

Prediction of Radial Head Subluxation and Dislocation in Patients with Multiple Hereditary Exostoses

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Background: Multiple hereditary exostoses (MHE) is a rare bone disease that results in growth of benign cartilage-capped tumors and a number of skeletal deformities. Forearm deformities are present in up to 60% of patients with MHE, and radial head subluxation or dislocation occurs in 20% to 30%. Radial head subluxation/dislocation results in a shortened forearm and loss of motion. The purpose of this study was to identify radiographic variables that are most predictive of radial head subluxation/dislocation in an effort to determine the need for prophylactic treatment.

Methods: We retrospectively reviewed the cases of consecutive patients with MHE treated in our center between April 2007 and December 2019. Radiographic measurements included the presence or absence of distal ulnar osteochondromas, total ulnar bow, total radial bow, and percent ulnar length. Participants were separated into 3 groups based on the status of the radial head: located, subluxated, and dislocated. Radiographic measurements were compared using a Kruskal-Wallis H test with Dunn post-hoc analysis. A prediction model was run using a binomial logistic regression, and a prediction matrix was created.

Results: A total of 88 patients were included in the study. There were significant differences in the located group compared with the dislocated group in terms of pronation, supination, and extension. The percent ulnar length, total ulnar bow, and total radial bow differed significantly between the located and dislocated groups ($p < 0.0001$); however, in the binomial regression analysis, only the percent ulnar length and total ulnar bow could be used to distinguish between the located group and the subluxated/dislocated group. Both of these measurements were significant predictors of subluxation/dislocation. There was no radial head subluxation/dislocation in patients with an ulnar bow of $<17^\circ$.

Conclusions: The data indicate that total ulnar bow and percent ulnar length are good predictors of radial head subluxation/dislocation. These 2 parameters can be utilized to monitor forearm deformity and guide timing for prophylactic treatment.

Level of Evidence: Prognostic Level III. See Instructions for Authors for a complete description of levels of evidence.

Multiple hereditary exostoses (MHE) is an autosomal-dominant gene disorder that affects 1 in 50,000 births¹. This condition results in the growth of benign cartilage-capped tumors that most commonly originate from the growth plates of long bones. Deformities are common in patients with MHE, including subluxation or dislocation of the radiocapitellar joint with a foreshortened forearm¹⁻⁴.

The prevalence of forearm deformities has been reported to be as high as 60% in patients with MHE². If these deformities are left untreated, they may result in radial head subluxation/dislocation (Fig. 1). Late management, after dislocation, often

does not restore function, particularly supination and pronation⁵. If radial head dislocation is prevented, then the loss of function can be mitigated. Determining which patients are at risk for radial head subluxation/dislocation is important because it gives surgeons an opportunity to intervene before it occurs.

Previous studies have identified osteochondroma location/number, ulnar length, and radial bow as risk factors for radiocapitellar dislocation^{3,6,7}. Another condition that results in radial head dislocation is a Monteggia fracture. Ulnar malunion has been linked to chronic radial head dislocation in patients with a Monteggia fracture⁸. Ulnar deformity has been suggested

Disclosure: The **Disclosure of Potential Conflicts of Interest** forms are provided with the online version of the article (<http://links.lww.com/JBJS/G668>).

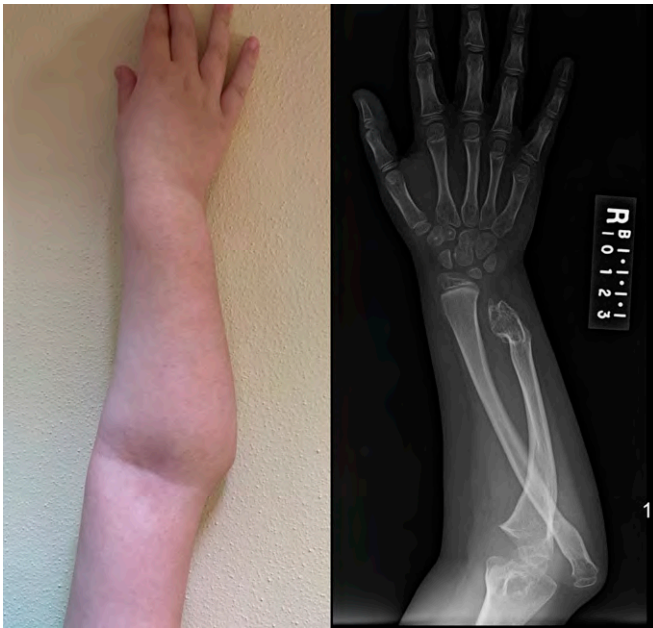


Fig. 1
Example of a radiograph and clinical presentation of a dislocated forearm in a patient with multiple hereditary exostoses.

to be a contributing factor to radiocapitellar dislocation in patients with MHE; however, we are not aware of any published studies providing evidence for this suggestion⁹.

