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Functional Assessment of Children and Adolescents with Symbrachydactyly

A Unilateral Hand Malformation

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Investigation performed at Shriners Hospitals for Children-Northern California, Sacramento, California, Texas Scottish Rite Hospital for Children, Dallas, Texas, Karolinska Institutet, Stockholm, Sweden, Children's Hospital of Los Angeles, Los Angeles, California, Lucile Packard Children's Hospital Stanford, Palo Alto, California, and Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio

Background: We studied children and adolescents with symbrachydactyly to determine whether hand function depends on digit opposability and whether scores for function and quality-of-life measures differ from population norms.

Methods: Participants were grouped on the basis of hand morphology: Group A lacked opposable digits, and Group B had ≥2 digits that were opposable. The groups were compared with each other and with norms with respect to pinch strength, the performance of bimanual activities and in-hand manipulation, and questionnaires regarding psychosocial status and the ability to perform activities of daily living (ADLs). Participants and parents also rated the appearance and function of the hand.

Results: Pinch strength was higher for participants in Group B (4.1 compared with 2.4 kg; $p = 0.008$), but the groups did not differ with respect to the proportion of participants outside of pinch norms. Participants in Group B were more likely to actively use their affected hand to perform bimanual activities ($p \leq 0.0009$), and to use normal or supination strategies to accomplish in-hand manipulation ($p = 0.031$). The groups did not differ in the proportion of ADLs rated "difficult" or "impossible," and both groups tested within normal limits for psychosocial function. Participants from both groups and their parents rated their satisfaction with hand appearance and function similarly high.

Conclusions: Participants with ≥2 opposable digits incorporated their hand better in bimanual activities and used more effective strategies to accomplish in-hand manipulation than those who did not. These groups reported no difference in the ability to perform ADLs or with psychosocial function, which was within the normal range. Children and adolescents with symbrachydactyly demonstrated and reported a high level of function in all domains of validated function tests. This study provides information to help parents of children with a unilateral hand malformation understand their child's potential function, and assist surgeons with recommending treatment.

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

Peer Review: This article was reviewed by the Editor-in-Chief and one Deputy Editor, and it underwent blinded review by two or more outside experts. The Deputy Editor reviewed each revision of the article, and it underwent a final review by the Editor-in-Chief prior to publication. Final corrections and clarifications occurred during one or more exchanges between the author(s) and copyeditors.

Symbrachydactyly is a typically unilateral hand malformation characterized by the failure of fingers to form, usually in the central hand plate, with the presence of rudimentary nubbins. Morphologies range from aphalangic to short-finger type, with the middle of the spectrum consisting of absent central digits and relative sparing of the border digits[1]. Symbrachydactyly may be associated with underdevelopment of the ipsilateral sternal portion of the pectoralis major and other structures of the chest wall (Poland syndrome)[2,3]. According to the Oberg, Manske, and Tonkin (OMT) classification of congenital hand malformations, symbrachydactyly is a failure of axis formation and

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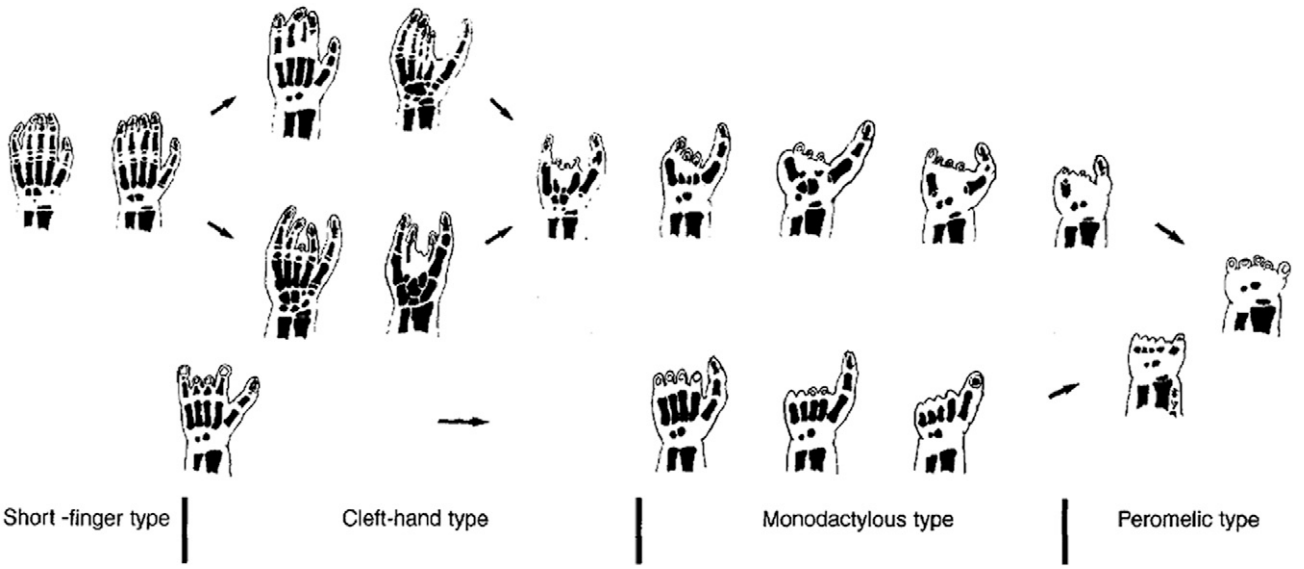

