Tendon Transfer Surgery in Upper-Extremity Cerebral Palsy Is More Effective Than Botulinum Toxin Injections or Regular, Ongoing Therapy

Ann E. Van Heest, MD, Anita Bagley, PhD, Fred Molitor, PhD, and Michelle A. James, MD

Investigation performed at Shriners Hospitals for Children: Chicago, Illinois; Greenville, South Carolina; Northern California, Sacramento, California; Salt Lake City, Utah; Shreveport, Louisiana; Tampa, Florida; and Twin Cities, Minneapolis, Minnesota

Background: For children with upper-extremity cerebral palsy (CP) who meet standard indications for tendon transfer surgery, we hypothesized that surgical treatment would result in greater functional improvement than treatment with botulinum toxin injections or regular, ongoing therapy.

Methods: Thirty-nine children with upper-extremity CP, who were four to sixteen years of age and surgical candidates for the transfer of the flexor carpi ulnaris to the extensor carpi radialis brevis, pronator teres release, and extensor pollicis longus rerouting with adductor pollicis release, were prospectively assigned, either randomly (twenty-nine patients) or by patient/family preference (ten patients), to one of three treatment groups: surgical treatment (Group 1); botulinum toxin injections (Group 2); or regular, ongoing therapy (Group 3). Seven centers participated. Assessment measurements included active range of motion, pinch and grip strength, stereognosis, and scores as measured with eight additional functional or patient-oriented outcome instruments. Thirty-four patients (twenty-five randomized and nine from the patient-preference arm) were evaluated twelve months post-treatment as the study cohort.

Results: For the primary outcome of the Shriners Hospital Upper Extremity Evaluation (SHUEE) dynamic positional analysis (DPA), significantly greater improvement was seen in Group 1 than in the other two groups (p < 0.001). Improvements in SHUEE DPA reflected improved supination and wrist extension during functional activities after surgical treatment. Group 1 showed more improvement in the Pediatric Quality of Life Inventory (PedsQL) CP module domain of movement and in the Canadian Occupational Performance Measure (COPM) score for satisfaction than Groups 2 and 3. Both Groups 1 and 3 showed more improvement in pinch strength than did Group 2.

Conclusions: For children with upper-extremity CP who were candidates for standard tendon transfer, surgical treatment was demonstrated to provide greater improvement, of modest magnitude, than botulinum toxin injections or regular, ongoing therapy at twelve months of follow-up for the SHUEE DPA, the PedsQL CP module domain of movement, and COPM satisfaction.

Level of Evidence: Therapeutic Level II. See Instructions for Authors for a complete description of levels of evidence.

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erebral palsy describes a group of permanent disorders in the development of movement and posture, causing activity limitations attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain^{1,2}. Because of abnormal tone patterns, the limb is positioned such that it reflects the imbalance of muscle forces; the most common pattern of upper-extremity positioning is elbow flexion, forearm pronation, wrist flexion, and thumb in palm.

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TABLE I Demographics	TABLE I	Demographics*
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	Surgery	Botulinum Toxin	Therapy	P Value
No. of patients	16	9	9	
Affected side = right	50.0%	66.7%	44.4%	0.61
Stereognosis† (no. of objects)	6.4	4.9	6.6	0.54
CTONI score†	85.2	85.0	82.0	0.86
Female	31.2%	11.1%	55.6%	0.13
Age† (yr)	9.3	9.0	10.2	0.66
Age distribution (no. of patients)				
4-7 yr	6	3	2	
8-10 yr	6	5	3	
11-13 yr	2	0	3	
14-16 yr	2	1	1	

*ANOVA was used to compare age, stereognosis, and CTONI scores among groups. Chi-square testing was used to compare distribution by sex and affected side among groups. †Values are presented as the mean.

The intent of addressing the wrist "palsy" position is to improve both patient self-esteem and hand function. Wrist-flexion positioning substantially interferes with grasp and release function. Surgical and nonsurgical options have been used to reduce deformity. Surgical reconstruction includes the transfer of the flexor carpi ulnaris (FCU) to the extensor carpi radialis brevis (ECRB), which removes the FCU as a spastic wrist flexor and augments wrist-extension power, thus improving wrist position³⁻⁵. Concomitant procedures may include pronator teres release, to decrease the pronation deformity^{6,7}, and release of the spastic adductor pollicis with extensor pollicis longus (EPL) rerouting, to decrease the thumb deformity⁸⁻¹⁰. Standard nonoperative treatment options include the use of botulinum toxin injections and therapy interventions. Botulinum toxin injections have been used for the chemical denervation of spastic muscles, which decreases the magnitude of deforming forces to provide better balance across malpositioned joints¹¹⁻¹⁶. Therapy interventions such as range-ofmotion and strengthening exercises, dexterity training, and splinting are most commonly used. The comparative effectiveness of surgical and nonsurgical treatment options has not been established, which was the impetus for this study.

