Radiographic Criteria for Undergoing an Ulnar Shortening Osteotomy in Madelung Deformity: A Long-term Experience From a Single Institution

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Background: There are no established guidelines on the age or the severity of deformity for which an ulna shortening osteotomy or ulna epiphysiodesis should be performed in children and adolescents with Madelung deformity. The purpose of this study was to identify radiographic criteria associated with the eventual performance for an ulna shortening procedure in this patient population.

Methods: We retrospectively identified 41 wrists in 31 Madelung patients (mean ± SD age 13.8 ± 3.2 y) subjected to surgical correction of their deformity between 1999 and 2013. We assessed established radiographic criteria (ulnar tilt, lunate subsidence, palmar carpal displacement, ulnar variance) at preoperative and postoperative visits. Univariate and multivariate analyses were carried out to determine which radiographic criteria were associated with the performance of an "ulnar shortening procedure" at the first (index) surgical procedure.

Results: Eleven wrists were subjected to an ulna shortening osteotomy at the index and 5 at subsequent procedures; 10 cases received an ulnar epiphysiodesis (mean age 13.4 ± 1.5 y). Ulnar shortening at the index procedure was associated with significantly higher preoperative lunate subsidence, ulnar variance, and palmar carpal displacement. Ulnar variance of > 5 mm and lunate subsidence > 4 mm resulted in a respective 67% and a 53% likelihood of undergoing ulnar shortening osteotomy; palmar carpal displacement over 22 mm resulted in a 50% likelihood for ulnar shortening. Patients who required a subsequent procedure (n = 8) showed a significant increase in palmar displacement between surgeries. None of the 10 cases with a primary ulnar epiphysiodesis received a subsequent ulnar shortening; none of those undergoing late ulnar shortenings had an ulna epiphysiodesis at their index procedure (at 10.3 ± 4.3 y).

Conclusions: Lunate subsidence, ulnar variance, and palmar carpal displacement were significant radiographic criteria for undergoing an ulnar shortening osteotomy at our institution. A shortening osteotomy may be prevented by early ulna epiphysiodesis in skeletally immature children older than 10 years of age.

Level of Evidence: Therapeutic level IV—case series.

Key Words: Madelung deformity, ulna shortening osteotomy, ulnar variance

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Madelung deformity refers to a characteristic deformity of the distal radius resulting from an abnormality of the volar-ulnar portion of the distal radial physis.[1] In some cases, a thickened radiolunate ligament has been found, as described by Vickers.[2–4] The volar-radial growth disturbance may eventually lead to a progressive, 3-dimensional wrist deformity including radiocarpal and radioulnar joint malalignment, usually noted during adolescence.

In patients with pain and functional limitations, surgical correction of the radius has been advocated; the present standard of care involves a dome osteotomy of the distal radius.[5,6] This procedure improves radiocarpal and radioulnar joint alignment acutely, with good-to-excellent short-term clinical and radiographic results.[7] However, in more advanced cases, operative treatment of the radius alone may be insufficient as patients with severe or longstanding Madelung deformity will have marked volar-ulnar shortening of the radius compared with the longer adjacent ulna. Depending on the severity of the radial growth disturbance, forearm length is significantly reduced compared with normal controls.[8] In situations where the ulna is much longer, an ulnar shortening osteotomy has been advocated to "rebalance" the wrist and restore more normal radiographic alignment.[9] Although this procedure may be performed concomitant to the dome osteotomy of the radius, ulnar shortenings are often performed as a second-stage procedure in later

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adolescence. In addition, ulna and/or distal radius epiphysiodeses may be appropriate to reduce the threat of recurrent deformity in younger patients, although at the risk of an even shorter forearm.

Currently, there are no established guidelines on when an ulnar shortening osteotomy should be performed. Moreover, there are similarly limited data on the indications and results of ulnar epiphysiodeses in children and young adolescents with Madelung deformity. Hence, the purpose of this retrospective investigation was to review radiographs of all cases with Madelung deformity undergoing an ulnar shortening procedure (ulnar shortening osteotomy or distal ulna epiphysiodesis) over a period of 14 years. We aimed to correlate established radiographic criteria with the eventual performance of ulna shortening procedures in our cohort.

METHODS

After obtaining Institutional Review Board approval, we performed a retrospective chart review of patients undergoing operative treatment for Madelung deformity between 1999 and 2013 at a single tertiary referral center. Madelung patients were identified using an electronic institutional database by searching for the term "Madelung." Only cases with surgical treatment were retrieved for study. Final inclusion criteria in this study were children and adolescents younger than 18 years of age who received either an ulnar shortening osteotomy or an ulnar epiphysiodesis in addition to other concomitant procedures such as radius dome osteotomy.

