Hand Function and Activity Performance of Children with Longitudinal Radial Deficiency

By Laurien M. Buffart, MSc, Marij E. Roebroeck, PhD, Wim G.M. Janssen, MD, Anneke Hoekstra, PT, Ruud W. Selles, PhD, Steven E.R. Hovius, MD, PhD, and Henk J. Stam, MD, PhD, FRCP

Investigation performed at the Departments of Rehabilitation Medicine and Plastic and Reconstructive Surgery, Erasmus MC, University Medical Centre Rotterdam, Rotterdam, The Netherlands

Background: The effects of treatment of children with longitudinal radial deficiency are generally evaluated by measuring grip and pinch strength and joint mobility. Insight into limitations of activities of children with radial deficiency is scarce. In this study, we used standardized instruments to assess impairments in hand function and activity limitations and explored the relationship between the two.

Methods: We evaluated the hand function of twenty children with radial deficiency who were between four and twelve years of age. Impairments in hand function were assessed by measuring grip and pinch strength and the active range of motion of the wrist and of the metacarpophalangeal and proximal interphalangeal joints of the second digit. Functional activities were assessed with use of the Assisting Hand Assessment (AHA), to measure the effectiveness of the affected hand, and the Prosthetic Upper Extremity Functional Index (PUFI), to evaluate the ease of activity performance. The relationship between hand function and activity performance and the relationship of those measures with the type of radial deficiency were determined.

Results: The average grip and pinch strengths were 36% and 30% of reference values. We found reductions in the active range of motion, particularly of the metacarpophalangeal and proximal interphalangeal joints. The mean AHA score was 85.5 points and the mean PUFI score was 81.8 points, with both measured on a 0 to 100-point scale. Grip and pinch strength, the active range of joint motion, and the sum scores on the two functional tests were related to the type of radial deficiency. Significant relationships were found between impairments in hand function and activity performance. There was a large variation in the activity performance of the children with poor strength, whereas a more linear relationship was found between the active ranges of motion of the wrist and finger joints and activity performance.

Conclusions: Despite marked impairments in hand function, children with radial deficiency performed functional activities fairly well. Relationships between impairments in hand function and limitation of activities were not linear. We recommend that evaluations of the results of treatment include assessment of both aspects of hand function.

Longitudinal radial deficiencies range from hypoplasia of the thumb to complete absence of the radius. Absence of the radius results in a lack of radial support to the wrist, often leading to a marked radial deviation at the wrist and a reduced range of motion of this pseudojoint. Also, skeletal abnormalities such as bowing of the ulna and abnormalities in radial-sided muscles, ligaments, and neurovascular elements may be present. Overall, depending on the severity of the longitudinal radial deficiency, these patients may have limited hand function[1].

In the clinical evaluation of hand function, it is current practice to focus on impairments by measuring grip and pinch strength and joint mobility[2]. A more complex aspect of function is the execution of a task or action by an individual, which is referred to as *activity* by the World Health Organization in the International Classification of Functioning, Disability and Health[3]. For the patient, limitations of activities may be more important than impairments in grip and pinch strength and joint mobility[4]. Because there may not be a direct relationship

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between impairments in hand function and associated activity limitations[5-7], insight into both aspects may be useful.

Research on activity limitations of children with longitudinal radial deficiency is scarce. In a recently conducted systematic review, Ho and Clarke concluded that hand performance during daily activities has been evaluated in only a few studies[2], and most of these evaluations were based on simple observation of the patients' performance of tasks[8,9] or unvalidated questionnaires[10]. Dexterity tests such as the Purdue pegboard[11] and Jebsen-Taylor[12] tests have been used, but they do not measure performance of functional activities in daily life. Several instruments have been developed to measure the upper-extremity function of children with different diagnoses[13]. Instruments such as the Assisting Hand Assessment (AHA)[14] and the Prosthetic Upper Extremity Functional Index (PUFI)[15,16] focus on bimanual activities rather than evaluating the two hands separately during the performance of certain tasks, as the Jebsen-Taylor test[17] and the Melbourne Assessment[18] do. Both the AHA and the PUFI were developed for the evaluation of children with a unilateral limb deficiency. The AHA was developed for children with neurological conditions, such as cerebral palsy and obstetrical brachial plexus palsy, and the PUFI was developed for children with a transverse reduction limb deficiency. In a previous study, we found evidence that the AHA and the Ease of Performance Scale of the PUFI provide valid and reliable results when used for children with longitudinal radial deficiency[19]. In the present study, we focused on assessing, with use of standardized instruments, hand function impairments and activity performance in a series of children with longitudinal radial deficiency. Furthermore, we explored the relationship between impairments in hand function and activity performance.