Fig. 1

The teratologic sequence of symbrachydactyly. An idea proposed by Muller in 1937 and adapted by Buck-Gramcko for symbrachydactyly[1]. The participants in the current study demonstrated the "cleft-hand" and "monodactylous" types. Children with "short-finger" and "peromelic" types were excluded. (Reproduced, with permission, from Buck-Gramcko D. Congenital malformations of the hand and forearm. Chir Main. 2002 Mar;21[2]:70-101. Copyright Elsevier Masson SAS. All rights reserved.)

differentiation of the hand plate (or, if it includes chest-wall anomalies, of the entire upper limb)[4]. It occurs in approximately 0.6 per 10,000 live births, predominantly in males (73%), and is more common on the left side (67%). Bilateral involvement, associated anomalies (other than chest wall), and a positive family history are all rare[5].

The description of function for people with symbrachydactyly can be conceptualized within the World Health Organization's framework of functional levels: body structure, activities, participation, and quality of life[6]. Interventions for symbrachydactyly are often based on the morphology of the affected hand (Fig. 1). For instance, for a child with a hand with either 5 short mobile digits or aphalangia, management focuses on activity training and supporting the development of healthy self-esteem. The child with a hand in the middle of the morphologic spectrum may benefit from surgery to improve opposition, grip strength, or digit mobility or stability, such as syndactyly release, web-space deepening, joint fusion, nubbin excision, phalanx lengthening, toe phalanx transfer, and neurovascular transfer of a toe or toes[7]. However, the functional benefit of these procedures is difficult to estimate, as the function of children with symbrachydactyly has not been well described.

Our primary hypothesis was that hand function in symbrachydactyly is based on the status of the border digits; specifically, that 2 stable, flexible, and opposable digits are necessary and sufficient for most functional tasks. Our secondary hypothesis was that children and adolescents with symbrachydactyly will show results similar to age-matched population norms on quality-of-life and hand-function questionnaires and that children and their parents will have similar opinions about the appearance and function of the child's affected hand.

TABLE I Demographic and Hand Characteristics of Participants

	Group A, N = 27	Group B, N = 26
Age* *(yr)*	12.5 ± 4.3	11.4 ± 3.5
Sex *(no.)*		
Male	11	17
Female	16	9
Affected hand *(no.)*		
Left	19	19
Right	8	7
Unaffected hand dominant *(no. [%])*	27 (100%)	26 (100%)
Poland syndrome *(no. [%])*	3 (11%)	6 (23%)
Left hand	1	3
Right hand	2	3
Prior hand surgery *(no. [%])*	5 (19%)	10 (38%)

*The values are given as the mean and the standard deviation.

Materials and Methods

We gathered information on body structure, upper-extremity activities of daily living (ADLs), and psychosocial domains for children with unilateral symbrachydactyly who had monodactyly or ≥2 digits that were non-opposable and/or were unstable (Group A) and those who had ≥2 digits that were opposable and stable (Group B).