The purpose of this study was to determine what radiographic measurements may be risk factors for radial head subluxation/dislocation. We hypothesized that ulnar bow is a significant risk factor for this pathological condition.

Materials and Methods

Institutional review board approval was obtained for this study. A retrospective chart review was performed for all patients with MHE treated in our clinic between April 2007 and December 2019. A total of 248 patients were seen. They were included in the study if they had anteroposterior and lateral radiographs of the radius and ulna. Patients were excluded from the study if the 2 radiographs of the forearm were not orthogonal. One arm was included for each patient to eliminate the effects of correlated data within groups. For patients in the located group, the side was selected randomly and the most recent radiograph was chosen unless they had had surgery, in which case the last radiograph prior to the surgery was used. For patients in the subluxated or dislocated group, the subluxated or dislocated side was used for analysis and the first radiograph displaying subluxation/dislocation was used. Basic demographics and range-of-motion data were collected from the charts. Range of motion included supination, pronation, elbow flexion, and elbow extension. Measurements were performed on the radiographs and included percent ulnar length, total radial bow, total ulnar bow, the presence of distal ulnar osteochondromas, and the status of the radiocapitellar joint (located, subluxated, or dislocated).

The calculation to determine percent ulnar length was done with a method that was similar to the one described by Jo et al.⁶: the length of the ulna from the tip of the styloid to the edge of the olecranon was divided by the length of the radius measured from the center of the proximal physis to the center of the distal physis, and the quotient was multiplied by 100 to give a percentage. Length measurements were obtained from the anteroposterior radiographs. The total ulnar bow and total radial bow were determined by measuring the angle of the proximal and distal mid-diaphyseal lines of the respective bones on orthogonal radiographs¹⁰. The Pythagorean theorem was applied to determine the true oblique-plane deformity. Figure 2 shows examples of how these measurements were performed. Some of the ulnae analyzed had a distal-third deformity (Fig. 3), and for these ulnae the distal mid-diaphyseal line was started just proximal to the apex of this distal-third bow. Additionally, the presence or absence of distal ulnar osteochondromas was recorded.

Intraclass correlation coefficients (ICCs) were calculated for a subset of the bow measurements done by an attending surgeon and a clinical scientist, which resulted in a good reliability with an ICC of 0.77.

The forearms were separated into 3 groups depending on the status of the radial head: located, subluxated, and dislocated.

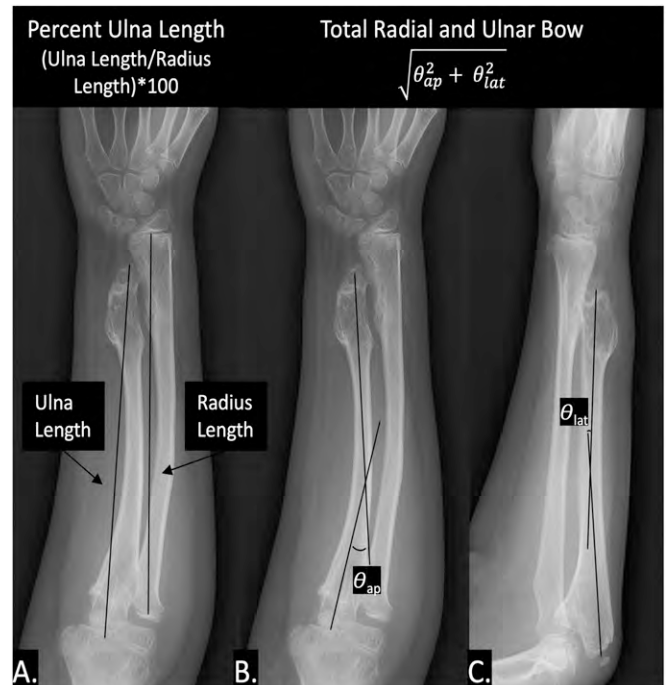


Fig. 2
Radiographic measurements. **Fig. 2-A** The percent ulnar length is the ratio of the ulna and radial lengths, providing a normalized ulnar length value. **Fig. 2-B** Radial and ulnar bow measured on the anteroposterior radiograph. **Fig. 2-C** Radial and ulnar bow measured on the lateral radiograph. The total ulnar bow and total radial bow were calculated by using the Pythagorean theorem.