We hypothesized that children with spastic hemiplegia who met standard clinical indications for FCU to ECRB tendon transfer, pronator teres release, and EPL rerouting with adductor pollicis release would have a greater reduction in impairment, limitations, and restrictions through surgical treatment compared with treatment by either three serial botulinum toxin injections or a standard therapy protocol, as measured with the use of validated functional assessment tools.

Materials and Methods

This multicenter prospective study was designed by a working group of hand surgeons and therapists who met on several occasions prior to initiation of the study to develop the following study design. The working group agreed on treatment of the most common deformity in children with upper-extremity cerebral palsy (CP): forearm pronation, wrist flexion and ulnar deviation, and thumb-in-palm positioning. The standard indications for surgical treatment agreed to were (1) pronation deformity; (2) deficient active wrist extension with adequate digital control, and wrist flexion deformity with the FCU as the primary deforming force; and (3) adduction of the thumb ray with flexion at the thumb metacarpophalangeal (MCP) joint, with the inability to make a fist with the thumb outside the flexed digits. The surgical experience and the clinical judgment of the treating surgeon were used in making these assessments. Surgical treatment included (1) for the loss of passive supination (i.e., pronation contracture), pronator teres tenotomy^{6,7}; (2) for the correction of wrist flexion deformity, FCU to ECRB transfer^{4,5}; and (3) for the correction of thumb deformity, adductor pollicis release and rerouting of the EPL from the third to the first extensor compartment⁸. Children meeting the standard indications for surgical treatment with pronator teres release, FCU to ECRB tendon transfer, and thumb adductor pollicis release with EPL rerouting ("tendon transfer surgery") were potential study participants.

This clinical trial was registered as number NCT00250081 (Comparison of Tendon Transfer, Botox Injections and Ongoing Treatment in Upper-Extremity CP) at ClinicalTrials.gov.

Children with spastic hemiplegic upper-extremity CP who were four to seventeen years of age, whose primary language was Spanish or English, who received treatment at a participating hospital, and who met the standard indications for tendon transfer surgery as described above were eligible for inclusion in the study. Exclusion criteria were a House¹⁷ score of 0 ("Does not use-Extremity not utilized in any capacity for completion of task"), previous upper-extremity surgery, and upper-extremity botulinum toxin injections within the previous twelve months.

Children who met the standard indications for tendon transfer surgery were prospectively randomized into one of three treatment groups (Fig. 1): surgical treatment (Group 1), comprising treatment with tendon transfer surgery and a standard occupational therapy (OT) protocol; botulinum toxin treatment (Group 2), a series of three botulinum toxin injections and the standard OT protocol; or regular, ongoing treatment (Group 3), involving treatment with the standard OT protocol alone.

After obtaining consent and testing dystonia¹⁸, stereognosis¹⁹, and cognitive abilities using the Comprehensive Test of Nonverbal Intelligence (CTONI)²⁰, the participating site conveyed the information to Shriners Hospitals for Children-Northern California, where an independent statistician chose a unique study identification number for each patient from a table of random numbers The Journal of Bone & Joint Surgery - JBJS.org Volume 97-A • Number 7 • April 1, 2015 SURGERY IN UPPER-EXTREMITY CEREBRAL PALSY COMPARED WITH BOTULINUM TOXIN INJECTIONS OR THERAPY



Fig. 1

Study design timeline and outcome assessment tools used. UECP = upper-extremity cerebral palsy, CTONI = Comprehensive Test of Nonverbal Intelligence, FCU = flexor carpi ulnaris, ECRB = extensor carpi radialis brevis, PT = pronator teres, EPL = extensor pollicis longus, and AROM = active range of motion.



Fig. 2

CONSORT (Consolidated Standards of Reporting Trials) flow diagram. PI = principal investigator.

associated with one of the three treatment groups. Because of a high rate of refusal of randomization²¹, a patient-preference option was added in 2010 to allow families who declined randomization to choose their child's treatment.

Thirty-four children with spastic hemiplegia completed the study protocol: sixteen in Group 1, nine in Group 2, and nine in Group 3 (Fig. 2). Four were lost to follow-up or were noncompliant with the protocol, and one failed to meet the inclusion criteria.

