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Clinical and radiological aspects of the forearm in children with congenital radioulnar synostosis: a cohort study

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Abstract

Introduction The upper limb functional limitations in congenital radioulnar synostosis may significantly affect the daily activities of patients. Classifications of the condition are descriptive and have limited practical application.

Purpose Determine a functionally significant quantitative criterion for anatomical changes in the forearm.

Material and methods 92 children (136 forearms) with congenital radioulnar synostosis were examined for limitations in activities of daily living (ADL), health-related quality of life measured with PedsQL questionnaire; pronation of the forearm and radiographic parameters. A comparative and correlation analysis, ROC analysis were performed to determine the relationship between the forearm pronation and limitations of ADL.

Results Statistically significant correlations were revealed between symptoms and the forearm alignment ($p < 0.01$, $r_{xy} = 0.5$); subluxation of the ulnar head and forearm alignment ($p < 0.001$, $r_{xy} = 0.6$); bowing deformity of the radius, forearm alignment and subluxation of the ulnar head and between the length of the forearm bones and bowing deformity of the radius ($p < 0.05$, $r_{xy} = 0.4$ and $r_{xy} = 0.5$). A statistically significant inverse correlation was revealed between symptoms and PedsQL scores ($p = 0.038$, $r_{xy} = -0.4$). Pronation of 45° was the threshold value of the forearm alignment with a high risk of ADL limitation. The area under the ROC curve corresponding to the relationship between symptoms and the forearm alignment was 0.955 ± 0.021 (95 % CI: 0.915–0.995). There was a statistically significant ($p < 0.01$) decrease in the lumen of the medullary canal in the middle third of the ulnar shaft with the radius lumen being unchanged. Dorsal subluxation of the ulnar head was detected In 30 % of cases.

Discussion The characteristics identified demonstrated changes in the forearm bones with functional impairments being correlated with the forearm pronation.

Conclusion The correlation between the patient's symptoms and the forearm alignment must be taken into account in the classification and when determining indications for surgical treatment distinguishing between functional (< 45° pronation) and dysfunctional (⩾ 45° pronation) options.

Keywords: radioulnar synostosis, child, classification

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INTRODUCTION

Congenital radioulnar synostosis (CRUS) is a rare developmental condition of the upper limb caused by failure of differentiation that leads to the joint enchondral ossification in utero at the stage of embryogenesis [1, 2, 3]. Embryology-based Oberg-Manske-Tonkin (OMT) classification was accepted as the new classification system for all congenital upper extremity anomalies. CRUS is classified as disruption of radioulnar (antero-posterior) axial differentiation of tissues [4, 5]. Synostosis of bones occurs at the level of the proximal radioulnar joint, and can extend to the distal third of the forearm [6]. Idiopathic congenital synostosis of the distal forearm bones is an occasional case and can have a different genesis [7, 8]. Functional limitations in CRUS can affect daily activities, with pronounced pronation of the forearm and bilateral lesions, in particular [9, 10].

The current classifications of CRUS are descriptive and based on radiological manifestations seen at the level of the proximal forearm and have limited practical application.

The purpose of the work was to determine a functionally significant quantitative criterion for anatomical changes in the forearm.

MATERIAL AND METHODS

The review of 92 patients (136 forearms) aged 2.5 to 17 years was performed at the National Medical Turner Research Center for Pediatric Trauma and Orthopaedics between 2010 and 2022. The patients were examined and/or treated for congenital radioulnar synostosis (Table 1).

Table 1

Characteristics of children

Description		Total	Presented with complaints		No complaints reported	
			abs.	%	abs.	%
Number of patients		92	77	83.7	15	16.3
Involved side, No. of cases	left	33	28	84.8	5	15.2
	right	15	10	66.7	5	33.3
	both	44	39	88.6	5	11.4
Graded by Cleary – Omer, No. of forearms	I	7	4	57.1	3	42.9
	II	11	6	54.5	5	45.5
	III	104	97	93.3	7	6.7
	IV	14	9	64.3	5	35.7
Males		60	47	78.3	13	21.7
Females		32	30	93.8	2	6.2
Age, completed years, Me [Q1–Q3]		7 [4–10]	6 [4–9]		8 [7–13]	

The design is a single-center retrospective cohort study for the first part and a case-control study for the second part of the work. The study was performed in accordance with STROBE recommendations and was divided into two consecutive parts. The purpose of the first part was to assess the relationship between patient complaints, upper limb function and clinical presentation of padiatric forearm with CRUS. The purpose of the second part was to analyze the radiological parameters in patients with unilateral CRUS. The study design is presented in Figure 1.