Materials and Methods

Patients

From March to October 2004, children with longitudinal radial deficiency between the ages of four and twelve years were consecutively recruited from the Department of Rehabilitation Medicine and the Department of Plastic and Reconstructive Surgery of Erasmus MC, University Medical Centre Rotterdam. To attain a sample of children with a variety of radial deficiencies in terms of type and severity, we approached the parents of all children with a unilateral deficiency and a sample of bilaterally affected children with different types of longitudinal radial deficiency. The parents of twenty-eight children were invited to enroll their child in the study, and twenty children participated (a 71% response rate). A lack of time was the main reason given by the parents for not participating in the study. The local ethics committee approved the study, and the parents of the children signed an informed-consent form.

The mean age of the children (and standard deviation) was 7.6 ± 2.4 years, and 80% were boys. Children who were bilaterally affected were underrepresented in this study: 35% of the children in the study were bilaterally affected compared with 49% of all children between the ages of four and twelve years who were seen for longitudinal radial deficiency at Erasmus MC. Information on the type of longitudinal radial deficiency, associated anomalies, and the surgical history was obtained from medical records. The type of longitudinal radial deficiency was classified according to the system of Bayne and Klug[20] as type I (a short distal part of the radius), type II (a hypoplastic radius), type III (partial absence of the radius), or type IV (total absence of the radius). Five children had a syndrome: four of them had Holt-Oram syndrome, and one had a VACTERL (vertebral anomalies, anal atresia, cardiac anomalies, tracheoesophageal fistula, renal anomalies, radial anomalies, lung anomalies) association. Nearly all children in the study had been treated surgically. Depending on the type of longitudinal radial deficiency, surgical procedures included soft-tissue distraction at the wrist level, centralization preserving wrist mobility, pollicization, and opponensplasty (Table I). Ulnar distraction lengthening is mostly performed in older children at our institution. Surgical intervention was followed by hand therapy with splinting and exercises.

Procedures

Impairments in hand function were evaluated by measuring the grip and pinch strength and the active range of motion of the joints of the hand that was used as an assisting hand. The unilaterally affected children used the affected hand as the assisting hand, and those with bilateral involvement used the more severely affected hand as the assisting hand. Functional activities were assessed with a functional test (the AHA[14]) and with a questionnaire (the PUFI[15,16]). Selection of these instruments was based on an extensive literature review[13] and evaluation of the validity and reliability of the instruments when used for children with longitudinal radial deficiency[19]. A certified hand therapist administered the AHA. The parents of the children filled out the PUFI.

Measurement Instruments

Grip and Pinch Strength

Power grip and tripod pinch strength were measured with the Lode hand-grip and pinch-grip dynamometers (Lode, Groningen, The Netherlands), which are similar in design to the Jamar dynamometer and the Preston pinch-grip dynamometer, respectively. The Lode dynamometer was used with the handlebars in position 2. During the measurements, the children were seated at a table and they were told to keep the elbow flexed without resting the arm or the grip handle of the dynamometer on the table. The mean of three maximum voluntary contractions was recorded. The reliability of these dynamometer measurements in healthy children (four to twelve years old) and adults and in adults with hand injuries has been found to be excellent[21,22].