Shriners Hospitals for Children Headquarters approved all consents prior to submission to local institutional review boards. Seven participating hospitals obtained institutional review board approval. A study manual, written by the working group, was used to standardize all treatment protocols and outcome measures at all sites. Lead surgeons, therapists, and coordinators met annually to review data collection and maintain consensus. The period of recruitment and follow-up was October 6, 2005, to May 17, 2013.

Outcome Measures

Validated outcomes measures (Fig. 1) were selected on the basis of the World Health Organization International Classification of Functioning, Disability and Health (WHO ICF)²².

Bodily Impairment

Defined as a problem with body function or structure, bodily impairment was measured by assessing active range of motion (ROM), grip and pinch strength, and stereognosis, and by parent and child-reported satisfaction with appearance.

Activity Limitation

Defined as difficulty encountered by an individual in executing a task or an action, activity limitation was measured by the Shriners Hospital Upper Extremity

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Activity Limitation Measure	Surgery			Botulinum Toxin			Therapy			
	N	Mean (Std. Dev.)	95% Confidence Interval	N	Mean (Std. Dev.)	95% Confidence Interval	N	Mean (Std. Dev.)	95% Confidence Interval	P Value
Shuee DPA* (%)	13	21.6 (9.4)††	15.91 to 27.32	8	5.3 (10.0)	-3.08 to 13.58	9	-1.0 (9.8)	-8.55 to 6.55	<0.00
SHUEE SFA (%)	15	6.5 (11.0)	0.37 to 12.56	9	1.0 (8.5)	-5.57 to 7.57	9	5.7 (11.4)	-3.13 to 14.46	0.46
Box and Blocks (no. of blocks)	15	2.0 (5.9)	-1.85 to 4.65	9	-0.22 (7.4)	-5.92 to 5.48	9	0.89 (3.30)	-1.62 to 3.39	0.80
AHA (points)	16	3.1 (5.4)	0.21 to 5.92	9	1.4 (4.6)	-2.08 to 4.97	9	1.4 (2.1)	1.16 to 4.40	0.69

Evaluation (SHUEE) spontaneous functional assessment (SFA) and dynamic positional analysis (DPA), the Box and Blocks test, and the Assisting Hand Assessment (AHA).

Participation Restriction

Defined as a problem experienced by an individual in involvement in life's situations, participation restriction was measured by the Pediatric Outcomes Data Collection Instrument (PODCI-parent); the Pediatric Quality of Life Inventory (PedsQL), both the standard version and the CP module; the Children's Assessment of Participation and Enjoyment (CAPE); and the Canadian Occupational Performance Measure (COPM).

Outcome measurement protocols are described in the Appendix. For each of the outcome measures used, a higher score is better. Therapists administering the assessments at each site were not blinded to the treatment group. However, for the measurement of function with the SHUEE and the AHA, the assessments were administered and videotaped at each site, with the grading of the videos performed by two AHA-certified therapists who were blinded to treatment.

Treatment Protocols

One universal therapy treatment protocol was used for all patients. Each child was fitted with a long opponens splint for daytime use for six weeks, and a hand splint for nighttime resting for six months. All patients received eight standardized therapy sessions. Exercises progressed over the twelve-month program from passive ROM to active ROM, and building to resistive exercises. All of the patients were also prescribed therapy exercises to be performed three times daily at home, with written and videotaped instructions.

Each child in Group 1 was treated with pronator teres release, FCU to ECRB tendon transfer, and adductor pollicis release with EPL rerouting. All participating surgeons agreed to treat all patients with adductor pollicis release without concomitant thenar release. If the patient had an MCP hyperextension deformity, the EPL rerouting insertion was modified to the first metacarpal head to avoid further hyperextension. To avoid excessive loss of wrist flexion, the FCU to ECRB tendon transfer was tensioned so that the wrist rested in neutral positioning. A long-arm cast holding the limb in maximum supination with slight wrist extension and thumb abduction was worn postoperatively for one month. The eight OT sessions were initiated after cast removal, with therapy conducted according to the universal protocol, with the exception of resistive strengthening, which was initiated twelve weeks after surgery.

Each child in Group 2 was treated with a series of three botulinum toxin injections into the pronator teres, the FCU, and the adductor pollicis. Dosing was 0.5 to 1.0 units/kg into the thumb adductor and 1 to 2 units/kg each into the pronator teres and the FCU, diluted at 100 units in 1 mL of normal saline solution. The maximum dose was the lesser of 50 units per site or 12 units/kg. Injections were performed with the child under general anesthesia, using muscle stimulation to verify the site of injection. Therapy sessions 1 to 3 were after the first injection; sessions 4 to 6, after the second injection; and sessions 7 and 8, after the third injection, according to the universal therapy protocol.