Inclusion criteria included radiologically verified congenital radioulnar synostosis, patients under 18 years of age. The study did not include patients with incomplete data and those after surgical treatment.

Exclusion criteria for the second part of the work included bilateral involvement of the upper limbs and the absence of preoperative radiological findings. Pronation alignment of the forearm measured in degrees was assessed clinically using a goniometer at an elbow flexion of 90°. Subluxation of the ulnar head was identified using radiographs of the forearm in a strictly lateral projection (Fig. 2).

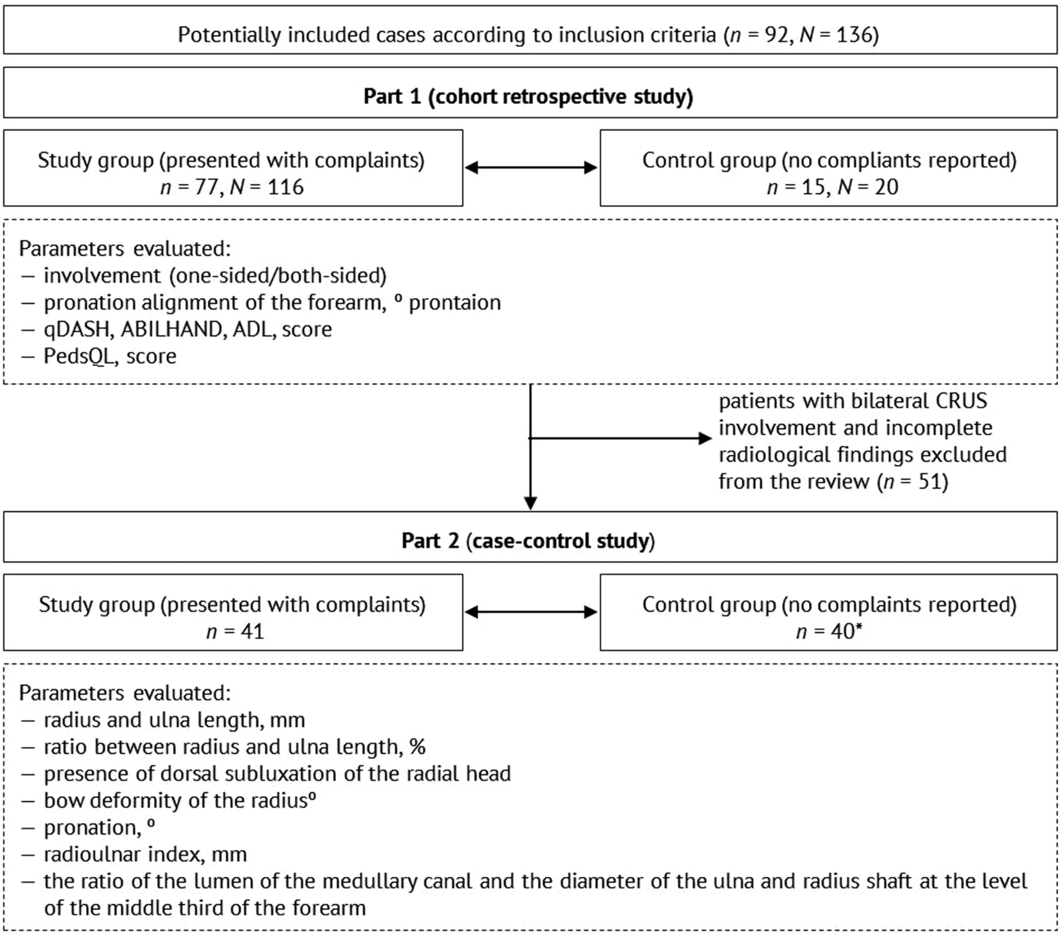

Fig. 1 Study Design Flowchart; * radiological findings of the contralateral healthy limb were unavailable in 1 case

