Volar Ligament Release and Distal Radial Dome Osteotomy for the Correction of Madelung Deformity: Long-Term Follow-up

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Background: Madelung deformity is a disorder of growth of the distal aspect of the radius that is usually recognized in late adolescence near skeletal maturity. It results in a characteristic wrist deformity, decreased wrist motion, and wrist pain. The purpose of this study was to evaluate long-term results in patients treated by volar ligament release and distal radial dome osteotomy for Madelung deformity.

Methods: Patients who had undergone volar ligament release and dome osteotomy for Madelung deformity at our institution from 1990 to 2002 and who were the subjects of a previous report on this treatment were contacted for clinical and radiographic evaluation at mid-term to long-term follow-up. Forearm and wrist motion was evaluated. Posteroanterior and lateral radiographs of both forearms were assessed for radial inclination, lunate subsidence, and arthritis changes. A Disabilities of the Arm, Shoulder and Hand (DASH) survey was completed.

Results: Twenty-seven patients underwent volar ligament release and distal radial dome osteotomy. Eight patients were either lost to follow-up or were unable to return for follow-up. Nineteen patients with thirty-one operatively treated wrists were available for follow-up. After further review, eighteen patients and twenty-six wrists were included in the study. The average age at the time of follow-up was twenty-five years (range, nineteen to thirty-one years), with an average length of follow-up of eleven years (range, seven to fourteen years). There was no change in radial inclination or in wrist motion between the immediate postoperative and long-term follow-up evaluations. There was a positive correlation between the amount of deformity correction based on more severe preoperative parameters and an increased arthritic grade at the time of follow-up. There was positive correlation between an increased DASH score and arthritis grade as well as a correlation between whole bone deformity and increased arthritis grade and DASH score.

Conclusions: Volar ligament release and distal radial dome osteotomy for Madelung deformity provides lasting correction of the deformity. Long-term follow-up shows maintenance of original radiographic correction with good to excellent functional outcome. Patients with radiographic evidence of more severe disease preoperatively and the whole bone variety of Madelung deformity have poorer radiographic outcomes and trend toward poorer functional outcomes.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

Madelung deformity is an uncommon condition with characteristic bowing of the distal end of the radius, resulting in a prominence of the ulnar head and a classic appearance of volar subluxation of the carpus. The condition was first completely described in 1887 by Madelung[1]. It accounted for <2% of the pediatric hand deformities in the series described by Flatt[2]. In many patients, Madelung deformity is inherited in an autosomal dominant pattern[3,4]. When associated with Leri-Weil dyschondrosteosis, which is a form of dwarfism marked by short stature and short forearms, it is

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associated with a SHOX deficiency (short stature homeobox-containing gene)[5]. Patients usually present in late adolescence with characteristic wrist deformity, decreased wrist motion, and wrist pain. The disorder is characterized by abnormal growth at the distal radial physis and an abnormal ligamentous structure that tethers the proximal carpal row in relative flexion with respect to the articular surface of the distal end of the radius[6].

Many reports on procedures for the treatment of this deformity have described corrective osteotomies, but none have specifically noted a release of the volar ligament combined with corrective osteotomy at the site of the deformity. In 2006, Harley et al. reported on the early results of volar ligament release with distal radial dome osteotomy, a procedure directed toward removing the deforming force on the volar ulnar distal aspect of the radius and correction of deformity with a multiplanar osteotomy[7-9]. Patients had good pain relief with improved wrist motion and improved radiographic parameters. We report the mid-term to long-term results for these patients and examine whether the initial clinical and radiographic improvements were maintained.