TABLE II Previous Surgical Reconstructive Procedures

Group A, N = 5	Group B, N = 10
Nubbin excision	Fusion of small finger proximal interphalangeal (PIP) joint, ring finger distal phalanx excision
1st web-space deepening, toe phalanx transfer to thumb	Syndactyly release
1st web-space deepening	1st web-space deepening and thumb rotational osteotomy
Nubbin excision	1st web-space deepening
Opponensplasty using abductor digiti minimi tendon transfer	Syndactyly release
	Nubbin excision and 1st web-space deepening
	2nd toe microvascular transfer
	Nubbin excision and mass removal from small finger
	Web-space deepening
	Syndactyly release

Study Participants

This study involved 53 participants (28 boys and 25 girls) with unilateral symbrachydactyly from 6 children's hospitals. The average age of the participants was 12 years (Table I). Participants were recruited from clinic appointments and by telephone and mail between August 2014 and December 2015, after institutional review board approval was obtained at each site. An additional 15 potential subjects were invited to participate but were unable to do so (7), did not give a reason for not participating (7), or did not want to participate (1).

Inclusion Criteria

Enrolled participants were children and adolescents with unilateral symbrachydactyly aged 6 to 20 years. Because surgical reconstruction is frequently performed before children with symbrachydactyly are old enough for validated function tests, and because, for the purpose of this study, subjects were classified on the basis of their digit opposability and stability regardless of prior treatment, 15 children who had previously undergone surgical reconstruction were included (Table II).

Exclusion Criteria

Participants were excluded from the study if they did not understand English, Spanish, or Swedish, if they had comorbidities that limited function, or if they had 5 mobile digits or no fingers on their affected hand.

Study Design

This was a cross-sectional, multicenter study.

Function Tests

Joint stability was measured on a 3-point scale: (1) stable to slight laxity, (2) laxity with a firm end point, and (3) unstable with no firm end point. Opposition of the border digits was classified on a 4-point scale: (1) monodactyly, (2) ≥2 digits but non-opposable, (3) ≥2 digits with opposition but not pad-to-pad contact, and (4) ≥2 digits with pad-to-pad opposition. Pinch strength was measured with a calibrated pinch meter with 3 trials in the pinch position of the participant's choice.

The Modified House Test (MHT) was used to assess the incorporation of the affected hand into bimanual tasks, which included opening a drawstring bag, opening a specimen jar, removing a marker cap, pulling a tape measure, and unfolding a piece of paper. The performance on each activity was scored on a 0 to 5 scale: (0) does not use, (1) poor passive assist, (2) passive assist, (3) poor active assist, (4) active assist, and (5) spontaneous use[8]. MHT performance was video-recorded for scoring purposes.

In-hand manipulation and unilateral function were measured using the Functional Dexterity Test (FDT)[9]. The FDT is a timed test in which participants turn over 16 pegs in a square board in a specific order using a single hand (Fig. 2). The test is intended to be performed according to specific instructions, including not using the board or supinating the forearm to assist in peg turning. If participants were unable to invert the peg according to these specifications, they were allowed to use spontaneously generated adaptive strategies and were categorized according to their strategy: normal, board-assisted, or supination. The time used to accomplish this test was not analyzed, as it was not likely to be valid considering the adaptive strategies allowed.

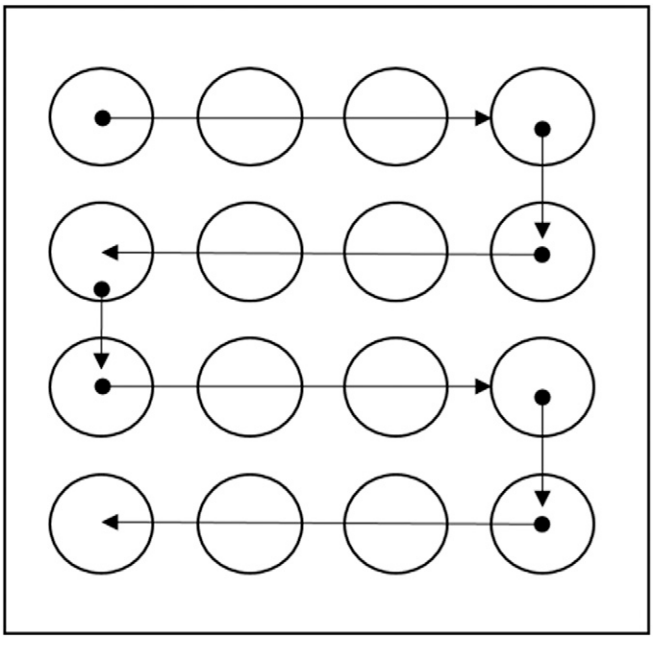

Fig. 2

The pattern of peg turning for the Functional Dexterity Test with the right hand. The participant starts at the top left corner, turns 4 pegs, moves down a row, and continues to zig-zag across the pegboard.