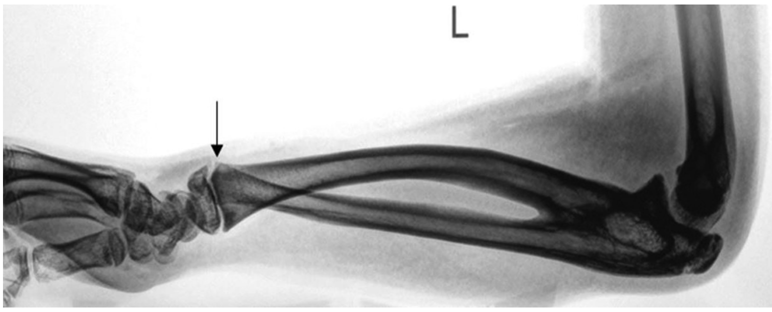

Fig. 2 Radiograph of the forearm of a patient with CRUS in a strictly lateral projection with the arrow indicating the dorsal subluxation of the ulnar head

The Hafner method was used for ulnar variance measurement in children under 11 years of age [11]. Ulnar variance measured in patients 12 years of age and older referred to the difference in height between the joint surfaces of the distal ulna and the distal edge of the radial sigmoid notch. Liu et al. reported measurements of the radius pronation angle using the special flexed posterior-anterior views of the X-ray image of the forearms [12]. Complaints about limitations in daily activity were assessed according to the 12-point ADL scale (activity of daily living) [13].

Subjective evaluation of the forearm function in children with CRUS consisted of a set of 12 questions regarding the basic activities of life [13]. The length of the forearm bones was measured using lateral radiographs. The ulna length was measured as the distance between the olecranon and the styloid process. Radial length was defined by the length measured between the tip of the radial styloid and the distal articular surface of the ulna. In younger children with radiological absence of ossification nuclei of the distal and proximal parts of the forearm bones, the length was measured from the edges of the metaphyses, and from the most distal and proximal points of the secondary ossification centers in the presence of ossification nuclei.

Statistical analysis was performed using IBM SPSS Statistics 26. The distribution of quantitative data was primarily assessed using the Kolmogorov – Smirnov test with the Lilliefors correction for a sample size of more than 50 and using the Shapiro – Wilk test for a smaller sample size. The sample size was not calculated in advance. A comparative analysis of radiological parameters was performed using the nonparametric Mann – Whitney test after preliminary assessment of the data distribution. A correlation analysis was performed using the Spearman criterion to analyze the relationships between radiological, clinical and functional parameters. Strength of relationship was assessed using the Chaddock scale. ROC analysis was performed to determine the cut-off point for predicting complaints and creating a binary classification on the basis of the forearm pronation alignment and limitations in activities of daily living. The study was approved by the institutional ethics committee and was conducted in accordance with the ethical standards outlined in the Declaration of Helsinki.

RESULTS

A higher prevalence of CRUS was found in male patients among individuals seeking surgical treatment (ratio 2:1). The mean age at the time of referral for surgery was 6 years. Morphotype III of Cleary and Omer classification was the most common (Table 1). Although an identical morphological type was observed in 70.5 % of cases of bilateral involvement, 13 patients out of 44 (29.5 %) with bilateral CRUS were diagnosed with different types as grouped by Cleary – Omer. Statistically significant differences in the forearm pronation alignment were identified between the group of patients who presented no limitations in activities of daily living, and those who did (Table 2). Radiological parameters of the second part of the study and quantification characteristics are presented in Table 3.

Table 2

Forearm bone alignment in the groups

Description	Total	Presented with complaints	No complaints reported	*p*
Pronation alignment of the left forearm, degrees, Me [Q1–Q3]	70 [30–90]	80 [50–90]	10 [10–20]	< 0.05*
Pronation alignment of the right forearm, degrees, Me [Q1–Q3]	85 [30–90]	90 [60–90]	10 [5–15]	< 0.05*