Fig. 3
Radiographic measurements when a distal-third bow is present. **Fig. 3-A** The coronal ulnar bow when the distal-third segment is used. **Fig. 3-B** The coronal ulnar bow when the distal mid-diaphyseal line is moved proximal to the distal-third apex. The bow in Fig. 3-A is higher than the bow in Fig. 3-B; however, the distal-third bow does not contribute to stability of the radiocapitellar joint. In this study, the measurement in Fig. 3-B was utilized for all arms with a distal-third bow.

Subluxation and dislocation were determined either with intraoperative arthrography or from radiographs using the methods outlined by Souder et al.¹¹. The range of motion and radiographic measurements were compared among groups using a Kruskal-Wallis H test to determine differences in group distributions. Post-hoc analysis was performed using the Dunn test for multiple comparisons. All post-hoc reported p values were adjusted for multiplicity. Pearson correlation coefficients were determined for the radiographic measurements.

A binomial logistic regression analysis was performed to predict radial head subluxation/dislocation. Binomial regres-

sion analysis uses any number of independent variables and creates a probability function that can be used to predict a binary outcome. The subluxation and dislocation groups were combined to create 1 subluxation/dislocation group because it is paramount to prevent both of these pathological conditions. The regression analysis was initially performed with percent ulnar length, total ulnar bow, total radial bow, and presence/absence of distal ulnar osteochondromas to determine which ones were significant risk factors. The significant risk factors were used to create the final probability function and were then analyzed across a range of values. The probability of radial head subluxation/dislocation was determined for each value of total ulnar bow and percent ulnar length. A matrix was created that shows the predicted status of the radiocapitellar joint (located versus subluxation/dislocated) in relation to the significant variables analyzed.

Significance for all tests was set at $\alpha = 0.05$. The binomial regression analysis was performed with R (R Core Team 2020; The R Project for Statistical Computing, www.R-project.org/) and the other statistical analyses were performed using GraphPad Prism, version 8.4.2 for MacOS (www.graphpad.com).

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Results

A total of 88 patients, 52 males and 36 females with a median age at the time of radiographic examination of 10.0 years (range = 2.5 to 43.8 years) were included in this study. The patients were divided into 3 groups: located ($n = 70$; 80%), subluxated ($n = 10$; 11%), and dislocated ($n = 8$; 9%).

Range-of-motion measurements were available for 63 forearms and were compared among the groups using a Kruskal-Wallis H test with Dunn post-hoc analysis (Fig. 4, Table I). There were significant differences between groups for supination and pronation (Figs. 4-A and 4-B). Post-hoc analysis revealed that supination was greater in the located group (median = 89°; range = 0° to 96°) compared with the dislocated group (median = 38°; range = 0° to 87°) but not compared with the subluxated group (median = 53°; range = 0° to 93°). Pronation was greater in the located group (median = 82°; range = 10° to 100°) compared with the subluxated group (median = 57°; range = 0° to 85°) and the dislocated group (median = 50°; range = 15° to 80°). There was no difference between the subluxated and dislocated groups for either supination or pronation. Flexion did not differ significantly among the groups (Fig. 4-C). Post-hoc analysis revealed significantly less extension (a lack of full extension) in the dislocated group (median = -8°; range = -35° to 0°) compared with the located group (median = 0°; range = -8° to 14°) but no difference between the subluxated group (median = 0°; range = -30° to 10°) and either the located or the dislocated group (Fig. 4-D).