Materials and Methods

A retrospective chart review was performed on the previously reported patients who underwent volar ligament release with dome osteotomy for the treatment of Madelung deformity at our institution from 1990 to 2002, with approval from the University of Texas Southwestern Medical Center institutional review board. Information gathered from the chart review included age at presentation, presenting symptoms, surgical procedures performed, preoperative and postoperative wrist and forearm motion, and preoperative and postoperative radiographic parameters, including radial inclination and lunate subsidence[7], as well as the primary location of the deformity in the radius[4]. Patients were invited to return for clinical and radiographic follow-up. Data collected included further surgical procedures performed, occupation, and whether any pain was present. Clinical evaluation assessed wrist motion, strength, and location of pain. Posteroanterior neutral and lateral radiographs of the forearms were measured for radial inclination and lunate subsidence, as described previously for this group of patients[7], as well as arthritis grade (grade 0 through 3), as described by Jupiter et al., at the radiocarpal joint[10]. Radiographs were also evaluated for whole bone involvement, defined by Zebala et al. as involvement of the radial diaphysis with bowing of ≥10° on the lateral radiograph or a radiocapitellar joint space of ≥4 mm (the distance between the radial head and the capitellum on anteroposterior and lateral radiographs)[4]. If the entire radius was deformed, the designation of "whole bone involvement" was given to a subset of patients. Each study patient also completed the DASH (Disabilities of the Arm, Shoulder and Hand) outcome questionnaire during his or her follow-up visit to provide validated outcome data on patient-reported disability. DASH subsets included function score (Q score), optional subset sports score (S score), and optional subset work score (W score). The S and W scores were optional as not all patients were employed or participated in sports.

Statistical Analysis

Individual and average clinical wrist motion and radiographic measurements made at the preoperative and postoperative evaluations, postoperative and long-term follow-up evaluations, and preoperative and long-term follow-up evaluations were compared with one-way analysis of variance (ANOVA) with a post hoc Tukey multiple comparison test. Changes in radiographic measurements were compared with a paired Student t test. The association between Q score (functional portion of the DASH score) and arthritis grade was compared with one-way ANOVA. Deformity location and arthritis score were compared with a chi-square test, with arthritis grade grouped by grades 0 to 1 or grade 2. Deformity location and Q score were compared using a chi-square test as well as deformity location and arthritis grade. The association between radial inclination and lunate subsidence with the arthritis score was estimated with the Spearman rho correlation coefficient. All tests were two-tailed with a p value of <0.05 considered significant.

Source of Funding

There was no external funding for this investigation.

Results

Twenty-seven patients underwent volar ligament release and distal radial dome osteotomy for Madelung deformity. Nineteen patients with thirty-one operatively treated wrists were available for follow-up. One patient with bilateral involvement was excluded because of an inadequate follow-up interval, and three patients who had surgery bilaterally with a follow-up interval of seven or more years for only one wrist had the other wrist excluded. Therefore, eighteen patients and twenty-six wrists with over seven years of follow-up were included in the study. There were two men and sixteen women, with fourteen right wrists and twelve left wrists. The average age at the time of follow-up was twenty-five years (range, nineteen to thirty-one years), with an average length of follow-up of eleven years (range, seven to fourteen years).

Six patients had additional reconstructive procedures after the index procedures. Ulnar shortening osteotomy, which was done in five patients, was the most common procedure. Of the five patients, one also underwent ulnocarpal ligament reconstruction, one had revision dome osteotomy, and one had a bilateral Darrach distal ulnar resection with ulnocarpal ligament reconstruction. One other patient underwent a Sauvé-Kapandji procedure (arthrodesis of the distal radioulnar joint with resection of a portion of the distal ulnar shaft). All of these revision procedures on the ulnar side of the wrist were in the first half of the series. In our current practice, if the distal ulnar physis is still open, an epiphysiodesis is done at the time of the index procedure. If the ulna remains too long and threatens impaction with the triquetrum, a shortening osteotomy is done at the same time. Fifteen patients reported being happy with the initial procedure, noting improvement with regard to motion, pain, and appearance postoperatively. This included the patient who had the Sauvé-Kapandji reconstruction. The three other patients did not report that the symptoms were worse after surgery, but they were ambivalent as to whether they had had any substantial improvement. Eight of the patients were employed in manual heavy labor. One patient was receiving Social Security disability benefits that were based on a claim that she could not work because of her wrists.

Clinical evaluation revealed that the patients had no significant loss in average wrist motion between the initial postoperative visit and the long-term follow-up evaluation. There was no significant difference in the average wrist extension or average forearm pronation between the initial postoperative evaluation and the long-term follow-up evaluation; however, significant improvements were detected in wrist flexion and supination (average, 11° and 14°, respectively; $p < 0.05$) (Table I).