Range of Motion

A standard goniometer was used to measure the active range of motion. We measured the total flexion-extension arc at the wrist. So that the measurements would be consistent for children with a thumb (either a hypoplastic or a pollicized thumb) and those without a thumb, we measured the total flexion-extension at the metacarpophalangeal and proximal interphalangeal joints of the second digit, defined as the first finger ulnar to the (pollicized or hypoplastic) thumb or as the first

TABLE I Characteristics of Participating Children

Characteristic	
Mean age (and stand. dev.) *(yr)*	7.6 ± 2.4
Sex*	
Boys	16 (80)
Girls	4 (20)
Type of radial deficiency*	
Type I	7 (35)
Unilateral	6 (30)
Bilateral	1 (5)
Type II	4 (20)
Unilateral	2 (10)
Bilateral	2 (10)
Type III	2 (10)
Unilateral	0 (0)
Bilateral	2 (10)
Type IV	7 (35)
Unilateral	5 (25)
Bilateral	2 (10)
Surgical treatment*†	
None	3 (15)
Distraction	6 (30)
Centralization	10 (50)
Opponensplasty	12 (60)
Pollicization	10 (50)

*The values are given as the number of children with the percentage in parentheses. †Most children had more than one surgical treatment.

finger in the children without a thumb. To explore relationships with activity performance, we used the sum of the active range of motion of the metacarpophalangeal and proximal interphalangeal joints of the second digit as an indicator of the active ranges of motion of the finger joints.

AHA

The AHA[14] is used to evaluate activity performance by means of an assessment of how effectively the child uses the assisting hand in bimanual play. The AHA is conducted during a semi-structured play session. Scoring focuses on spontaneous arm use. A video recording is made of the children, standardized according to the criteria of AHA, English research version 4.1, and the scoring is based on that video[23]. In the manual, the toys described for children between the ages of five and twelve years are slightly different from those described for children between the ages of eighteen months and five years. Items of the AHA refer to six domains: general usage, arm usage, grasp-release, fine motor adjustment, coordination, and pace. The quality of performance is scored on a 4-point scale, with 4 points indicating effective, 3 points indicating somewhat effective, 2 points indicating ineffective, and 1 point indicating "does not do." Raw sum scores range from 22 to 88 points and are transformed to scaled scores ranging from 0 to 100 points, representing a percentage distribution within the scale. The AHA has proved to be valid and reliable for children with cerebral palsy, obstetrical brachial plexus palsy[7,14], and radial deficiency[19].

PUFI

The PUFI[15,16] was developed for children with transverse reduction limb deficiencies. It is used to evaluate the extent to which a child actually uses the prosthesis for daily activities, the comparative ease of task performance with and without the prosthesis, and the perceived usefulness of the prosthesis. In the present study, we only addressed the ease of performing activities, which was scored on a 5-point ordinal scale ranging from "no difficulty" to "cannot do."

The young-child version (ages three to six years) of the PUFI includes twenty-six upper-extremity bimanual activities, whereas the older-child version (seven years of age or older) includes thirty-eight bimanual activities. For both versions, scaled sum scores range from 0 to 100 points. Higher scores correspond to less difficulty with performance. The PUFI has been found to have good validity and reliability when used for children with longitudinal radial deficiency[19].

Statistical Methods

The mean values and standard deviations for grip and pinch strength, the active range of motion, and the sum scores of the AHA and PUFI are presented. In addition, the percentage of the reference value is given for grip and pinch strength (except for the pinch strength of four-year-old children because of the unavailability of reference values) and the active range of motion[24-26]. Because the AHA and PUFI are used to assess age-specific activities, we assumed that children without a disability would receive a score of 100 points.

To find out which specific activities were difficult for children with longitudinal radial deficiency to perform, we studied the individual activities that were applicable to >50% of the children. We considered activities to be easy to perform when they were given a score of "no difficulty" or "some difficulty" and difficult to perform when the score was "great difficulty," "with help," or "cannot do."

Differences in hand function and activity performance among children with different types of longitudinal radial deficiency were tested with use of analysis of variance. In these analyses, we expressed grip and pinch strength as a percentage of reference values in order to correct for age. We calculated the Pearson correlation coefficient (r_p) to determine the relationship between hand function and activity performance. Relationships were considered significant when the p value was <0.05.