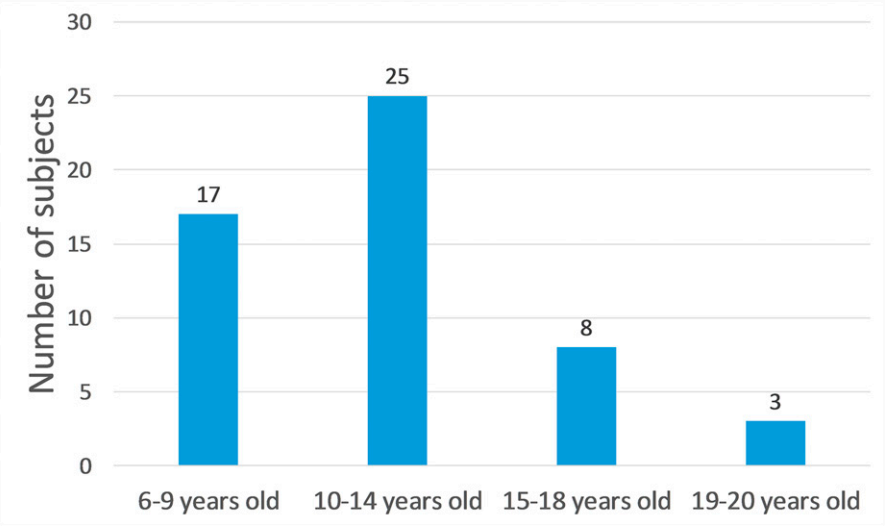

Fig. 3

The age distribution of the 53 participants in the study.

Questionnaires

The ABILHAND-Kids questionnaire asks the child and parent to rate each of 21 ADLs that typically require bimanual upper-extremity function as "impossible," "difficult," "easy," or "?" ("?" indicates that the subject had not attempted the task in the last 3 months or the parent was unsure of his or her child's ability to perform it; "?" responses were not included in the analysis of ABILHAND-Kids results). Participants >18 years of age completed the questionnaire independently. Each ADL has a difficulty ranking: (1) purely unimanual, (2A) achievable with unimanual steps, (2B) requiring stabilization with contralateral digit activity, or (2C) requiring bimanual digit activity[10].

The Quality of Life in Neurological Disorders (Neuro-QoL) pediatric short-form questionnaires assess the psychosocial domains of stigma, depression, anger, and interaction with peers. Participants score how often each of 8 statements per domain describes their feelings on a 5-point scale: (1) never, (2)

Fig. 4-A

Fig. 4-B

Pinch demonstrated by a participant in Group A who is unable to oppose (**Fig. 4-A**) and a radiograph of the same participant (**Fig. 4-B**).

TABLE III Distribution by Tip Pinch Assessment

Tip Pinch	Group A, N = 27	Group B, N = 26	Total, N = 53
Within or above normal range *(no.)*	13	17	30
Below normal *(no.)*	14	9	23