* statistically significant differences identified between the groups

Table 3

Radiological findings in patients with CRUS

Description		Unilateral CRUS involvement
Subluxated ulnar head, abs. (%)		19 / 48 (39.5 %)
Radius length, % relative to the normal limb, M ± SD		91.6 ± 5.4
Ulnar length, % relative to the normal limb, M ± SD		94.1 ± 6.2
Bow deformity of the radius, degrees, M ± SD		21.36 ± 6.05
Ulnar length, % relative to the radius, M ± SD	involved limb	103.4 ± 5.5
	normal limb	102.8 ± 4.2
The lumen of the medullary canal at the level of the middle third of the ulnar shaft, % of the diameter of the shaft at the level, M ± SD	involved limb	37.3 ± 8.1
	normal limb	45.9 ± 9.7
The lumen of the medullary canal at the level of the middle third of the radial shaft, % of the diameter of the shaft at the level, M ± SD	involved limb	41.9 ± 8.2
	normal limb	45.5 ± 9.5
Ulnar variance, mm, Me [Q1–Q3]	involved limb	0.74 [−1.48–1.52]
	normal limb	−0.84 [−2.56–0]
Pronation angle, degrees		11.06 ± 0.47

Dorsal subluxation of the ulnar head was seen in 30 % of all observations (41 out of 136 forearms) with greater proportion observed with Cleary-Omer type III CRUS noted in 37.5 % (39 out of 104 forearms). The length of the ulna and radius was 91.6 ± 5.4 % and 94.1 ± 6.2 % relative to the length of the intact bones of the contralateral forearm, respectively. There were no statistically significant differences in the ratios between the forearm bone lengths (ulna relative to radius) of the healthy and affected limbs. A statistically significant ($p < 0.01$) decrease in the lumen of the medullary canal and thinning of the ulna was revealed, with a relatively maintained diameter of the radius as compared with the healthy limb (Fig. 3).

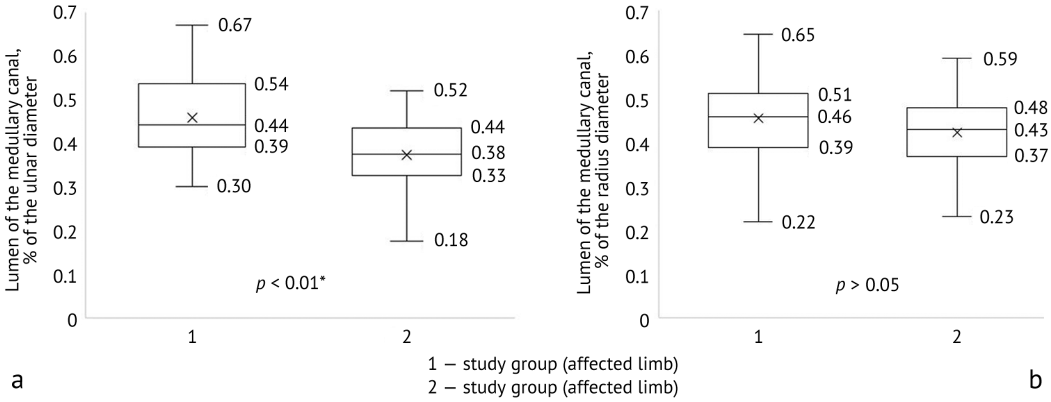

Fig 3 Comparison of the mean measurements of the lumen of the medullary canal at the level of the middle third of the forearm relative to the diameter of the ulna (a) and theradius (b): * statistically significant differences between the groups ($p < 0.01$)

Spearman's correlation coefficient showed statistically significant ($p < 0.01$, $r_{xy} = 0.5$) positive association between presentation of complaints and the forearm alignment measured with the Chaddock scale. An inverse correlation of moderate tightness ($p = 0.038$, $r_{xy} = -0.4$) measured with the Chaddock scale between complaints and PedsQL scores indicating general health. There were no statistically significant correlations between complaints and the affected side, gender, age, functional scales ADL, Failla, ABILHAND, qDASH and the total PedsQL score.

Statistically significant ($p < 0.05$) positive correlations of moderate tigntness ($r_{xy} = 0.4$ and $r_{xy} = 0.5$) measured with the Chaddock scale were identified between the bow deformity of the radius, the forearm pronation alignment and subluxated ulnar head, between the length of the radius and ulna and the bow deformity of the radius (Fig. 4). No statistically significant correlations were found between the forearm pronation alignment and the patient's age or ulnar variance.