Results

Grip and Pinch Strength and Range of Motion

Two children were unable to perform the power grip, and six children without a thumb were not able to perform the tripod pinch. Table II presents the results of grip and pinch-strength testing and the active ranges of motion of the wrist and the metacarpophalangeal and proximal interphalangeal joints of the second digit. Compared with reference values, children

TABLE II Grip and Pinch Strength and Active Ranges of Motion

	Mean and Standard Deviation					
	Total Group	Type I	Type II	Type III	Type IV	P Value
Grip strength						
Newtons	34.9 ± 23.0	44.6 ± 18.0	47.4 ± 34.6	14.7 ± 5.6	19.6 ± 6.0	0.01
% of normal	36 ± 23	54 ± 22	36 ± 18	17 ± 3	18 ± 6	
Pinch strength						
Newtons	11.3 ± 5.2	14.2 ± 5.8	11.4 ± 5.4	7.0	7.8 ± 1.7	0.09
% of normal*	30 ± 15	41 ± 16	26 ± 12	—	20 ± 9	
Active range of motion						
Wrist						
Degrees	114.1 ± 30.6	137.9 ± 20.6	131.9 ± 23.8	96.3 ± 8.8	85.1 ± 17.2	0.001
% of normal	95 ± 26	115 ± 17	110 ± 20	80 ± 4	71 ± 14	
2nd digit metacarpophalangeal joint						
Degrees	82.4 ± 39.0	112.9 ± 31.2	96.3 ± 44.8	54.5 ± 13.4	52.0 ± 16.2	0.006
% of normal	72 ± 34	98 ± 27	84 ± 39	47 ± 12	45 ± 14	
2nd digit proximal interphalangeal joint						
Degrees	66.6 ± 37.6	99.4 ± 2.0	80.8 ± 29.4	52.5 ± 67.2	29.7 ± 17.8	<0.001
% of normal	67 ± 38	99 ± 2	81 ± 29	53 ± 67	30 ± 16	

*The three four-year-old children were excluded because of missing normative values.

with longitudinal radial deficiency showed reduced grip and pinch strength (36% and 30% of normal strength) and a moderately reduced active range of motion of the metacarpophalangeal and proximal interphalangeal joints of the second digit (72% and 67% of normal motion). The average flexion-extension arc of the wrist was 95% of normal. The average range of motion of the second digit was 149.0° ± 69.2°. Children with a more severe type of longitudinal radial deficiency had less grip strength ($p = 0.01$) and less mobility of the wrist ($p = 0.001$), the second metacarpophalangeal joint