TABLE IV ABILHAND-Kids Results by Group

		Rating*							
		Impossible		Difficult		Easy		% Answered	
ADL	Difficulty	Group A	Group B	Group A	Group B	Group A	Group B	Group A	Group B
Opening a jar of jam	2B		1 (5%)	10 (42%)	9 (41%)	14 (58%)	12 (55%)	89%	85%
Buttoning trousers	2B	1 (4%)		9 (35%)	9 (38%)	16 (59%)	15 (63%)	96%	92%
Buttoning a shirt/ sweater	2A	1 (4%)		8 (31%)	9 (36%)	17 (65%)	16 (64%)	96%	96%
Rolling-up a sleeve on a sweater	2A			11 (41%)	5 (21%)	16 (59%)	19 (79%)	100%	92%
Unscrewing a bottle cap	2B			5 (19%)	9 (39%)	22 (81%)	14 (61%)	100%	89%
Fastening the snap of a jacket	2A			8 (30%)	5 (22%)	19 (70%)	18 (78%)	100%	89%
Opening a bag of chips	2C			4 (15%)	5 (19%)	23 (85%)	21 (81%)	100%	100%
Zipping-up trousers	2A			6 (22%)	2 (8%)	21 (78%)	24 (92%)	100%	100%
Washing the upper body	1				4 (15%)	27 (100%)	22 (85%)	100%	100%
Sharpening a pencil	2C			4 (15%)		22 (85%)	26 (100%)	96%	100%
Squeezing toothpaste onto a toothbrush	2A			2 (7%)	2 (8%)	25 (93%)	23 (92%)	100%	96%
Filling a glass with water	2A	1 (4%)			2 (8%)	26 (96%)	24 (92%)	100%	100%
Unwrapping a chocolate bar	2A			2 (7%)	1 (4%)	25 (93%)	25 (96%)	100%	100%
Switching on a bedside lamp	1			1 (4%)	1 (4%)	26 (96%)	25 (96%)	100%	100%
Taking a coin out of a pocket	1			2 (7%)	1 (4%)	25 (93%)	24 (96%)	100%	96%
Opening the cap of a toothpaste tube	2A			1 (4%)		26 (96%)	26 (100%)	100%	100%
Taking off a T-shirt	2A			1 (4%)		26 (96%)	26 (100%)	100%	100%
Zipping-up a jacket	2B			1 (4%)	1 (4%)	26 (96%)	24 (96%)	100%	96%
Opening a bread box	2A			1 (5%)	1 (6%)	19 (95%)	17 (94%)	74%	69%
Putting on a backpack/ schoolbag	2A					27 (100%)	26 (100%)	100%	100%
Putting on a hat	2A					27 (100%)	26 (100%)	100%	100%

*The values are given as the number of participants and, in parentheses, the percentage of the total respondents in each group who provided this rating for the specific ADL.

TABLE V Distribution by Score (5 or <5) for Each Component of the Modified House Test (MHT)

MHT Task	No. of Participants		P Value*
	Group A	Group B	
Drawstring			<0.0001
<5	19	2	
5	8	24	
Jar			0.0009
<5	19	6	
5	8	20	
Tape			<0.0001
<5	17	2	
5	10	24	
Paper			<0.0001
<5	19	4	
5	8	22	
Marker			0.0003
<5	15	2	
5	12	24	

*Fisher exact test.

rarely, (3) sometimes, (4) often, or (5) always. For children <10 years of age, parents served as a proxy[11].

Children <18 years of age and their parents completed a 10-point Likert scale regarding the function and appearance of the affected hand.

Participants were grouped on the basis of hand morphology. Group A consisted of children with monodactyly or ≥2 digits that were unstable and/or were not opposable (opposition scores of 1 or 2); Group B included children with ≥2 stable and opposable digits (opposition scores of 3 or 4).

Statistical Analysis

Pinch strength was analyzed by comparing mean participant pinch strength and interquartile range (IQR), after 3 trials, with age and sex-matched tip pinch-strength norms[12,13]. A Fisher exact test was used to test for an association between the groups and whether tip pinch strength was more than 1 standard deviation (SD) outside of population norms. A Wilcoxon test was used to compare pinch strength between the groups.

The association between the group and an MHT score of 5 or <5 for each component of the MHT was evaluated using a Fisher exact test, and a Wilcoxon test was used to test if the average of the 5 MHT scores differed significantly between the groups.

The proportion of "difficult" or "impossible" ABILHAND-Kids tasks was calculated for each participant, and a Wilcoxon test was used to compare the proportions between Groups A and B. Because the 21 ADLs are rated in terms of increasing bimanual involvement, whether the proportion of participants rating a task as "difficult" or "impossible" differed on the basis of ADL rating and group was evaluated[10]. A Wilcoxon test was used to compare the proportion of participants rating a task as "difficult" or "impossible" between groups A and B.

A Fisher exact test was used to test associations between FDT strategy and group.

Parental and child scores for appearance and function were compared between the groups and between children and parents within each group, using Wilcoxon rank sum tests. A Bonferroni adjustment was used to control the type-I error rate.

Results

Morphology and Opposability

Hand morphology, laterality, hand dominance, age distribution, and Poland syndrome prevalence in our sample are summarized in Table I and Figure 3. On the basis of opposability and digit stability, 27 (51%) of the participants were categorized as Group A (Figs. 4-A and 4-B) and 26 (49%), Group B.