TABLE I Average Wrist Motion

Parameter	Preop.*† *(deg)*	Initial Postop.* *(deg)*	Follow-up* *(deg)*	Comparison of Postop. and Follow-up Measurements *(p value)*
Wrist flexion	66 ± 18	58 ± 10	69 ± 14	0.01
Wrist extension	48 ± 17	56 ± 14	58 ± 17	0.97
Supination	50 ± 25	61 ± 34	75 ± 21	0.04
Pronation	71 ± 19	72 ± 21	70 ± 19	0.50

*The data are given as the mean and the standard deviation. †Preoperative data were reported in a previous study[7].

From the initial postoperative to long-term follow-up evaluations, eight wrists had a loss of extension (range, 5° to 15°), but two had improvement compared with the preoperative findings. Nine forearms had loss of pronation (range, 5° to 30°) between the postoperative and long-term follow-up examinations, with three that had improvement compared with the preoperative findings. Four wrists had loss of flexion (range, 10° to 15°) between the initial postoperative and the follow-up evaluations, but only two had lost motion compared with the preoperative findings. Only two forearms had loss of supination (10° and 15°) between the initial postoperative and long-term follow-up evaluations, but both had improvement compared with the preoperative findings. Of the patients who had lost motion, four had additional procedures.

Radiographic evaluation (Fig. 1) revealed that there was no significant difference between average initial postoperative and follow-up measurements of radial inclination ($p = 0.7$). There was a small, but significant, difference in average initial postoperative and follow-up measurements of lunate subsidence, with a mean loss of correction of 2 mm ($p < 0.05$) (Table II).

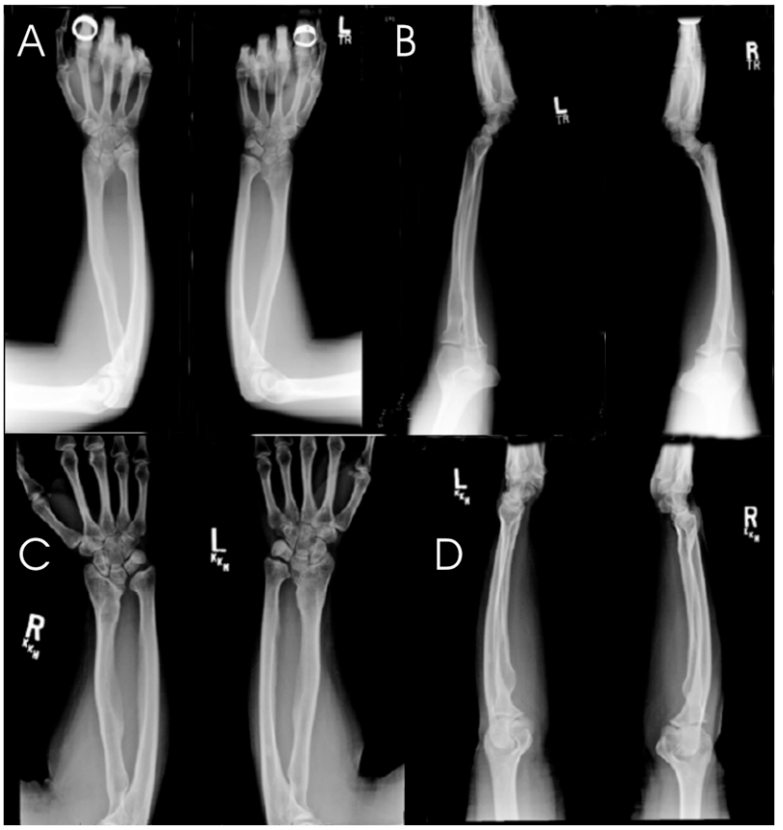

Fig. 1

Figs. 1-A through 1-D A sixteen-year-old girl with a four-year history of bilateral wrist pain. **Figs. 1-A and 1-B** Preoperative posteroanterior and lateral radiographs. **Figs. 1-C and 1-D** Postoperative posteroanterior and lateral radiographs made at the ten-year follow-up evaluation. The radiographs demonstrate improved radiographic parameters including improved alignment of the hand on the forearm. The procedure, however, does not address deformity of the whole arm.