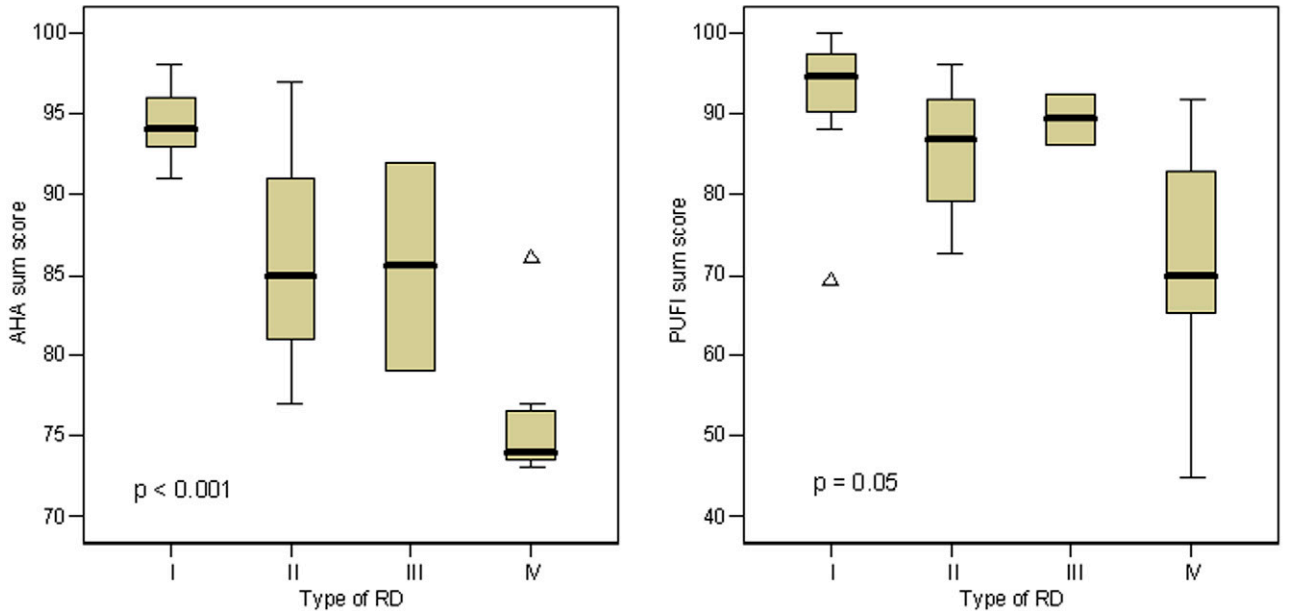

Fig. 1

Sum scores of the AHA (effectiveness) and the PUFI (ease of performance) and their relationships with the type of radial deficiency (RD). Medians (bold lines) with the lower and upper quartiles (lower and upper edge of the boxes) and the range (whiskers) are presented; the triangles represent outliers.