Hand Function

Pinch strength averaged over 3 trials differed significantly between the groups (2.4 kg [IQR, 1.7 to 3.4 kg] for Group A and 4.1 kg [IQR, 2.4 to 4.5 kg] for Group B; p = 0.008). Three participants from Group A could not pinch the meter: 2 had non-opposable border digits, and 1 had a single, unstable digit. Table III shows the number of participants who scored below normal tip pinch values for their age and sex. The groups did not differ significantly with respect to the proportion of participants whose pinch was weaker than normal (p = 0.27). Five participants (1 from Group A and 4 from Group B) exceeded the norm by 1 SD. The unaffected hand was dominant for all 53 participants.

Twenty (77%) of the participants in Group B had pad-to-pad opposition. In the remaining 6 (23%) of the participants, the border digits were opposable but could not achieve pad-to-pad contact. Three participants in Group A had non-opposable border digits (2 with stable digits and 1 with an unstable ulnar digit); the remainder of the participants in Group A had monodactyly or unstable border digits.

Bimanual Function

ABILHAND-Kids responses are shown in Table IV. Groups A and B did not differ significantly with respect to the proportion of ADLs rated "difficult" or "impossible" (p = 0.85). The groups were not differentiated by the complexity of hand involvement of ADLs (purely unimanual [difficulty = 1], p = 0.398; achievable with unimanual steps [difficulty = 2A], p = 0.347; requiring stabilization with contralateral digit activity [difficulty = 2B],

TABLE VI Distribution by Functional Dexterity Test (FDT) Strategy

FDT Strategy	Group A, N = 27	Group B, N = 26	Total, N = 53
Board-assist *(no. [%])*	22 (81%)	14 (54%)	36 (68%)
Supination *(no. [%])*	5 (19%)	8 (31%)	13 (25%)
Normal *(no. [%])*	0 (0%)	4 (15%)	4 (8%)

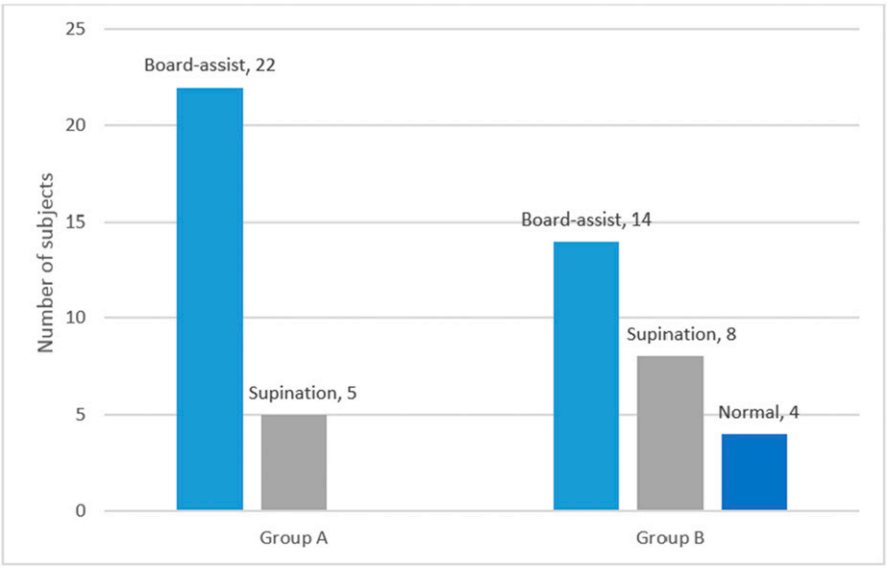

Fig. 5

Functional Dexterity Test (FDT) strategies used by both groups. Participants in Group A (presence of monodactyly or ≥2 digits that are non-opposable and/or unstable) predominantly used a strategy of board assistance (n = 22) in completing the FDT, followed by supination (n = 5). In Group B (presence of ≥2 stable and opposable digits), a majority of participants also used a board-assist strategy (n = 14), but Group B had a nearly equal proportion of those who used supination (n = 8) or the normal strategy (n = 4).

p = 0.55; or requiring bimanual digit activity [difficulty = 2C], p = 0.49). Only 2 (4%) of 54 parents marked a total of 4 ADLs as impossible (opening a jar of jam [2B], buttoning a shirt or sweater [2A], buttoning up trousers [2B], and filling a glass with water [2A]) for 1 child from each group, aged 11 and 13.