The radiographic severity of Madelung deformity can be determined using predefined, validated criteria.[10,11] As a consequence, we measured the following radiographic imaging criteria of each of the 41 wrists in the 31 patients included in this analysis: (1) ulnar tilt, (2) lunate subsidence, (3) palmar carpal displacement, and (4) ulnar variance.[12] These criteria were assessed on anteroposterior (AP; ulnar tilt, lunate subsidence, ulnar variance; Figs. 1A, C, D) and lateral (palmar carpal displacement; Fig. 1B) wrist or forearm radiographs taken at multiple outpatient clinic visits. Given the fact that some patients traveled a long distance and underwent their first (index) surgery months after the first clinical consultation, we considered the most recent preoperative radiographs as the "baseline" whenever available. The median interval between baseline radiographs and the index procedure was 46 days. Postoperative radiographs and additional (preoperative/postoperative) radiographs of those who received subsequent surgery were also evaluated. The following demographic data were also collected: age at initial visit, age at surgical procedure(s), age at postoperative follow-up visit(s); bone age at the initial visit according to Greulich and Pyle[13]; sex, hand dominance, side of deformity, laterality (unilateral vs. bilateral), deformity type according to Zebala et al[8]; and other underlying or associated conditions.

Preoperatively, all patients had ulnocarpal wrist pain unresolved by rest, anti-inflammatory medications, splinting, and/or therapy. All had progressive, painful restriction of forearm rotation localized to their distal radioulnar and ulnocarpal joints. All had failed nonoperative management for a minimum of 6 months before surgical intervention. Most had documented progressive deformity with growth before surgical intervention. Radius dome osteotomy is almost always performed in primary cases to correct the radial tilt and inclination. The radial dome osteotomy will result in some improvement in ulnar variance as rotation of the distal radial epiphyseal segment will translate the lunate facet distally. Depending on the severity of the remaining radiographic ulnar (positive) variance and the age of the patient, either an ulnar shortening osteotomy alone, ulnar epiphysiodesis (if distal ulna physis is open), or both were performed at the first (index) procedure.

The electronic search retrieved 40 patients who were treated operatively for a Madelung deformity. Surgery was performed by either 1 of 2 fellowship-trained pediatric orthopaedic hand surgeons. However, after exclusion of cases with insufficient radiographic documentation, a total of 31 patients with 41 wrists were included in this study. All except 2 patients were female, and there were 20 left wrists affected. Twenty-two patients (71%) had bilateral Madelung deformity, although at present only 10 patients in this series received surgery on both sides. According to the criteria published by Zebala and colleagues, 35 wrists (85%) in 27 patients had distal radius involvement compared with 6 wrists (15%) in 4 patients with entire radius involvement. Eleven cases underwent ulnar shortening osteotomy at the index and 5 cases at a subsequent procedure (Table 1), for a total of 16 ulnar shortening osteotomies in this series of 41 wrists. Overall, ulnar epiphysiodesis was performed in 13 wrists; 9 patients (10 wrists) received the procedure at the index operation and 3 patients (3 wrists) at a subsequent procedure. The mean $\pm$ SD chronologic age at the time of the index procedure was 13.8 ± 3.2 years; the mean bone age at the baseline radiographic examination was 13.5 ± 2.9 years. A comprehensive overview of all patients is presented in online Appendix 1 (Supplemental Digital Content 1, http://links.lww.com/BPO/A33).

Statistical Analysis

Distributions were summarized as mean $\pm$ SD or percentage. Two-sample *t* tests were used to compare groups and paired *t* tests were used to test for within-person changes over time. These comparisons were repeated using nonparametric Wilcoxon tests and the results were very similar; thus, only the *t* tests are reported. The statistical assumption of independent observations was partially violated because of the subgroup of patients contributing data from both wrists. However, in a sensitivity analysis using generalized estimating equations to account for clustered data, we confirmed that the results and conclusions were substantively unchanged. Likelihood ratio tests from logistic regression were used for multivariate analysis of factors associated with ulna shortening osteotomy and also a composite