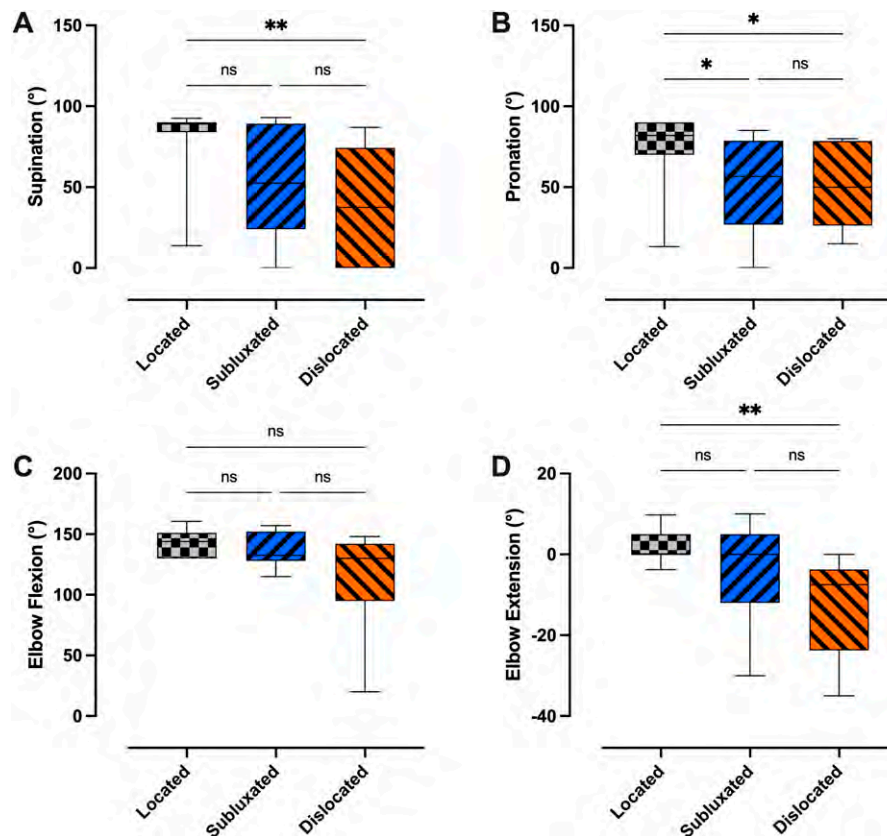


Fig. 4

Box and whisker plots showing the median, interquartile range, and 5th and 95th percentiles for the range-of-motion measurements. *P < 0.05, **p < 0.01, and ns = not significant.

Radiographic measurements were compared among the groups using a Kruskal-Wallis H test with Dunn post-hoc analysis (Fig. 5, Table I). There were significant differences between groups for percent ulnar length (Fig. 5-A). Post-hoc analysis revealed that percent ulnar length was greater in the

located group (median = 109%; range = 98% to 119%) compared with the subluxated group (median = 98%; range = 92% to 104%) and the dislocated group (median = 92%; range = 72% to 100%). There was no significant difference between the subluxated and dislocated groups for percent ulnar length.

TABLE I Median Values (Range) for Radiographic and Range-of-Motion Parameters

Parameter	Group			Chi-Square Test Statistic	Multiplicity-Adjusted P Value		
	Located (N = 70/47*)	Subluxated (N = 10/10*)	Dislocated (N = 8/6*)		Located vs. Subluxated	Subluxated vs. Dislocated	Located vs. Dislocated
Radiographic							
Percent ulnar length	109 (98-119)	98 (92-104)	92 (72-100)	38.89	<0.0001	>0.9999	<0.0001
Total ulnar bow	13° (4°-25°)	22° (17°-30°)	22° (19°-28°)	37.83	<0.0001	>0.9999	<0.0001
Total radial bow	15° (7°-41°)†	17° (13°-30°)	28° (21°-47°)	20.36	0.1162	0.1932	<0.0001
Range of motion							
Supination	89° (0°-96°)	53° (0°-93°)	38° (0°-87°)	13.19	0.0666	0.8492	0.0055
Pronation	82° (10°-100°)	57° (0°-85°)	50° (15°-80°)	13.10	0.0105	>0.9999	0.0344
Flexion	144° (130°-162°)	133° (115°-157°)	130° (20°-148°)	5.244	>0.9999	0.6139	0.0798
Extension	0° (-8°-14°)	0° (-30°-10°)	-8° (-35°-0°)	13.78	0.2901	0.1992	0.0013

*The N values are for radiographic/range-of-motion measurements. †N = 69.