Treatment for the children in Group 3 was initiated after enrollment in the study and performed according to the universal therapy protocol.

Statistical Analysis

Activity measurements (defined by the WHO ICF) were chosen as the primary outcomes, as assessed by the AHA, the SHUEE SFA, the SHUEE DPA, and the Box and Blocks test. The AHA²³ was used for a sample-size calculation, with an 8-point change considered meaningful; this indicated that sixteen patients per group were needed. As part of an interim data analysis, a significant change from baseline to twelve months was found with the SHUEE DPA between Group 1 and Groups 2 and 3 (p < 0.01); therefore, the study was ended as the SHUEE DPA was the primary outcome. Using our current SHUEE DPA data, power analysis showed sufficient power (100%) and a required sample size of eight patients per group.

Secondary outcomes included the bodily impairment measures of grip and pinch strength and parent and child-reported satisfaction scores, and the participation-restriction measures of PODCI global, PedsQL, and COPM performance and satisfaction scores.

Participant characteristics were compared among the three treatment groups at baseline using ANOVA (analysis of variance) for age, stereognosis, and CTONI scores, and the chi-square test for distribution by sex and affected side (right or left) (Table I).

Descriptive statistics for all outcome measures at baseline and at the twelve-month follow-up were generated for each treatment group (see Appendix). Changes between baseline and final follow-up were compared across the three treatment groups using ANOVA. ANOVA was selected because none of the demographic characteristics were found to differ by treatment group. To discount the potential confounding effect of sex, the one variable that approached significance at the $p \le 0.10$ level, the analyses were repeated with MANOVA (multivariate ANOVA). The findings for the MANOVA tests mirrored those for the ANOVAs. Results for the primary outcome measures are presented in Table II. Results for the secondary outcomes, post-hoc analysis for multiple comparisons was performed using a Bonferroni correction, with significance determined when $p \le 0.004$, because fourteen comparisons were made.

Source of Funding

This study was supported by a Shriners Hospitals for Children clinical research grant.

Results

Baseline Comparisons

B aseline comparisons of age, stereognosis, CTONI, sex, and affected-side distribution showed that there were no statistically detectable differences among the groups (Table I).

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Comparison of the three treatment groups pre-treatment and twelve months post-treatment on the basis of the Shriners Hospital Upper Extremity Evaluation (SHUEE) Dynamic Positional Analysis (DPA) scores.

Comparison within Treatment Groups

Descriptive statistics for pre-intervention and twelve-month postintervention values are shown for all outcome measures for each of the three treatment groups in a table in the Appendix. Bodily impairment measures showed increased forearm supination, increased wrist extension, decreased wrist flexion, increased grip strength, and increased pinch strength for the surgical treatment group (see Appendix).

Comparisons Among Treatment Groups

Primary Outcomes: Activity Limitations

Group 1 (the surgical treatment group) showed more improvement in the SHUEE DPA from baseline to the twelve-month follow-up than did Group 2 or Group 3 (Table II and Fig. 3). Group 1 had a mean improvement of 21.6% in the SHUEE DPA, from 66% at baseline to 88% at the twelve-month follow-up. Group 2 had a mean improvement of 5.3%, from 61% at baseline to 66% at the twelve-month follow-up. Group 3 had a mean decline of 1.0%, from 64% at baseline to 63% at the twelvemonth follow-up. There were no significant differences among the groups for the SHUEE SFA, Box and Blocks, or AHA.

Secondary Outcomes: Bodily Impairment and Participation Restriction

Comparison of the three treatment groups showed a significantly greater increase in mean pinch strength of the affected hand in Group 1 and Group 3 relative to Group 2 (p = 0.004) (see Appendix). Group 1 had a mean increase in pinch strength of 0.7 kg, and Group 3 had a mean increase of 0.4 kg. Group 2 had a mean decrease in pinch strength of 0.6 kg.

No significant differences were found among the three treatment groups in terms of the PODCI global and the PedsQL standard version.

Four domains of the PedsQL CP module (school, fatigue, eating, and speech) showed no significant differences among the three groups. In the movement domain, Group 1 had a greater change than Group 2 or Group 3 (p = 0.002). The mean increase in Group 1 was 26.4%. Group 2 and Group 3 both had a mean decrease of 3.3%.

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No significant differences among the groups were measured for the COPM performance score. For the COPM satisfaction score, Group 1 had a greater change than both Group 2 and Group 3 (p = 0.002). Group 1 had a mean increase of 4.4, Group 2 had a mean increase of 2.0, and Group 3 had a mean increase of 0.9.