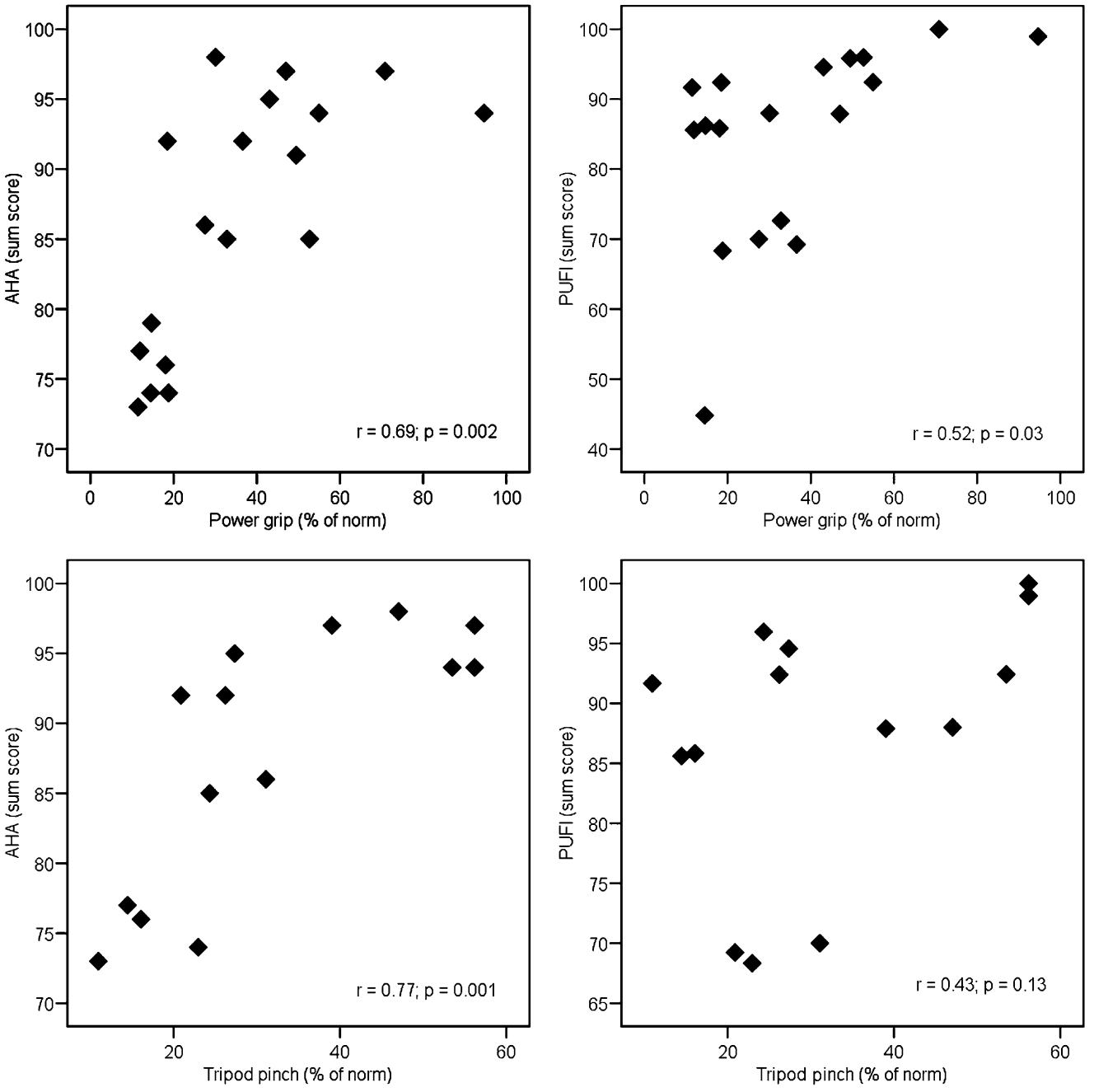

Fig. 2-A

Scatterplots showing the power grip and tripod pinch strength (as percentages of normal) (Fig. 2-A) and the active ranges of motion (AROM) of the wrist and fingers (in degrees) (Fig. 2-B) in relation to the sum scores on the AHA and PUFI. r = Pearson correlation coefficient.

($p = 0.006$), and the second proximal interphalangeal joint ($p < 0.001$).

Functional Activities

The AHA score for the effectiveness of the affected hand was 85.5 ± 9.2 points, and the PUFI score for ease of performance was 81.8 ± 14.6 points. Figure 1 shows that children with a more severe type of longitudinal radial deficiency used the hand less effectively ($p < 0.001$) and had more difficulty in performing activities ($p = 0.05$). Regardless of pollicization, the hand was used the least effectively for the AHA items of "moves forearm," "varies type of grasp," "manipulation," and "moves fingers," particularly by children who had a more severe type of longitudinal radial deficiency. For these items, 45%, 40%, 35%, and 30% of all participating children, respectively, used the hand ineffectively. The three most difficult activities on the younger-child