A statistically significant ($p < 0.001$) positive correlation of noticeable tightness measured with the Chaddock scale ($r_{xy} = 0.6$) was revealed between dorsal subluxation of the ulnar head and severity of the forearm pronation alignment (Fig. 4).

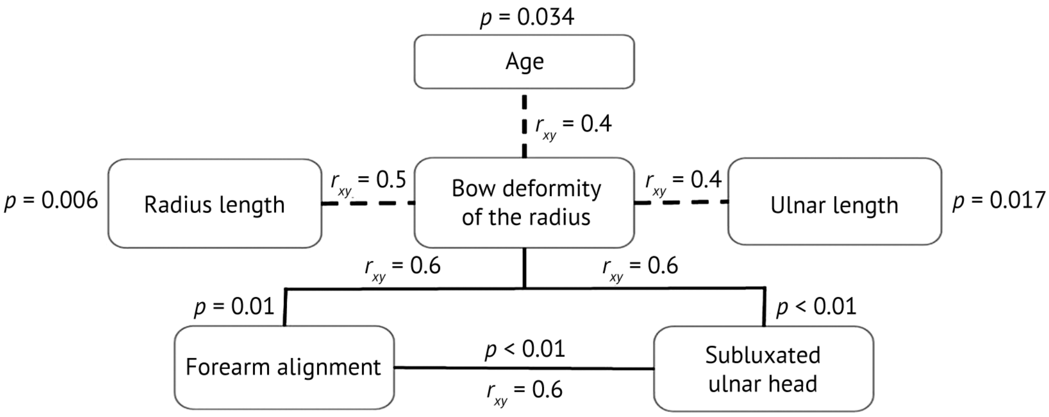

Fig. 4 Results of Spearman correlation analysis. Positive correlations are marked with solid lines, reverse correlations are marked with dotted lines

With a statistically significant correlation between limited activities of daily living and the forearm alignment an ROC analysis was performed to determine the minimum threshold value of the forearm alignment with higher values indicating the increased likelihood of having complaints. No correlations between the involvement side (unilateral or bilateral CRUS) and complaints about limited activities of daily living were identified at the previous stage of statistical data processing, the forearm with a greater pronation alignment was selected as more clinically significant for ROC analysis; the pronation alignment of one forearm was taken into account with the same forearm alignment. The area under the ROC curve corresponding to the relationship between the prediction of the complaints and the severity of pronation alignment of the right forearm measured 0.955 ± 0.021 degrees with 95 % CI: 0.915–0.995 (Fig. 5). The resulting model was statistically significant ($p < 0.001$). The threshold value of the forearm alignment at the cut-off point was 45° pronation. If the forearm alignment was equal to or greater than this value, the patient was predicted to have a higher risk of complains about limited activities of daily living. The sensitivity and specificity of the method were 91 % and 100 %, respectively.

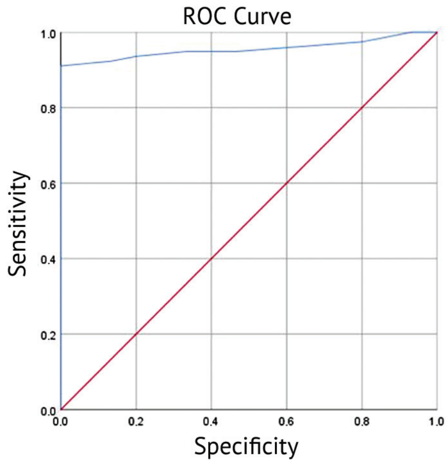

Fig. 5 Results of ROC analysis for binary classification of CRUS depending on the forearm pronation alignment in degrees

DISCUSSION

Most common classifications of patients with CRUS published in research works (Table 4) are based on radiological findings of the condition. The authors of the most popular classification [10] reported its limited use in evaluation of the limb function and making tactical decisions regarding treatment approaches [14, 15, 16].