TABLE II Average Radial Inclination and Lunate Subsidence

Parameter	Preop.*†	Postop.*‡	Follow-up*‡	Change Between Preop. and Postop.*§	Change Between Postop. and Follow-up*
Radial inclination *(deg)*	44 ± 11	32 ± 11	32 ± 9	12 ± 11#	0 ± 5
Lunate subsidence *(mm)*	8 ± 5	4 ± 4	6 ± 5	4 ± 5#	−2 ± 3#

*The values are given as the mean and the standard deviation. †Average age at the time of the preoperative measurement was twelve years (range, eight to sixteen years). ‡Average age at the time of the postoperative measurement was thirteen years (range, nine to seventeen years), with an average follow-up time of twenty-three months (range, seven to forty-nine months). §Average age at the time of the long-term follow-up was twenty-five years (range, nineteen to thirty-one years), with average follow-up time of eleven years (range, seven to fourteen years). #The difference was significant ($p < 0.05$).

Arthritis scores for the radiocarpal joint[10] at the time of follow-up showed two wrists with a grade of 0 (no arthritic changes), eleven wrists with Grade-1 changes (slight joint-space narrowing), and thirteen with Grade-2 changes (marked joint-space narrowing with osteophyte formation). None had Grade-3 changes, described as complete loss of cartilage and joint space with osteophyte and cyst formation. Increased preoperative deformity for both radial inclination and lunate subsidence parameters showed a significant association ($p < 0.05$) with increased arthritis score at the time of follow-up (Figs. 2-A and 2-B). On evaluation of the average change in radial inclination and lunate subsidence between the preoperative and the follow-up evaluations, there was a trend between increasing arthritis grade and increased correction when the two wrists with no arthritic change were excluded, but the difference was not significant. We were unable to identify a specific level of correction that could predict an increased arthritis grade.

Evaluation of the deformity on radiographs revealed that fifteen arms had whole bone involvement of the radial deformity and eleven of the wrists had only distal involvement of the radius, as described by Zebala et al.[4]. There was a significant association between whole bone deformity and increasing arthritis grade ($p < 0.05$) (see Appendix). There was also a positive relationship between whole bone deformity and increasing DASH Q score ($p < 0.05$) (Fig. 3).

The DASH survey yielded complete data for the functional portion of the survey (Q score) but incomplete data for the sports and work scores due to patient response (S and W scores). The Q scores of our patients were within norms, with the exception of three patients who had whole bone involvement. The average Q score was 12 for our nineteen patients, with only three patients who had a score of >24.7, which is outside normative data. Normative data for an adult are a mean Q score (and standard deviation) of 10.1 ± 14.6[11]. DASH scores