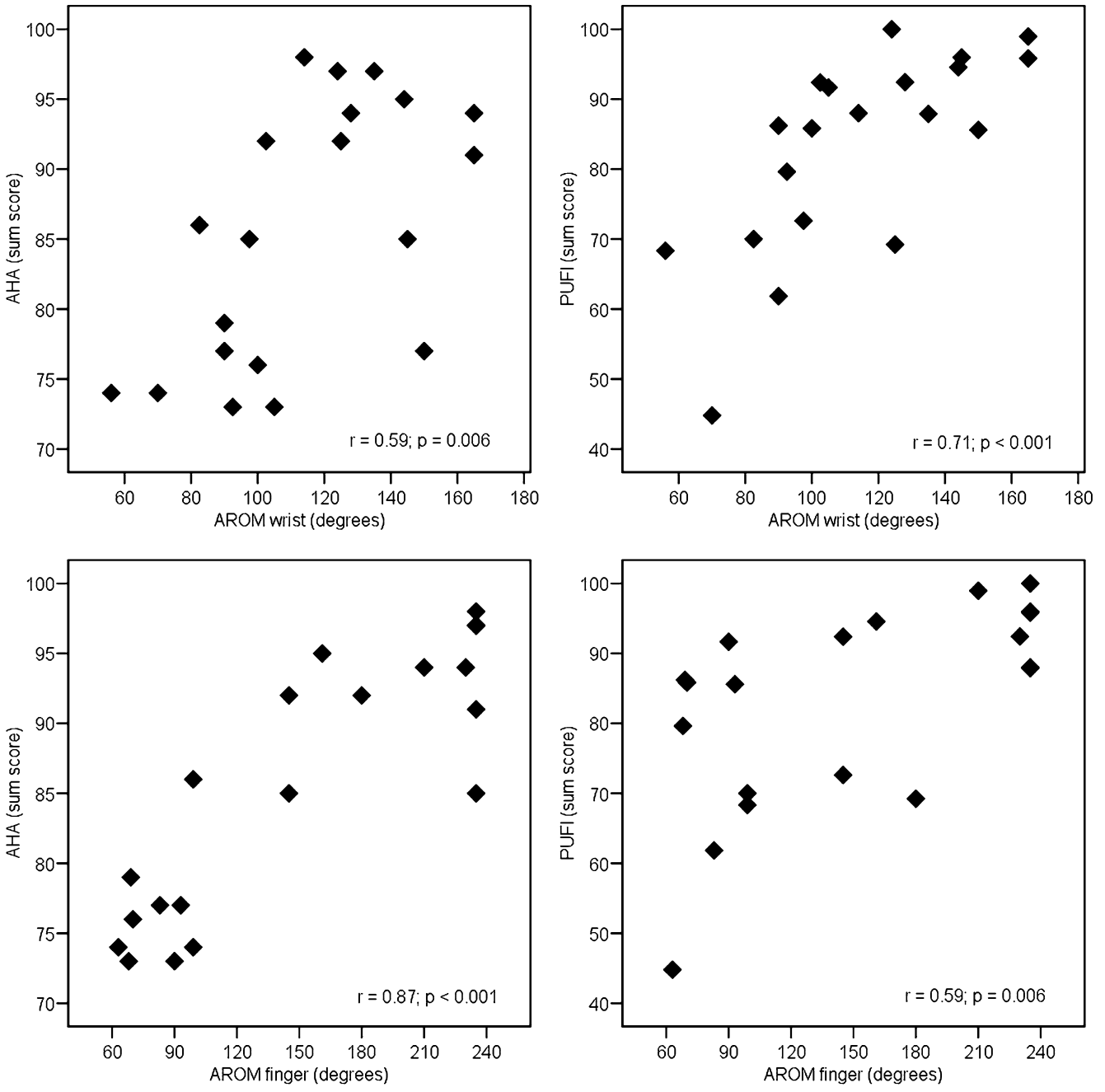

Fig. 2-B

version of the PUFI were "button up shirt," "spread jam on cracker," and "remove clothes from doll"; 71%, 56%, and 43% of the younger children experienced difficulties with these activities. Of the older children, 44% had difficulty with buttoning a shirt; 36%, with putting on mittens; and 33%, with cutting meat.

Relationship Between Hand Function and Activity Performance

Most measures of strength and active range of motion correlated significantly with the performance of activities. Grip strength correlated significantly with activity performance as measured with both the AHA ($r_p = 0.69$, $p = 0.002$) and the PUFI ($r_p = 0.52$, $p = 0.03$). Pinch strength correlated significantly with the AHA score ($r_p = 0.77$, $p = 0.001$), but not with the PUFI score ($r_p = 0.43$, $p = 0.13$) (Fig. 2-A). The active range of motion of both the wrist and the second digit correlated significantly with the AHA score ($r_p = 0.59$, $p = 0.006$, and $r_p = 0.87$, $p < 0.001$) and the PUFI score ($r_p = 0.71$, $p < 0.001$, and $r_p = 0.59$, $p = 0.006$) (Fig. 2-B). Scatterplots of the relationships between grip and pinch strength and functional

activities showed a nonlinear pattern. The scatterplots showed that children with high grip or pinch strength performed the functional activities well. However, while some children with low grip or pinch strength also had low scores for functional activities, others with low strength performed functional activities as well as the children with greater strength. Scatterplots of the relationships between the active ranges of motion of the wrist and finger joints and functional activities showed a more linear pattern, indicating that children with greater active ranges of motion of the wrist and finger joints had higher scores for functional activities.

Discussion

In this study of children with longitudinal radial deficiency, we measured hand function at two levels of the International Classification of Functioning, Disability and Health[3]: the function level and the activity level. We assessed the function level by measuring grip and pinch strength and the active ranges of motion, and we measured activity performance with a function test and a questionnaire. We found reduced grip and pinch strength and less mobility of the metacarpophalangeal and proximal interphalangeal joints of the second digit compared with the published normative values[24-26]. Manske et al. also found reduced strength and joint mobility in children with longitudinal radial deficiency[12]. They reported grip and pinch-strength values that were, respectively, 21% and 23% of normative values, which are lower than our average values but comparable with the 18% and 20% that we found for children with type-IV longitudinal radial deficiency. The active ranges of motion of the metacarpophalangeal and proximal interphalangeal joints of the second digits in the present study, except for the proximal interphalangeal joints of children with type-IV longitudinal radial deficiency, were greater than the ranges found by Manske et al. Goldfarb et al. found that the average total active range of motion per digit of children with type-III or IV longitudinal radial deficiency was 42% of normal[27], a value that was comparable with that of the children with type-III or IV deficiency in our study. Despite these deficits in grip and pinch strength and active range of motion, particularly in children with the more severe type of deficiency, children with longitudinal radial deficiency generally performed most activities rather easily, as indicated by high sum scores on the AHA and the PUFI. Even though the overall activity scores were relatively high, children with longitudinal radial deficiency had difficulty with a number of specific activities, such as buttoning a shirt, spreading jam on a cracker, putting on mittens, or cutting meat.