FIGURE 1. A–D, Radiographic characteristics of a Madelung case are presented. A, The AP radiograph shows pathognomonic findings such as an ulnar-tilted distal radius, a proximally migrated carpus, and relative hypertrophy of the distal ulna compared with the radius; note the radius radiolucency where the presence of a Vickers ligament is anticipated. The technique of measuring the "ulnar tilt" is shown. The "ulnar tilt" is the complement of the angle between the longitudinal ulna axis and a line proximal to the lunate and scaphoid. B, The lateral radiograph shows the volar radius tilt with a consecutive increase in the palmar carpal shift. This shift is defined by measuring the "palmar carpal displacement" between the longitudinal ulna axis and the most volar aspect of the lunate. C, An AP radiograph of another patient shows the "lunate subsidence." It is the distance between a perpendicular line to the longitudinal ulna axis and the proximal pole (most proximal point) of the lunate. D, The "ulnar variance" is often difficult to measure in Madelung deformity. After a line is placed along the longitudinal axis of the ulna, a perpendicular line at the most distal aspect of the articular surface of the ulna is drawn. Thereafter, the longitudinal distance from this perpendicular line to the most ulnar aspect the radial plateau is defined (C and D, Reprinted from Farr and Bae,[11] with permission from Elsevier).

ulna shortening and/or ulna epiphysiodesis procedure outcome. These analyses allowed us to test whether a factor added independent predictive information beyond the information provided by other factors in the model. P-values are 2-sided and considered statistically significant when <0.05.

RESULTS

Radiographic Baseline Results

The mean ulnar tilt at baseline was 40.0 ± 11.3 degrees, the mean lunate subsidence was 3.1 ± 4.4 mm, the mean palmar displacement was 18.4 ± 10.0 mm, and the mean ulnar variance was +3.4 ± 4.1 mm. However, patients who received an ulnar shortening at the index procedure showed significantly higher ulnar variance and lunate subsidence compared with those receiving ulnar shortening at a subsequent procedure (Table 1). After radius dome osteotomy with or without ulnar shortening osteotomy (n = 35), all radiographic parameters except ulnar variance decreased significantly after the index procedure. Considering cases with radius dome osteotomy and ulnar shortening osteotomy (n = 9), a significant improvement in ulnar tilt from 43.4 ± 8.5 to 26.3 ± 11.6 degrees, lunate subsidence from 8.1 ± 4.4 to 2.1 ± 4.1 mm, palmar carpal displacement from 25.1 ± 9.1 to 18.9 ± 7.2 mm, and ulnar variance from 8.2 ± 2.8 to 3.2 ± 4.1 mm was observed (Table 2).

In wrists that were subjected to a subsequent procedure (n = 8), there was a significant increase in palmar displacement ($P = 0.04$) between postoperative radiographs after the index procedure and radiographs before the subsequent procedure; ulnar tilt, lunate subsidence, and ulnar variance did not change significantly between the index and the subsequent procedures (Table 2). This significant increase in palmar displacement was also observed within the subgroup that received an ulnar shortening at a subsequent procedure (n = 5, $P = 0.03$). Interestingly, none of the 10 cases with a primary ulnar epiphysiodesis had persistent pain necessitating subsequent surgery. Moreover, none of the 5 cases undergoing a late ulnar shortening was subjected to an ulnar

TABLE 1. Age and Radiographic Measurements at the Index Surgical Procedure for Patients Who Were Subjected to an Ulna Shortening Osteotomy

	Timing of Ulna Shortening Osteotomy [Mean (± SD)]		
	At the Index Procedure (N = 11)	At a Subsequent Procedure (N = 5)	*P*
Age	15.5 (± 3.5)	10.3 (± 4.3)	0.02
Bone age	14.1 (± 2.2)	10.0 (± 5.1)	0.04
Ulnar tilt	40.0 (± 10.9)	36.0 (± 5.8)	0.46
Lunate subsidence	6.4 (± 5.4)	−0.5 (± 3.4)	0.02
Palmar displacement	23.9 (± 8.9)	14.0 (± 8.4)	0.06
Ulnar variance	6.8 (± 4.1)	0.1 (± 2.2)	0.004

TABLE 2. Changes in Radiographic Baseline Measurements Between the Surgical Procedures

	Change in Measurements Directly After Undergoing the Index Procedure				Change in Measurements Between Procedures (N = 8)	
	RD ± US (N = 34)*		RD + US (N = 8)*			
	Mean (± SD)	P†	Mean (± SD)	P†	Mean (± SD)	P†
Ulnar tilt	−15.1 (± 9.4)	< 0.001	−17.5 (± 12.5)	0.006	+1.9 (± 10.2)	0.62
Lunate subsidence	−2.3 (± 3.7)	0.001	−6.2 (± 3.4)	0.002	+1.9 (± 3.8)	0.20
Palmar displacement	−4.3 (± 8.2)	0.005	−6.6 (± 6.4)	0.02	+5.0 (± 5.5)	0.04
Ulnar variance	−1.3 (± 3.9)	0.07	−5.2 (± 4.9)	0.02	+1.0 (± 2.6)	0.30

*These sample sizes are one fewer than the actual number of cases because 1 case had no postindex procedure measurements.
†Paired *t* test.
RD indicates radius dome osteotomy; US, ulna shortening osteotomy.

epiphysiodesis at their index procedure performed at a mean age of 10.3 ± 4.3 years.