Table 4

Classifications of CRUS

Authors, year, reference	Description
Tachdjian, 1990 [17]	Type I: “true” CRUS or type without radial head. radiographic radial head unobservable and osseous synostosis with the ulna. The radius is bowed, its thickness is greater than the thickness of the ulna Type II: with dislocated radial head. The malformed head of the radius is displaced posteriorly. Proximal bone fusion. Type III: There is no bony fusion, but there is a pronounced fibrous band attached to both bones limiting rotational movements. The rarest type
Cleary, Omer Jr, 1985 [14]	Type I: Fibrous ankylosis with normal radial head, without bone changes, but with limited movement and forearm shortening Type II: bony synostosis, radial head formed, centered Type III: bony synostosis, radial head displaced posteriorly Type IV: bony synostosis, the radial head displaced anteriorly
Wilkie, 1914 [18]	Type I: fusion of the medullary canals of the radius and ulna, the radius is larger and longer than the ulna Type II: anterior or posterior dislocation of the radial head, bony synostosis of the proximal shafts of the forearm bones

The morphological characteristics we identified indicated changes in the forearm bones including the proximal radioulnar joint. A decrease in the lumen of the medullary canal at the level of the middle third of the shaft and changes at the distal radioulnar joint, correlate with modern concepts, taking into account the stages of embryogenesis and the classification of malformations of the upper extremities graded by the OMT classification [4, 5].

In our series, subluxation of the ulnar head was observed in 30 % of cases. The subluxation was seen in the most common type III synostosis as graded by Cleary – Omer [19, 20, 21, 22]. It was noted that the more pronated the forearm was, the more common dorsal subluxation of the ulnar head observed (Fig. 4). We suggested that it might be caused by a more intense growth of the ulna and its relative “overlengthening.” The proximal radial physis and the distal ulnar physis are responsible for 20 % of the forearm growth [23, 24]. “Mute” proximal radial physis in type III CRUS is likely to be responsible for a slowdown in growth rates leading to delayed bone growth and causing a bow deformity of the radius [25], progression of incongruity in the distal radioulnar joint and subluxation.

Then there is a question: is the discrepancy between the longitudinal dimensions of the radius and ulna bones truly anatomical or is it a projection distortion on the radiograph? Epner et al. [26] and Palmer et al. [27] reported the relative radial and ulnar variance being dependent on the position of the forearm. The pronation visually increases the ulnar variance, while the supination, on the contrary, reduces it. Jung et al. [28] studied radiographs of the wrists of 120 healthy volunteers and determined maximum ulnar variance when gripping in pronation and minimum ulnar variance when relaxed in supination. Standardization of study designs or computed tomography of the forearm in patients with CRUS can be a solution to interpretation of the findings.

Yeh et al. [29] recommended to standardize the measurement of ulnar variance with neutral rotation radiographs of the wrist. However, neutral rotation radiographs of the wrist cannot be anatomically produced in patients with radioulnar synostosis. Although the authors found a statistically significant difference in ulnar variance between the pronated and neutral positions, this difference may not be clinically significant [29]. The presence of radial bone deformity would not normally allow restoration of rotational movements even with the synostosis being separated and grafts implanted [30, 31, 32]. The absence of statistically significant differences in the forearm bone length ratios (ulna relative to the radius) indicates a proportional shortening of both bones of the affected forearm. This disproportion may indicate partial preservation of the function of the proximal physes of the radius having growth potential of 20–25 % [23, 24]. The absence of statistically significant correlations between the severity of the forearm pronation and the patient’s age may indicate static changes that do not progress over time. Pathological changes in CRUS in children involve the whole forearm structures, and are not limited to changes at the level of the proximal radioulnar joint.

Limitations of the study A small sample size of patients with no active complaints for limited activities of daily living with CRUS being an incidental finding. If the forearm position is described as an average physiological one, patients may not seek consultation with a doctor or keep outpatient appointments at the place of residence. This would contribute to a representative sample.

CONCLUSION

We offer to give consideration to the dependence of the patient’s presentation of complaints and the forearm position specified in the CRUS classification, and when determining the indications for surgical treatment of pediatric patients with this condition, distinguishing functional (< 45° pronation) and dysfunctional (⩾ 45° pronation) options.

Conflict of interest *The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.*

Funding *The study was performed without external funding.*

Ethical review *The study was approved by the local ethics committee and was executed in compliance with the Declaration of Helsinki.*

Informed consent *All patients signed a voluntary informed consent form.*

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