Group B participants were significantly more likely to score higher than Group A participants for each MHT task (score of 5 versus <5), and the 2 groups also showed significantly different median values for the averaged MHT scores ($p < 0.0001$), with participants from Group B scoring higher (Table V).

Unilateral Function

FDT strategies are shown in Table VI and Figures 5 through 8. Group A participants predominately used board-assist (22 of 27

Fig. 6

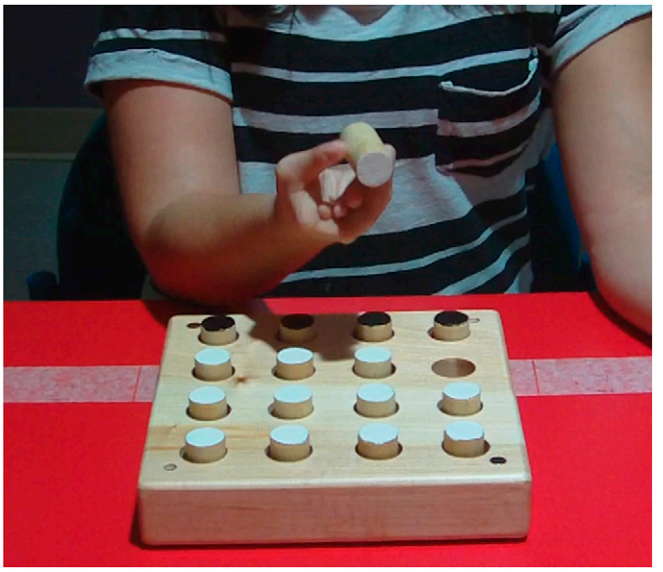

Fig. 7

Fig. 6 A participant completing the Functional Dexterity Test using the board-assist strategy. The edges of the peg hole and the board are used to stabilize the peg while it is flipped. **Fig. 7** A participant completing the Functional Dexterity Test using the supination strategy. The participants generally picked up the peg using their border digits or thumb and palm, and then used supination and gravity to flip the peg.

Fig. 8

A participant completing the Functional Dexterity Test using the normal strategy. Participants who used the normal strategy had ≥3 stable and opposable digits. They were able to stabilize and flip the peg without supination or board assistance.

participants, 81%) or supination (5 participants, 19%) strategies. Participants in Group B were more likely to use the normal or supination strategies ($p = 0.031$). In Group B, 14 (54%) of the 26 participants used the board-assist strategy, 8 (31%) used supination, and 4 (15%) used normal in-hand manipulation. These 4 participants each had ≥3 digits that were stable, mobile, and opposable (a subgroup of Group B).

Psychosocial Assessment

Neuro-QoL results for stigma, depression, anger, and peer relationships were within 1 SD of population norms for 52 (98%) of the 53 participants. No participant in Group A tested outside of the normal range; 1 participant in Group B was scored by her parent outside of population norms for all domains.

Median and quartile values for ratings of appearance and function by group are shown in Table VII. There were no significant differences between Groups A and B in the median participant or parent scores. Although, on the basis of raw p values, Group B participants rated their hand appearance significantly higher than did their parents ($p = 0.044$), this difference did not remain significant after the adjustment for multiple tests ($p = 0.176$).

TABLE VII Child and Parent Likert Scores*

	Group A	Group B	Overall
Appearance			
Child	8 (6, 10)	8 (7.25, 10)	8 (6, 10)
Parent	8 (6.5, 10)	8 (5, 9)	8 (6, 10)
Function			
Child	8 (8, 9)	8 (6, 10)	8 (6, 10)
Parent	8 (6, 9.5)	8 (8, 10)	8 (7, 10)

*The values are given as the median score, with the 25th and 75th quartile values, respectively, in parentheses.

Discussion

This study sought to evaluate function in several domains, including psychosocial well-being, for children and adolescents with symbrachydactyly. We classified participants on the basis of the presence of at least 2 stable, opposable digits (Group B) compared with those with monodactyly or multiple digits that were either unstable or non-opposable (Group A). We included children with previous surgical treatment, and accounted for this by grouping them according to opposability and stability, regardless of whether they had previously undergone surgery.