Ulnar Shortening and Ulnar Epiphysiodesis

Lunate subsidence and ulnar variance were both strongly associated with an ulnar shortening osteotomy at the index procedure in unadjusted analysis (lunate subsidence $P = 0.002$, ulnar variance $P = 0.001$; Table 3). However, they are strongly correlated with each other ($r = 0.83$) so that neither is significant when adjusted for each other ($P = 0.67$ for lunate subsidence, $P = 0.13$ for ulnar variance). Older age at the index procedure and greater palmar carpal displacement showed weaker but still significant associations with ulnar shortening in unadjusted analysis (Table 3). Hence, ulnar variance > 5 mm, lunate subsidence > 4 mm, and palmar carpal displacement > 22 mm resulted in 67%, 53%, and 50% likelihoods of ulnar shortening, respectively. In multivariate analysis, only lunate subsidence ($P = 0.04$) and ulnar variance ($P = 0.006$) remained significant after adjusting for age and palmar carpal displacement.

DISCUSSION

Various surgical techniques have been described to address the anatomic abnormalities found in Madelung deformity.[2,5,9,14–18] Surgical treatment is typically reserved for adolescents with pain, functional limitations, and/or progressive deformity failing nonoperative treatment and should be tailored to each affected wrist. Radius dome osteotomy is a useful procedure to reliably correct the increased radial tilt present in almost all cases.[7] However, Madelung deformity is a complex 3-dimensional condition and all structural components of the wrist should be considered to improve pain, motion, and esthetic appearance. Although some increased radial length may be gained using the dome osteotomy, larger discrepancies between the radius and the ulna may be addressed by ulnar shortening procedures, especially if ulnar-sided wrist pain is the main complaint.

Little information is available, however, to guide the surgeon on when to perform an ulna shortening osteotomy. To the best of our knowledge, there are no guidelines available that specify at which age and magnitude of relative ulnar overgrowth a shortening procedure is advised. Although a definitive ulnar shortening may be performed in late-presenting adult cases, indications for younger patients are more difficult to establish, given the potential remaining growth and risk for recurrent deformity.[19] It is believed that the adolescent growth spurt is a major factor influencing deformity progression as the majority of patients referred to tertiary care institutions are in their teenage years.

Our radiographic data suggest that patients who primarily underwent ulnar shortening were older, had worse deformity parameters with respect to ulnar length (variance/subsidence), and also showed significantly increased palmar carpal displacement (Fig. 1B). Most of the 11 ulnar shortening cases were performed for > 5 mm positive ulnar variance (8 cases, 73%), 4 mm lunate

TABLE 3. Baseline Characteristics Obtained at the Index Procedure

	Ulna Shortening Osteotomy			Any Ulna Shortening Procedure*			
Variables†	Yes (N = 11)	No (N = 30)	P	Yes (N = 19)	No (N = 22)	P	Ulna Epiphysiodesis (N = 8)
Age	15.5 (± 3.5)	13.2 (± 2.9)	0.04	14.6 (± 3.1)	13.2 (± 3.3)	0.17	13.2 (± 1.6)
Bone age	14.1 (± 2.2)	13.3 (± 3.2)	0.47	13.8 (± 2.0)	13.3 (± 3.6)	0.61	13.4 (± 1.7)
Ulnar tilt	40.0 (± 10.9)	40.1 (± 11.6)	0.99	38.8 (± 9.3)	41.1 (± 12.8)	0.51	37.1 (± 6.9)
Lunate subsidence	6.4 (± 5.4)	1.9 (± 3.3)	0.002	4.3 (± 5.0)	2.1 (± 3.6)	0.11	1.4 (± 2.6)
Palmar displacement	23.9 (± 8.9)	16.3 (± 9.8)	0.03	22.9 (± 7.2)	14.5 (± 10.6)	0.006	21.5 (± 3.9)
Ulnar variance	6.8 (± 4.1)	2.2 (± 3.3)	0.001	4.6 (± 4.3)	2.4 (± 3.6)	0.07	1.7 (± 2.5)

Data are presented independently for those who received an ulna shortening osteotomy or any ulna shortening procedure (osteotomy or epiphysiodesis) or epiphysiodesis alone at the index procedure.