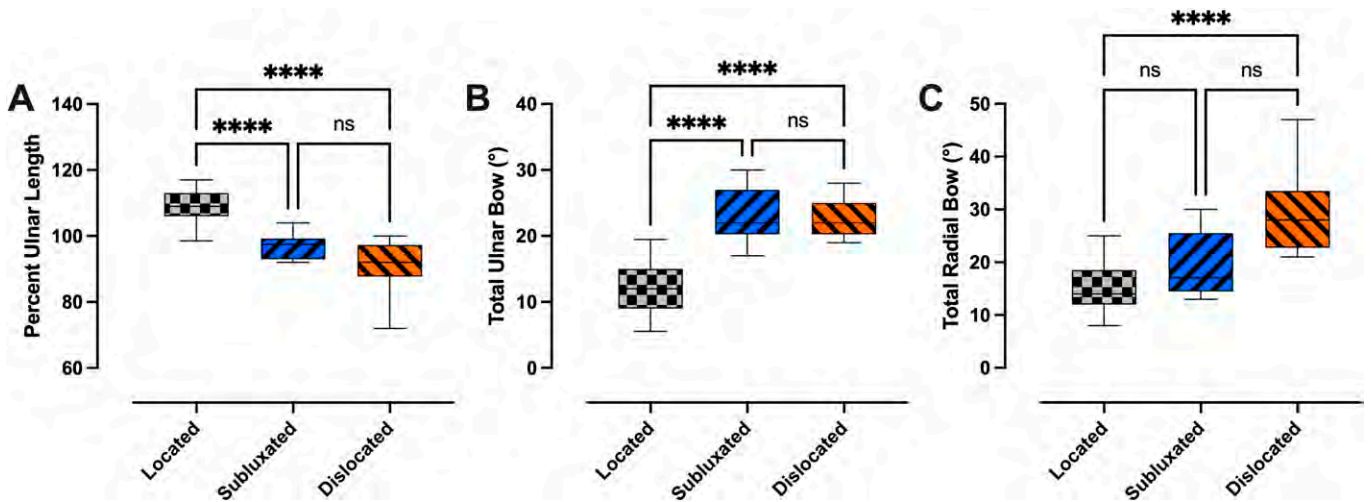


Fig. 5
Box and whisker plots showing the median, interquartile range, and 5th and 95th percentiles for the radiographic measurements. ****P < 0.0001 and ns = not significant.

There were significant differences between groups for total ulnar bow (Fig. 5-B). Post-hoc analysis revealed that total ulnar bow was less in the located group (median = 13°; range = 4° to 25°) compared with the subluxated group (median = 22°; range = 17° to 30°) and the dislocated group (median = 22°; range = 19° to 28°). There was no difference between the subluxated and dislocated groups. Post-hoc analysis revealed that total radial bow was significantly greater in the dislocated

group (median = 28°; range = 21° to 47°) compared with the located group (median = 15°; range = 7° to 41°) but did not differ between the subluxated group (median = 17°; range = 13° to 30°) and either the dislocated or the located group (Fig. 5-C).

The 3 radiographic parameters showed significant correlations with one another, with varying strengths (Fig. 6). The percent ulnar length had a moderate to strong negative correlation with total ulnar bow and total radial bow. The ulnar

