Radiographic indicators of a shared epiphysis in radial polydactyly

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Abstract

Background Thumb epiphyses cannot be visualized on radiographs in infants with radial polydactyly, making it difficult to classify by Wassel type. The purpose of this study was to identify radiographic features that distinguish a separate epiphysis from a shared epiphysis. This may assist in operative planning and establishing prognosis.

Methods The charts of 34 radial polydactyly patients treated with surgical reconstruction from 2008 through 2012 were retrospectively reviewed. Measurements of the most proximal bones involved in the duplication, including length, width at shaft, width at base, distance between radial and ulnar thumb, and angle between radial and ulnar thumb, were taken from PA radiographs of the thumb by four blinded individuals. The interclass correlation coefficient was calculated to determine inter-observer reliability. Operative notes were reviewed to distinguish between shared and separate epiphyses. Several indices were created from these measurements.

Results Radiographic measurements showed high inter-observer reliability. There were statistically significant differences between patients with separate and shared epiphyses for indices for the width shaft index, interspace distance, the angle×interspace distance, and the angle×interspace index.

Conclusion Radiographic differences exist between children with separate and shared epiphyses. In patients with shared

epiphyses, the radial thumb tends to be smaller, closer to the ulnar thumb, and less divergent. Threshold values were identified for predicting the status of the epiphysis based on the angle×interspace distance and the angle×interspace index. These values may be used to help determine in advance of surgery if a shared epiphysis exists.

Keywords Congenital · Thumb · Radiograph · Duplication · Classification · Wassel

Introduction

Radial polydactyly is one of the more common congenital hand differences. [2] Optimal surgical correction requires precise identification of the structures present in each of the thumb components. The Wassel classification scheme is commonly used for categorization and pre-operative planning. There are seven Wassel types. [10]

Wassel types I, III, and V by definition have a shared epiphysis. True Wassel types II, IV, and VI have a completely separate phalanx, including the epiphysis, at the level of duplication. Unfortunately, the epiphyses are usually not ossified at the age of surgery and it cannot be easily determined from radiographs if there are shared or separate epiphyses for each phalanx. Thus, it is difficult to definitively differentiate a type I from a type II, a type III from a type IV, and a type V from a type VI with radiographs alone (Fig. 1).

The purpose of this study was to identify radiographic features that distinguish a separate epiphysis from a shared epiphysis, which would allow more accurate classification of Wassel type and improve pre-operative planning and establishing prognosis. Duplications with shared epiphyses are less likely to require osteochondroplasty of the phalanx or metacarpal proximal to the duplication and have a better outcome. [1,4]

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Fig. 1 Wassel types: *left*—I or II, *center*—III or IV, *right*—V or VI

Materials and Methods

After receiving approval from the Institutional Review Board, a retrospective chart review was performed of 37 patients diagnosed with radial polydactyly. Inclusion criteria included any child age 0–5 years with radial polydactyly that was treated with surgical reconstruction. An AP or PA radiograph of the thumb was needed in order to be included in the study. Exclusion criteria included previous thumb surgery, patients treated non-operatively, and patients with inadequate radiographs. All patients were seen and treated between 2008 and 2012 at our institution. Thirty-four patients were found to be acceptable for the study. One patient was excluded because no radiographs were available and two were excluded because the available radiographs did not have AP or PA views.

Radiographs of all 34 thumb duplications were reviewed and 8 measurements were taken on each radiograph. The most proximal duplicated phalanges from the radial and the ulnar thumbs were identified and measured. For Wassel types I and II, measurements were taken from both distal phalanges. In types III and IV, measurements were taken from both proximal phalanges, and for types V and VI, they were taken from both metacarpals. A line was created down the longitudinal axis of each phalanx (e.g. proximal phalanges for types III and IV). The length of the phalanx along the axis, the width of the shaft at the midway point, the transverse width of the base, and the angle between the intersection of the axes were measured. The interspace distance was the shortest distance between the proximal bases of the duplicated phalanges (Fig. 2).

Measurements were completed by four individuals (one attending, two medical students, and one resident) in 34 radiographs in 32 patients. Two patients had bilateral radial polydactyly. Inter-observer reliability was calculated by using Interclass Correlation Coefficient (ICC) for each type of measurement. The measurements of the four researchers were averaged for each measurement in each patient. These mean values were used for comparison between the separate epiphysis and shared epiphysis groups.

Once review of all radiographs was complete, the operative notes of patients were reviewed in order to determine if a

shared epiphysis was found intra-operatively. The operative notes were also used to determine if the radial thumb or ulnar thumb was removed.

Patients were divided into separate and shared epiphysis groups based on their intraoperative diagnosis. Indices and calculated variables were constructed to combine more than one relationship into a single value. The creation of ratios and indices allowed the use of radiographic films that were not calibrated for absolute or actual distances, such as with printed radiographs.

Indices were calculated for the radial and ulnar sided lengths, base widths, and shaft widths. Three additional variables, interspace index, angle times interspace distance ($\text{angle} \times \text{IS}$), and angle times interspace index ($\text{angle} \times \text{IS index}$), were calculated.