*Patients who received any type of ulnar shortening (ulna shortening osteotomy and/or ulna epiphysiodesis) are considered.

†Age is age at the time of the procedure, and for other variables (including bone age) at the time of the baseline preoperative radiograph.

subsidence (9 cases, 82%), and 22 mm palmar displacement (9 cases, 82%), respectively (online Appendix 1, Supplemental Digital Content 1, http://links.lww.com/BPO/A33). The finding of increased palmar carpal displacement highlights the importance of evaluating both AP and lateral radiographs to characterize all elements of the deformity and thus optimize treatment. Furthermore, we observed that patients undergoing a secondary ulnar shortening showed a significant increase in palmar displacement, even after their index intervention. Hence, we hypothesize that not only the relatively long ulna (measured by variance/subsidence) but also the sagittal radioulnar incongruence are main contributors toward the development of pain and eventual performance of ulnar shortening at least in our cohort. These results are supported by the findings of Salon et al,[20] who emphasized the importance of a sufficient "coverage" of the lunate by the ulnar head in the lateral plane (> 80%) after ulna shortening.

The performance of subsequent surgery for recurrent deformity in cases without primary ulnar epiphysiodesis raises the question of when to perform a distal ulnar epiphysiodesis. Our results showed that none of the 8 wrists that were subjected to secondary procedures (5 of which were subjected to a subsequent ulna shortening) received ulnar epiphysiodesis at the index procedure. In contrast, none of the 10 wrists that had ulnar epiphysiodesis at the index procedure (performed at mean age 13.4 ± 1.5 y) required another intervention. The exact age to recommend this procedure remains to be defined. However, patients who underwent secondary ulnar shortenings had a significantly lower mean age at their index procedure compared with those receiving ulnar shortening directly at the index intervention (10.3 ± 4.3 vs. 15.5 ± 3.5 y, Table 1). This suggests that younger symptomatic patients with more growth remaining are more likely to require secondary surgery. On the basis of our experience, we believe that ulnar epiphysiodesis may be considered in skeletally immature children older than 10 years of age. This may be preventive of recurrent deformity, pain, and limited function that necessitate subsequent surgery.

As with many other deformities evolving during childhood and adolescence, a systematic treatment approach is preferred. Therefore, we suggest that patients with Madelung and refractory pain should first undergo surgical correction of their palmar and ulnar-tilted radius using the dome osteotomy technique. A soft-tissue release of the thickened radiolunate ligament (Vickers ligament) is performed at the same time. This procedure can reliably restore hand-forearm alignment with respect to radius inclination in the coronal plane, and palmar tilt in the sagittal plane. Depending on the relative length of the ulna and the age of the patient, further concomitant procedures may be warranted in the index operation, including (1) distal ulnar epiphysiodesis in younger patients (10 to 14 y) and/or (2) an ulnar shortening osteotomy in older patients (above 14 y) with increased ulnar variance and palmar displacement (> 5 and > 22 mm, respectively). As highlighted in Table 3, patients presenting with a lunate subsidence and/or ulnar variance of around 2 mm and/or a palmar carpal displacement of around 16 mm would most likely never undergo an ulnar shortening osteotomy at our tertiary referral center.

Although this study included a relatively large number of patients treated solely by pediatric hand surgeons with a standardized surgical technique, a number of limitations bear mention. Almost all radiographs were performed at a tertiary care pediatric hospital; however, subtle variations in technique and projection errors (particularly on lateral radiographs) cannot be completely ruled out. No reliable pain scores were collected, which could be correlated with the radiographic results. Therefore, the surgical recommendations proposed here are predominantly based on our radiographic observations, and furthermore, may not be applicable to the general population with Madelung deformity. However, it should be emphasized that surgery was indicated in all cases for refractory ulnar-sided wrist pain, which subjectively improved in our patients postoperatively; the 5 patients (8 wrists) who received a second intervention had recurrent pain with growth. Finally, long-term clinical and radiographic results are not available; additional investigations with longer follow-up are needed to characterize the durability of the radiographic correction reported.

On the basis of this radiographic study, lunate subsidence, ulnar variance, and palmar carpal displacement were strongly associated with the eventual necessity of an ulnar shortening osteotomy in Madelung deformity in our experience. Ulnar shortening osteotomy may be considered in symptomatic Madelung patients older than 14 years of age with > 5 mm of ulnar positive variance and 22 mm of palmar displacement after radial dome osteotomy. Furthermore, distal ulnar epiphysiodesis may be considered in patients between 10 and 14 years of age at the time of distal radial osteotomy to avoid secondary ulnar procedures.

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