholars$^{[10]}$ have suggested that the osteotomy point should be located at the maximum bending point, and the distance between the osteotomy point and the distal end of the ulna should be larger than 42% of the total length of the ulna. We suggest that the proximal part is selected, avoiding the maximum curvature as much as possible. Four Schanz pins were placed in the same plane in parallel to each other.

A patient with type IIb deformity is shown in Fig. 4 (A-C). The two groups of Schanz pins were located at the maximum bending plane of the ulna arch, and the nails were not positioned in parallel to each other or in the same plane. During the lengthening process, the joint of the external fixator became loose, and the dorsal angle of the ulnar arch gradually increased. Another patient with a type I deformity is shown in Fig. 4 (D-F). Although the Schanz pins avoided the maximum bending plane of the ulna arch, they were positioned in parallel to each other; however, they were not located in the same plane, so the force line was skewed, and the ends of the osteotomy region were separated during the extension process, leading to nonunion. Although the joints of the fixator were pressurized and locked, they were still loose during the extension process. The monorail external fixator should be used to effectively avoid this situation.

Whether exostoses should be removed is still debated. Akita$^{[11]}$ et al. found that exostosis resection can significantly improve the rotational function of the forearm but that it affects the US, RRA and CS values very little. We believe that the resection of exostoses can open the epiphysis of the distal ulna, enable the ulna to obtain a certain growth potential, reduce the effect of local tissue on the radius and correct deformities of the wrist. In this study, 37 patients underwent exostosis resection. The function of the forearm significantly improved.
Our study presents some limitations. This study is a retrospective study, with a small sample size and a short follow-up time. The clinical efficacy and complications remain to be further verified by a long-term prospective randomized controlled study.

Conclusions

Ulnar lengthening with unilateral external fixation is a safe and effective procedure for the treatment of HME. For complications, monorail fixators are more reliable than multi-joint fixators, as they can effectively avoid the occurrence of ulnar deviation.

Abbreviations

HME: Hereditary Multiple exostosis; US: ulna shortening; RAA: radial articular angle; CS: carpal slip; FFU: Final Follow-up; IL, Improved length.

Declarations

Ethics approval and consent to participate

This study was approved and supervised by the ethics committee of the Children's Hospital of Chongqing Medical University and was performed in accordance with the ethical standards of the 1964 Declaration of Helsinki. Informed written consent was obtained from all patients.

Consent to participate

Informed written consent was obtained from all patients.

Consent for publication

All the patient data used in this study were approved by the guardian, and the informed consent forms were signed.

Availability of data and material

The datasets analyzed in the study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

Funding

No funding was provided

Authors' contributions
Chao Zheng analyzed the data and wrote the paper. Huanli Han performed the statistical measurements. Yujiang Cao designed and evaluated the manuscript.

Acknowledgements

We thank all of the patients involved in the study. We thank the Department of Radiology of the Children's Hospital and Chongqing Children's Hospital for providing X-ray images.

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