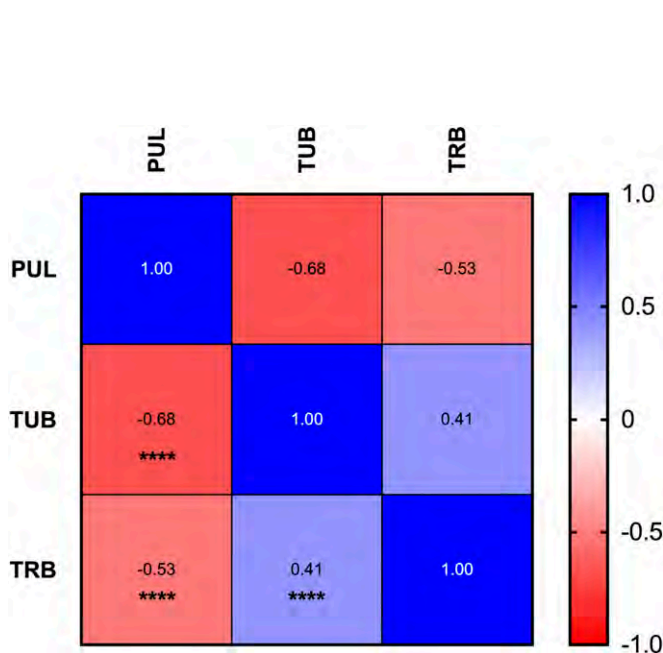


Fig. 6
Fig. 6 Correlation matrix containing r values for the radiographic parameters. There are several significant correlations, ranging from moderate to strong. Very weak correlation, r = 0.00 to 0.19; weak correlation, r = 0.20 to 0.39; moderate correlation, r = 0.40 to 0.59; strong correlation, r = 0.60 to 0.79; and very strong correlation, r = 0.80 to 1.00. Negative values indicate a negative correlation and positive values indicate a positive correlation. ****P < 0.0001, PUL = percent ulnar length, TUB = total ulnar bow, and TRB = total radial bow. **Fig. 7** Scatterplot of percent ulnar length versus total ulnar bow.

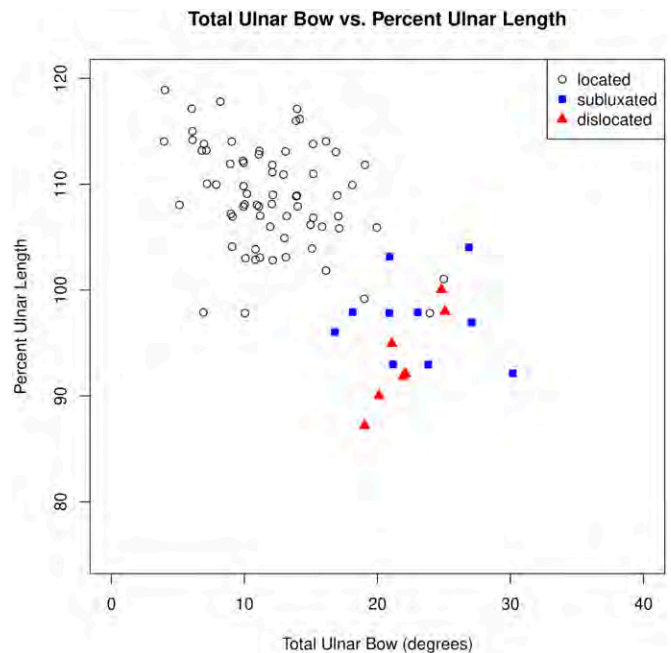


Fig. 7
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TABLE II Binomial Regression Model Used to Predict Radial Head Subluxation/Dislocation

	Coefficient	Std. Error	Z Value	P Value
Initial model with all variables*				
Percent ulnar length	-0.467	0.2165	-2.157	0.031
Total ulnar bow	0.337	0.1460	2.309	0.021
Total radial bow	0.094	0.1199	0.785	0.432
Distal ulnar osteochondroma	13.21	3,171	0.004	0.997
Final model with only significant predictors*				
Percent ulnar length	-0.471	0.1869	-2.518	0.012
Total ulnar bow	0.333	0.1387	2.402	0.016

*The initial model was created with all variables, and the final model was created with only the significant predictors.

and radial bows additionally had a moderate positive correlation with each other.

Figure 7 illustrates the relationship between total ulnar bow and percent ulnar length among all 3 groups. The located group tended to have long straight ulnae, while the subluxated and dislocated groups had both increased total ulnar bow and decreased percent ulnar length. No arms with a straight ulna ($<17^\circ$ of total ulnar bow) experienced radial head subluxation/dislocation. Additionally, no arms with a long ulna ($>104\%$ ulnar length) experienced radial head subluxation/dislocation.