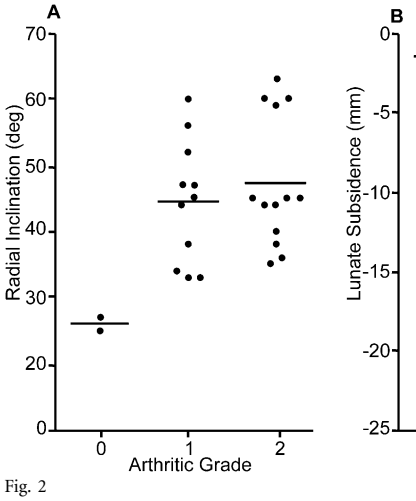

Fig. 2

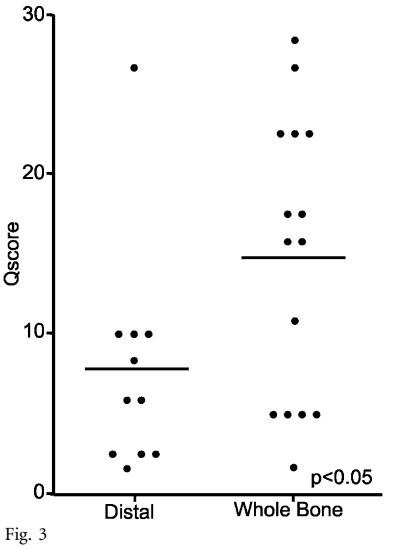

Fig. 3

Figs. 2-A and 2-B Graphs showing relationships between the deformity and the arthritis grade. **Fig. 2-A** A greater preoperative radial inclination is associated with a higher arthritis grade (with grade 0 indicating no arthritis and grade 2, advanced arthritis[10]) at the follow-up evaluation. **Fig. 2-B** Greater preoperative lunate subsidence is associated with a higher arthritis grade at the follow-up evaluation. The bars show the means, and the dots represent individual patient data. **Fig. 3** Correlation between increasing Q score (decreased function) and the location of the deformity (either distal or whole bone). Patients with a distal deformity had lower Q scores and higher function than patients did with whole bone deformity. The bars show the means, and the dots represent individual patient data.

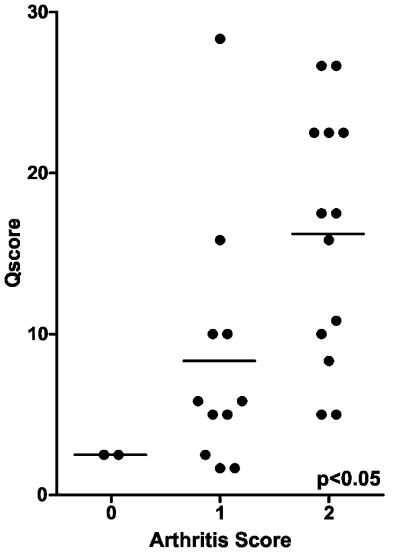

Fig. 4

A higher Q score (decreased function) is associated with increasing arthritis grade at the time of follow-up. The p value is one-way ANOVA. The bars show the means, and the dots represent individual patient data.

at the time of follow-up were evaluated with regard to the different radiographic parameters. The Q score (the functional portion of the DASH questionnaire) was positively correlated with arthritis grade but not radial inclination or lunate subsidence. The higher the average arthritis grade is, the greater the decrease in functional level (Fig. 4).

Discussion

The results of surgical treatment for Madelung deformity in the symptomatic young person should be compared with what is known about the natural history. However, we have information only about the symptomatic subset and anecdotal information about the rest. It is not known what the natural history of Madelung deformity is or whether there is a threshold of deformity that likely is associated with pain or symptoms at middle to late adulthood. Fagg[3] reported that he treated nineteen adult patients in their forties with Madelung deformity for wrist pain. Two patients had surgery for ulnar-sided wrist pain, but the other patients did not require surgical intervention; outcomes were not reported.

Schmidt-Rohlfing et al.[12], in a study of thirty-one patients with Madelung deformity, reported that five patients (16%) had surgery because of chronic pain. Ninety percent of their patients over sixteen years old were employed, but 42% reported disadvantages when working and 32% reported that their deformity had influenced their choice of employment.

Nielsen[13] reported twenty-six patients between six and sixteen years old with the diagnosis of Madelung deformity who were seen because of pain and loss of motion. Half of the patients had surgical intervention because of continued pain at the age of fourteen to fifty-seven years.

Overall, the majority of patients with Madelung deformity in these series did not require surgical intervention, and the pain resolved in many after adolescence. This has led to the recommendation of waiting until skeletal maturity to see whether the symptoms resolve prior to undertaking surgical intervention[12,13]. As reported by Schmidt-Rohlfing et al., however, many of the patients had some disability.

Many different procedures have been described to treat symptomatic Madelung deformity, including multiple variations on wedge osteotomies of the radius with or without shortening of the ulna[14-18], primarily procedures on the ulnar side of the wrist such as Darrach procedures and ulnar shortening osteotomies[13,19,20], correction utilizing Ilizarov techniques[21], and distal radial physiolysis[6]. Of all of these, the only procedure that addresses the site of the deformity is distal radial physiolysis as described by Vickers and Nielsen[6]. Physiolysis is reserved for patients identified early, with growth potential remaining, and with a discrete area of the growth plate that can be removed without compromising support for the lunate. Most patients with symptomatic Madelung deformity present after skeletal maturity and are not candidates for the physeal release. If there is no potential for remaining growth to correct the deformity after physeal release, then deformity correction by osteotomy is appropriate. Since the distal end of the radius is deformed in three planes, we selected a dome osteotomy to improve triplanar orientation of the distal radial articular surface.