Indices for length, width base, and width shaft were defined as the ratio of the length, width of the base, and width of the shaft, respectively, of the radial component divided by the ulnar components. Interspace index was defined as the interspace distance divided by the ulnar length. Angle $\times$ IS was defined as the thumb angle times the interspace distance. Angle $\times$ IS index was defined as the angle times the interspace index (Fig. 2).

The mean of each index was calculated for both the separate epiphysis and shared epiphysis groups and the level of significance was calculated. Two-tailed *t* tests were calculated for the indices comparing the separate and shared epiphysis groups. Each index was then plotted on a distribution graph for comparison of the separate and shared groups. A receiver operator characteristic (ROC) curve was calculated for angle $\times$ IS and angle $\times$ IS index. An ROC curve serves as a standard way of describing the accuracy of diagnostic tests, and was used to test the sensitivity and specificity of our measurements and indices at various thresholds.

Results

Of the 34 thumb radiographs reviewed, 21 % had a shared epiphysis and 79 % had separate epiphyses. The Wassel type

Fig. 2 Measurements and indices: R =radial, u =ulnar, WS and ws =width of shaft, WB and wb =width of base, IS =interspace distance, $Angle$ =thumb angle; length index= RL/ul , width base index= RWB/uwb , width shaft index= RWS/uws , interspace index= IS/ul , angle $\times$ IS=angle $\times$ IS, angle $\times$ IS index=angle $\times$ interspace index, size estimate radial= $RL(\frac{1}{2}RWS+\frac{1}{2}RWB)$, size estimate ulnar= $ul(\frac{1}{2}uws+\frac{1}{2}uwb)$

distribution in the 34 patients were: type I—6 %, type I—24 %, type III—12 %, type IV—41 %, type V—3 %, type VI—0 %, type VII—15 %. All five of type VII had separate epiphyses. In four of type VII, the duplication was at the level of the proximal phalanx. In one of type VII, the duplication was at the level of the interphalangeal joint. Of the 34 thumbs, 3 patients had the radial thumb retained. In the remaining 31 cases, the ulnar component was retained.

ICC of each measurement was calculated. The absolute ICC of radial length, ulnar length, radial width base, ulnar width base, radial width shaft, ulnar width shaft, interspace, and thumb angle were 0.987, 0.973, 0.967, 0.977, 0.923, 0.965, 0.987, and 0.973, respectively. The mean absolute ICC was calculated to be 0.969 indicating very high inter-observer reliability.

There were statistically significant differences between groups with respect to mean width shaft index, interspace distance, angle $\times$ IS, and angle $\times$ IS index. The mean width shaft index difference suggested that the smaller the radial thumb, the more likely it was to have a shared epiphysis. The mean interspace difference shows that a small interspace distance is more likely to have shared epiphysis and a large interspace distance is more likely to have a separate epiphysis. The angle $\times$ IS value shows that the more divergent and the further away the duplicated bones are from each other the more likely they are to have a separate epiphysis. The angle $\times$ IS index is a similar measure to angle $\times$ IS, but it utilizes an index, which eliminates length measurement units (Fig. 3).

A ROC curve was calculated for the angle $\times$ IS and the angle $\times$ IS index. Cutoff values for the likelihood of shared or separate epiphyses are shown (Tables 1 and 2). For the angle $\times$ IS, values less than 24.93 predict shared epiphyses, while values greater than 31.41 indicate the epiphyses are separate (Table 1). For the angle $\times$ IS index, values less than 2.87 predict shared epiphyses, while values greater than 3.87 indicate the epiphyses are separate (Table 2).

Discussion

Radial polydactyly is one of the more common congenital hand differences. [2,3,7] In the USA, it is more common in Asian and Native American populations, and it is seen with equal frequency in Caucasians and Afro-Americans. [8] The level of splitting is variable, and all the thumb structures can be involved as a combination of duplicated, shared, hypoplastic, or missing parts.

The classification type helps predict what kind, if any, of joint changing procedures need to be performed at surgery. While there are a number of classification systems, the Wassel classification scheme is commonly used. [10,11] The epiphyses are usually not ossified at the time of surgery, and therefore cannot be visualized on routine radiographs. In pre-operatively defined Wassel types II and IV, the split segment can either have a shared or separate epiphysis. If they are shared, the thumbs would be classified differently. While not essential, knowing the status of the epiphyses prior to surgery is useful.

Islam et al. reviewed 158 duplicated thumbs, with review of the radiographic, intra-operative, and pathological findings. [5] Based on their intra- and post-operative findings, 23 % of the patients changed into a different classification compared to the preoperative diagnosis. In a separate study, Islam et al. performed contrast arthrography in 20 patients at the start of their corrective procedure. [6] They were able to identify shared epiphyses and alterations in the proximal joint surfaces. Seven of the patients (35 %) would have been switched to a new Wassel type.

Chung et al. studied 159 duplicated thumbs and proposed a new classification scheme based on the anatomical joint relationship at the level of duplication. Their categories were duplicate epiphyses, shared epiphyses, osteochondroma type, and hypoplastic type.