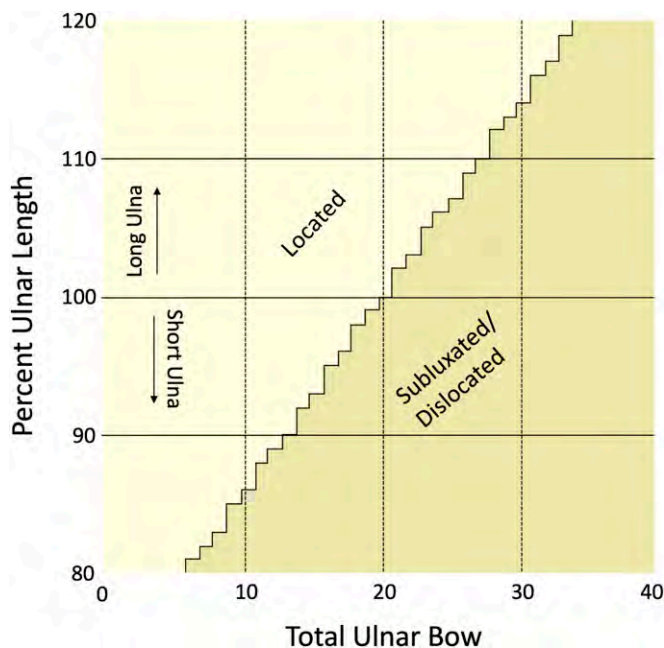


Fig. 8
Prediction matrix for subluxation and dislocation. The prediction model was run for each value of total ulnar bow between 0° and 40° and percent ulnar length between 80% and 120%. This allows a clinician to determine the risk of subluxation/dislocation according to where the values for these parameters fall on this matrix.

A binomial regression model was developed to predict radial head subluxation/dislocation (Table II). The radial head dislocation and subluxation groups were combined to form a single cohort for the regression analysis because prophylactic treatment would be based on preventing subluxation and dislocation. The initial model was run with percent ulnar length, total ulnar bow, total radial bow, and the presence of distal ulnar osteochondromas; however, total radial bow and the presence of distal ulnar osteochondromas were not significant risk factors and thus were removed. The final model was built using just percent ulnar length and total ulnar bow since both were significant risk factors for radial head subluxation/dislocation. The model was then used to predict radial head status across a range of percent ulnar length and total ulnar bow values, resulting in a prediction matrix (Fig. 8).

The prediction matrix using total ulnar bow and percent ulnar length to predict whether the radial head is located or subluxated/dislocated was able to correctly identify 97% of the cases in our series. The model highlighted a couple of interesting relationships. First, we found cases in which the ulna was longer than the radius. This seemed to impart a protective mechanism on the radiocapitellar joint. Second, the model predicted that, with an ulna of the same length as the radius (percent ulnar length = 100), total ulnar bow of $>20^\circ$ will lead to radial head subluxation/dislocation. Finally, as percent ulnar length decreases, the amount of the total ulnar bow tolerated before radial head subluxation/dislocation occurs also decreases; however, no radiocapitellar joints were subluxated or dislocated in patients with a total ulnar bow of $<17^\circ$ in our series. This model can be used as a tool to indicate which patients would benefit from prophylactic treatment to prevent radial head subluxation/dislocation.

Discussion

MHE is a rare bone disease with a variety of upper-limb deformities including radiocapitellar instability¹. Radiocapitellar dislocation has been associated with loss of function and motion^{12,13}. In our study, individuals who had a dislocated or subluxated radial head demonstrated impaired range of motion. Supination and pronation were reduced in

the dislocated group compared with the located group. A lack of full elbow extension was also identified in the dislocated group, which had significantly less elbow extension compared with the located group. Elbow flexion did not differ among groups. Additionally, pronation was reduced in the subluxated group when compared with the located group. These findings are important as they demonstrate that loss of motion occurs in elbows that are not dislocated, and therefore prophylactic treatment should be provided before they reach the subluxated stage. Furthermore, these findings support the decision to combine the subluxated and dislocated groups for regression analysis.

Additionally, the range-of-motion results agree with the findings of Noonan et al. in that patients with MHE and radiocapitellar dislocation have loss of motion¹². However, the survey performed by Noonan et al. found that these patients do not have pain or limitations in activities of daily living. Peterson, citing multiple sources, suggested that surgeons should aggressively attempt to prevent these deformities⁹. In our own experience, we have found that families/patients want to prevent dislocation of the radial head because of loss of function and motion as well as for cosmetic reasons. Parents with MHE have expressed to us that although they have “learned to live with the deformity,” they do not want the same for their children. This was one of the motivations for us to perform this study.