Despite significant linear correlation coefficients between grip and pinch strength and activity performance, the scatterplots in Figures 2-A and 2-B showed a nonlinear pattern. This pattern indicates a large variation in the activity performance of children with low grip or pinch strength, and a good activity performance by children with high grip or pinch strength. Although the small sample size in the present study does not allow us to draw strong conclusions regarding this point, the similarity between the scatterplots for strength and activity performance suggests that low grip and pinch strength does not always accompany poor activity performance. This finding may imply that high levels of grip and pinch strength are not prerequisites for the performance of functional activities. In future studies, it would be worthwhile to investigate whether other functions, such as a logarithmic function, might better describe these relationships. In fact, in a preliminary analysis, we found that this may be true for the association between grip and pinch strength and the AHA score, but this finding was not consistent for the other parameters. The nonlinearity of the relationships between grip and pinch strength and activity performance stresses the importance of assessing both aspects of hand function to perform an adequate evaluation.

While the scatterplots of strength and functional activities indicate a nonlinear pattern, the relationships between joint mobility and functional activities seemed to follow a more linear pattern. This suggests that improving mobility throughout the whole range of motion might have positive effects on the performance of functional activities. Longitudinal studies, including measurements of finger stiffness, should confirm whether improving or maintaining the ranges of motion of the wrist and fingers would have positive effects on activity performance.

The present study had some limitations. Since the incidence of children with longitudinal radial deficiency is low, the results of the present study are based on a small number of patients. Moreover, children with longitudinal radial deficiency are a heterogeneous group that includes several types of longitudinal radial deficiency, unilaterally or bilaterally affected children, and children who have undergone different types of surgical interventions. However, the purpose of this study was not to describe the outcome of surgery, but to explore the relationship between impairments in hand function and activity performance. Therefore, in our opinion, the heterogeneity of the study sample did not weaken our conclusion that the relationship between impairments in hand function and activity performance is not linear. Future studies with larger sample sizes are needed to study the influences of bilateral involvement, absence or deficiency of the thumb, and mental and physical agility.

Although the measurement instruments used in the present study were carefully selected, some comments should be made. Despite the difficulties that younger children have with handling the Lode grip and pinch dynamometers, we chose to use these devices since they are often used in clinical practice and have been found to be reliable in other patient groups[21]. At present, no other valid and reliable instruments are available to measure grip and pinch strength of children in clinical practice[28]. Furthermore, because we studied bilaterally affected children with instruments developed for children with neurological and unilateral conditions, our results should be interpreted with caution. However, we chose to use the AHA and PUFI to assess the activity performance of children with longitudinal radial deficiency because we had found evidence of their validity and test-retest reliability for the evaluation of children with that disorder[19]. Since both the AHA and the PUFI are generic instruments, we might have missed specific prob-

lems of children with longitudinal radial deficiency. For example, because of a reduced arm length, children with longitudinal radial deficiency may have problems with cleaning themselves after using the toilet. Also, adequate opposition of the thumb may be a problem. The additional use of patient-specific indices might be useful to objectively evaluate these specific problems[29].

In conclusion, the results of the present study suggest that the relationship between impairments in hand function and limitations in activities is not linear. We therefore recommend that evaluations of the results of treatment include assessment of both function and activity. Future studies of larger samples are necessary to provide detailed insight into the relationship between the two aspects of hand function and into the performance by specific subgroups of children with different types of longitudinal radial deficiency. ■

Laurien M. Buffart, MSc
Marij E. Roebroeck, PhD
Wim G.M. Janssen, MD
Anneke Hoekstra, PT
Ruud W. Selles, PhD
Steven E.R. Hovius, MD, PhD
Henk J. Stam, MD, PhD, FRCP
Departments of Rehabilitation Medicine
(L.M.B., M.E.R., W.G.M.J., A.H., R.W.S., and H.J.S.)
and Plastic and Reconstructive Surgery
(R.W.S. and S.E.R.H.),
Erasmus MC,
University Medical Centre Rotterdam,
P.O. Box 2040,
3000 CA Rotterdam,
The Netherlands.
E-mail address for L.M. Buffart: l.buffart@erasmusmc.nl

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