Six percent of their patient changed categories based on the operative findings. Their incidence of shared epiphyses corresponded with our findings. Duplications with shared

Fig. 3 Comparison of means with statistically significant differences. m =mean

Table 1 ROC curve coordinates—angle index

Index	Sensitivity, % (for predicting separate epiphyses)	Specificity, %
10.79	100	57.1
15.90	100	71.4
24.93	100	85.7
31.41	96.3	100
40.66	92.6	100
52.36	88.9	100
62.23	77.8	100

Table 2 ROC curve coordinates—angle×IS index

Index	Sensitivity, % (for predicting separate epiphyses)	Specificity, %
0.90	100	57.1
1.63	100	71.4
2.88	100	85.7
3.87	96.3	100
4.64	92.6	100
5.63	88.9	100
7.91	77.8	100

epiphyses are less likely to require osteochondroplasty of the head of the phalanx or metacarpal proximal to the duplication. [1]

Ogino et al. studied their long-term results in 113 patients. Patients with Wassel types III and V duplications had less favorable outcomes. They found no differences based on whether articular cartilage shaving was performed, but state that longer follow-up is needed to determine its influence. [9]

Thumbs duplicated at the metacarpophalangeal joint and classified pre-operatively as Wassel type IV were studied by Horii et al. Of the 106 cases, 16 % shared a wide cartilaginous connection between the duplicated segments and 68 % had separate epiphyses. Patients with separate epiphyses had a higher percentage of good results (80 %) compared with those with a shared epiphysis (50 %). Thumbs with shared epiphyses were more likely to require bone alignment. [4]

Physical exam can also aid in establishing the status of the epiphysis. Thumbs with separate epiphyses are more likely to have independent relative mobility or passive motion, be hypoplastic, or have the duplicated segment originate more proximally. [1] Ultrasound and MRI can also be used. Ultrasound is highly practitioner dependent and not commonly available for musculoskeletal evaluation. MRI is expensive, requires sedation for younger children, and is difficult to interpret. Arthrography has also been reported. [6] Radiographs are easily obtained, relatively inexpensive, and are commonly ordered by the referring physicians.

Surgeons can often predict the status of the epiphyses from physical exam and initial review of the radiographs based on the relative size and the position of the duplicated segments. For instance, when the origin of duplicated segments is mid-shaft, the more distal components of the phalanges are clearly separate. Our approach, however, quantifies the differences in size and position, and provides an additional predictive tool when needed.

We found several identifiable differences between thumb duplications with shared epiphyses and those with separate epiphyses, and that it is possible to better identify the exact Wassel type based on radiographs. Prior to this study, there were no published methods to easily differentiate pre-operatively a Wassel I from a Wassel II, a Wassel III from a Wassel IV, or a Wassel V from a Wassel VI.

Our study shows that a small interspace distance is more likely to have shared epiphysis and a large interspace distance is more likely to have a separate epiphysis. Based on our width of shaft data, we also showed that when the thumbs are close to the same size they are more likely to have separate epiphyses. When one phalanx is much smaller than the other, they are more likely to share an epiphysis. Further, our study shows that the more divergent and the further away the duplicated bones are from each other the more likely they are to have a separate epiphysis. These observations may be quantified by using the angle $\times$ IS and the angle $\times$ IS index.

Both the angle $\times$ IS value and the angle $\times$ IS index value can identify either a shared or separate epiphysis in most cases. If calibrated measurements are available, the angle $\times$ IS should be used since the calculations are based on two measurement and one multiplication (the angle of the components multiplied by the distance between the base of the bones).

In the age group studied, obtaining true AP or PA radiographs was difficult, and there were differences in the amount of rotation of the hands. Further study will be needed to determine if better standardization of the radiographs will decrease the size of the indeterminate zone. Other limitations of our study include a relatively small sample size, and the absence Wassel VI patients. This method will need to be tested on independent surgeons to see if their predictions improve over physical exam. The method is also initially time consuming.

Differences in size, amount of divergence, and distance of separation of the thumb components can be used to help predict the status of the epiphysis. If the product of the angle (degrees) and the interspace distance (millimeters) is less than 25 there is a shared epiphysis, and if that value is greater than 31 there are separate epiphyses. If calibrated distances cannot be measure, the angle $\times$ IS index can be used. Using these simple measurements and guidelines will help surgeons more accurately predict the Wassel type, predict outcomes, and strengthen their pre-operative plan.

Conflict of Interest Michael H. Johnson, MD declares that he has no conflict of interest.

Roshan T. Melvani, BLA declares that he has no conflict of interest.

Neil N. Patel, BLA declares that he has no conflict of interest.

Christine J. Cheng, MD, MPH declares that she has no conflict of interest.

Richard L. Hutchison, MD declares that he has no conflict of interest.

Statement of Human and Animal Rights All procedures followed were in accordance with the ethical standards of our Institutional Review Board and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent No patient identifiers being used; the IRB decided that informed consent was needed. No informed consent was obtained per IRB guidelines.

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