Assessing pinch strength was challenging because of participants' unique hand morphology and atypical pinch position. Since we considered pinch strength a surrogate for effectiveness of prehension, we measured it in the position chosen by the participant in order to achieve the highest force. We compared this to tip pinch norms because the position of pinch was generally more similar to this than to palmar or key pinch. Fifty-two percent of the Group A participants (14 of 27) and 35% of the Group B participants (9 of 26) had lower-than-normal pinch strength; the difference between the groups was not statistically significant. We did not assess other important types of prehension, such as the ability to grasp items of varying sizes, because most children with this condition will default to the contralateral, normal hand for activities requiring adaptable grasp strength.

The MHT showed that Group B participants were more likely to have normal bimanual function. This may represent the improved biomechanics associated with stable, opposable border digits that could not be detected by pinch strength alone. Group A participants were more likely to use their affected hand passively, but they were able to complete most tasks with adaptation (e.g., using their body or the table as a stabilizer). Since the MHT differentiated between children with and without the ability to oppose, it could serve as a study tool for preoperative and postoperative functional assessment in patients undergoing surgical procedures intended to achieve opposition.

The most complex hand function that this study aimed to measure was in-hand manipulation, using the FDT, a validated test of dexterity. There was a floor effect when participants tried to complete the FDT by the rule set, which disallowed the use of supination or the board. We therefore chose to qualitatively assess their adaptive strategy to turn the pegs. A pattern of strategies emerged, with increasingly complex hand function from board-assist to supination and, finally, normally. Group A participants were more likely to use the board-assist strategy, since they were presumably unable to manipulate in-hand. As with the MHT, the FDT could serve as a useful test to measure the effect of surgery on hand dexterity. Only participants with ≥3 mobile, stable, and opposable digits were able to complete the FDT in a normal fashion, as the FDT is largely reliant on tripod pinch for successful completion. Our study had only a small sample ($n = 4$) of these participants, but future, larger, studies of these hand types may yield a finer delineation of the functional and/or psychosocial benefits that this improved dexterity may impart.

The ABILHAND-Kids questionnaire showed no difference between the groups in the proportion of ADLs marked "difficult" or "impossible." It is not surprising that children and adolescents with symbrachydactyly find some ADLs difficult. Nevertheless, only 2 parents (of 1 participant in each group), marked a total of 4 ADLs as "impossible" (opening a jar of jam, buttoning a shirt or sweater, buttoning up trousers, and filling a glass with water). However, because the ABILHAND-Kids is completed by parents, it may not truly represent the child's abilities. In a previous study of children with unilateral hand malformation (below-the-elbow deficiency), the senior author (M.A.J.) showed that parents tend to underestimate their child's upper-extremity physical function compared with child self-reports[14]; it is possible that parents with symbrachydactyly also underestimate their child's abilities.

Fifty-two of 53 children tested within the normal range for psychosocial status. Since the Neuro-QoL was originally developed for neurological conditions[11], we modified a single word in the test (changing "illness" to "my hand") to make the questions more understandable to participants. Because it was designed for conditions that are generally more debilitating than symbrachydactyly, it may have a ceiling effect in this population. Our finding that most children with a unilateral hand malformation have normal psychosocial well-being is consistent with a previous study of children with unilateral below-the-elbow deficiency by the senior author, which found nearly normal scores on a quality-of-life index[15].

Likert ratings of both participants' and parents' opinions on the function and appearance of the affected hand were high for both groups. In combination with the Neuro-QoL data, we believe that this represents high levels of self-acceptance. Children in Group B rated their hand's appearance higher than did their parents, although the significance of this finding did not remain after adjustment for multiple tests. The study mentioned above also showed that parents under-rate quality of life and function compared with children with below-the-elbow deficiency[14].

It is possible that the function of the 15 subjects who were unable or declined to participate differs from that of the participants, a potential limitation of this study.

This study provides information regarding the functional and psychosocial status of children and adolescents with a unilateral hand malformation, and suggests that stable, opposable border digits enable complex hand function, including incorporation of the hand into bimanual activities and the capacity for increased in-hand manipulation. This functional difference did not impart a benefit that was measurable by our psychosocial tests. This study provides information to help parents estimate their child's potential with respect to function and quality of life, and to guide families and clinicians in the selection of treatment options and the measurement of their effectiveness. Our findings may be applicable to children with other unilateral hand anomalies that limit prehension, such as ulnar or radial longitudinal deficiency or brachydactyly. ■

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