Few studies have large numbers of patients for long-term follow-up of the different surgical interventions[6,16,19]. The main reasons for surgery included ulnar-sided wrist pain, loss of motion, and clinical appearance. Surgical intervention provided improvement with regard to pain and motion, but there is little information on functional outcomes.

In our series, we combined the approaches of release of the volar ligament with a corrective distal radial osteotomy that allows multiplanar correction of the distal radial deformity. Release of the volar ligament allows the proximal carpal row to assume a more normal position in relationship to the distal end of the radius. The distal radial osteotomy allows realignment of the articular surface in all three planes and addresses all components of the deformity. Measurement of the three-dimensional deformity in this condition on two-dimensional radiographs is difficult because of the inability to visualize consistent osseous landmarks. McCarroll et al. described a method and nomenclature for quantifying the deformity[22-24]. We used a simplified system of radial inclination and lunate subsidence that we find useful in this condition, as reported in our short-term follow-up study of our patients[7]. To be able to compare our results, we chose to continue with this form of radiographic measurement. At the time of short-term follow-up, the procedure provided good correction of the deformity with resolution of pain and improved wrist motion[7]. In the present study, we showed that, at the time of the long-term follow-up, the deformity had not recurred and improvements in motion were maintained.

We report on patient outcome with a validated outcome measure, the DASH, which none of the previous studies addressed. For the general population, the mean DASH Q score (and standard deviation) is 10.1 ± 14.6[11]. In our study, only three of nineteen patients had a Q score of >24 (outside normative

data), with a highest score of 28. The majority of our patients reported a functional outcome equivalent with normative data.

We found a correlation between an increased DASH score and arthritis grade as well as between an increased amount of correction based on radiographic parameters and arthritis grade. It is possible that the procedure itself could be the cause of the increasing arthritis grades. Larger corrections may produce increased forces across the wrist, leading to arthritic changes. However, wrists with increased radial inclination and lunate subsidence preoperatively had increased arthritis grades at the time of follow-up. Our data showed no significant correlation between initial deformity and the amount of correction achieved at the time of follow-up.

In our series, patients with whole bone deformity had worse radiographic outcomes and a trend toward more abnormal DASH scores and clinical outcomes. We agree that it is important to identify patients with whole bone deformity because they are more likely to have increased radiocarpal arthritis grade on radiographs, and they may be more likely to have more abnormal DASH scores than do those with isolated distal deformity. Zebala et al.[4] reported that there is not one single deformity in Madelung deformity but a spectrum of deformity with a milder group presenting with distal radial deformity and another more severe group having involvement of the entire radius, or whole bone deformity. They found that the overall deformity was more severe in the patients with whole bone deformity including shorter stature, worse radiographic parameters, and decreased wrist motion. As discussed by Zebala et al., the surgical procedures only address the distal end of the radius and therefore may be less effective for the whole bone deformities.

There are several weaknesses with the current study. As with the other reports of Madelung deformity, we had a small patient population and had several patients lost to follow-up. Although the length of follow-up is longer than that in most other reports, it is still relatively short. The major weakness, however, is that we did not know whether our intervention was affecting the natural history of the disease. We showed that the patients with more severe disease preoperatively did worse radiographically and showed a trend toward worse function at the time of follow-up, even with surgical intervention. These patients may have had even worse outcomes without intervention; however, without a control group, we cannot know. It is, and will be, impossible to know with certainty which patients with Madelung deformity will develop symptoms, whose symptoms will persist, and whether the intervention that is done in adolescence or early adulthood can influence the long-term function and comfort of this group of patients. It is important to follow the patients who have had surgical intervention to ascertain that the intervention has not led to a worsening of symptoms or deformity over the long term. Our findings with regard to worse outcomes for more severe disease and whole bone deformity may be helpful in preoperative discussion and counseling for patient expectations.

In conclusion, volar ligament release with distal radial dome osteotomy provides lasting correction of Madelung deformity at intermediate to long-term follow-up.

Appendix

eA A figure showing the correlation between deformity location and arthritis score is available with the online version of this article as a data supplement at jbjs.org. ■

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