Risk factors for radiocapitellar instability in individuals with MHE have been described previously^{3,4,6,7,14}. These risk factors include proportional ulnar length (in relation to the radius on radiographs and to ulnar height clinically), radial bow, and the number/location of osteochondromas (specifically in the distal part of the ulna). We chose to radiographically re-examine radial bow, ulnar length, and the presence/absence of distal ulnar osteochondromas. In addition, we investigated the effect of ulnar bow. The impetus to evaluate ulnar bow comes from our knowledge about malunited Monteggia fractures and their relationship to radiocapitellar instability¹⁵. Classically, in Monteggia fractures, the radial head dislocates in the direction of the ulnar bow¹⁶. In patients with MHE, we noted that the radial head dislocation followed the same pattern and we wanted to investigate its radiographic measurement as a potential risk factor for radial head subluxation/dislocation.

Recently, Hreha et al. studied the normal ulnar bow in both planes using a distance measurement¹⁷. They determined that 7 ± 2 mm of bow was normal in the coronal plane and 6 ± 3 mm was normal in the sagittal plane. We chose to use an angular method for 2 reasons. First, the deformity of the ulna varies in patients with MHE, with the convexity occurring on both the radial and the ulnar sides; using the method described by Hreha et al., a convex ulnar bow of the same angular degree would have a larger distance measurement than a convex radial bow. Second, we did not include the distal ulnar bow because this does not mimic a Monteggia deformity, and the method described by Hreha et al. could not accommodate for this.

We found that the total ulnar bow and percent ulnar length were independent predictors of radial head subluxation/dislocation. Our initial data analysis demonstrated that total ulnar bow, percent ulnar length, and total radial bow all dif-

fered significantly between the dislocated/subluxated group and the located group. However, total radial bow was not a significant predictor of radial head subluxation/dislocation in the binomial regression analysis. This differs from the findings presented by Jo et al.⁶ and may be explained by the addition of the total ulnar bow variable, the measurement methodology, and inclusion of patients with radial head subluxation in our study. In summary, the more growth retardation of the ulna, the fewer degrees of bowing will be associated with subluxation/dislocation. However, no radiocapitellar joint subluxated or dislocated in patients with an ulnar bow of $<17^\circ$ regardless of the relative length of the ulna. This finding raises questions about the need for ulnar lengthening and leads us to believe that treatment of the ulnar bow deformity (correction of total ulnar bow) may obviate the need to lengthen the ulna (correction of percent ulnar length).

This study was a retrospective, cross-sectional review. Ideally, a prospective, longitudinal study would be performed to follow patients and observe progression of the forearm deformities. Additionally, our study included a small number of subluxated/dislocated elbows compared with the located cohort. However, we believe that the differences observed between the groups will be helpful in guiding clinical prognostication.

We excluded patients because of inadequacy of radiographs—for example, if 2 views were obtained but they were the same view (2 coronal images of the ulna). We have now changed our protocol to obtain a coronal and lateral view of the elbow centered on the forearm with the forearm in neutral position. This has allowed us to ensure 2 orthogonal views of the forearm.

Finally, 3 of our cases did not fit into our prediction. Two patients were predicted to have a dislocation but were found to have a located radial head radiographically. One of these patients had a total ulnar bow of 24° and a percent ulnar length of 98%, and the other had a total ulnar bow of 25° and a percent ulnar length of 101%. Two possible reasons why these patients did not fit in the model are (1) our analysis does not account for the directionality of the bow, which may change the threshold for radial head subluxation/dislocation, and (2) radiographic measurement error as these cases were on the border between located and radial head subluxation/dislocation. The third patient, who had a total ulnar bow of 18° and a percent ulnar length of 98%, was predicted to have a located radial head but was found to have a dislocation radiographically. Again, this could be due to radiographic measurement error because this case was also on the border between located and subluxation/dislocation or there could have been an additional factor that contributed to this dislocation. Previous studies have suggested that osteochondroma load in the forearm may affect the radiocapitellar joint^{7,14}. Our analysis included the presence of distal ulnar osteochondromas but did not take into account additional locations of osteochondromas or the severity of involvement.

In conclusion, there are 2 radiographic measurements that can be utilized to determine the stability of the radiocapitellar joint in patients with MHE. Total ulnar bow is a new radiographic measurement that can help predict radiocapitellar dislocation and may have implications for the prophylactic treatment of radial head subluxation/dislocation. We recommend

serial clinical and radiographic examinations to monitor for radial head subluxation/dislocation. Our predictive model (Fig. 8) can help determine which patients are at risk for radial head subluxation/dislocation. This prognostication may help surgeons identify patients who would benefit from prophylactic treatment. ■

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