Discussion

This study shows that tendon transfer surgery was more effective than botulinum toxin injections or regular, ongoing therapy in children with upper-extremity CP who met standard surgical indications for intervention in terms of improving pinch strength, dynamic limb positioning while carrying out functional tasks as measured by the SHUEE DPA, and as assessed by the participation measures of PedsQL CP and COPM satisfaction. The functional improvements were of modest magnitude. The improvements in DPA reflect the improved wrist extension and forearm supination positioning during functional tasks after FCU to ECRB tendon transfer and pronator teres release. The primary benefits of tendon transfer surgery are improved joint positioning, particularly wrist extension and supination, which has some secondary effects on improved function and participation; however, other functional benefits, such as improved dexterity, were not assessed in this trial.

Similar improvements in bodily impairment measures have been previously reported^{4-6,8,9}, including improvements in wrist extension, at the price of a loss of wrist flexion^{24,25}. In this study, the FCU to ECRB transfer was tensioned with the wrist in neutral to avoid excessive loss of wrist flexion, which could create extensor habitus, as has been previously reported^{5,26,27}. This surgical technique was successful in improving grip strength because of the improved wrist posture. With the improvement in wrist extension, loss of wrist flexion was noted. Although the forearm and wrist positions were improved in the surgically treated group, significant changes in thumb position were not noted. A review of the assessment tools used showed difficulty in measuring changes in thumb position and function. Further investigation into more sensitive assessment tools for thumb position and function is recommended.

Although the SHUEE DPA measured changes in both wrist and forearm position during activities in our study patients, these changes were not reflected by improvement in AHA results. The scale for each of the twenty-two subtests of the AHA is small (14; in our patients, treatment rarely caused changes of more than one point). Other studies have also questioned whether a true change in the clinical use of the limb is reflected in a score change in the AHA²³. The AHA may not be as sensitive to joint-position changes as is the SHUEE DPA. Whether the instruments, such as the AHA or the thumb assessments, were not sensitive enough to detect change, or whether substantial changes actually did not occur, remains an issue for additional investigation.

Although this study used appropriate standardized outcome measures, the results of this study may differ from those of other evaluations of the same type of interventions that used different outcome measures. For example, Koman et al.¹¹ reported on seventy-three children with upper-extremity CP who

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were treated with botulinum toxin injections in a prospective, randomized, double-blind, placebo-controlled clinical trial and concluded that, after a series of three injections, children who received botulinum toxin injections showed an improvement in the Melbourne assessment at twenty-six weeks compared with the children who received the placebo. In their study, the botulinum toxin and placebo injections were given at enrollment, eight weeks, and twenty weeks, as deemed clinically appropriate, with assessment at twenty-six weeks for children with a wide variety of clinical presentations, many of whom were not surgical candidates. Although Koman et al. reviewed the short-term effects of botulinum toxin and placebo injections, our study looked at three injections of similar doses of botulinum toxin and similar evaluation periods, but measured the effects six months after the last injection had been administered. Our study used different assessment tools than did Koman et al. The major detriment to botulinum toxin injections reported in our study was a loss of grip and pinch strength, even six months after the last injection; grip and pinch strength were not measured by Koman et al. Thus, different evaluation tools, a different length of follow-up, and different patient populations may account for the positive results after botulinum toxin injections reported by Koman et al. compared with the negative results reported in the present study. Similarly, other studies^{12,13,15,16} indicating the benefits of botulinum toxin injections used different evaluation tools and different injection protocols than the present investigation.

In a 2010 Cochrane review¹⁴ of ten randomized controlled trials of botulinum toxin injections, the authors concluded that "a combination of botulinum toxin injections and therapy is more effective than therapy alone in reducing (bodily) impairment, improving activity level outcomes, but not for improving quality of life. When compared with placebo or no treatment, there is moderate evidence that botulinum toxin alone is not effective." The authors¹⁴ acknowledge the wide variations in dose frequency and in outcome measurement tools, and recommend further research into longitudinal outcomes, the timing and effect of repeated injections, and the most effective adjunct therapies. The Cochrane review reported significantly decreased spasticity with the use of botulinum toxin injections but reported that most trials did not include measurement of the loss of grip and pinch strength. The present study adds new information to the literature in that loss of strength persisted and no significant improvements in measurements of bodily impairment, activity limitations, or participation restrictions were found for the patients who received botulinum injections.

The strengths of our study include the study design. Agreement among seven hand surgeons to use the same surgical protocol allowed standardization across multiple sites. The multisite study design presents data from multiple surgeons, providing wider applicability of the results. In addition, standardized validated outcome tools were used for assessment. These tools were carefully selected to assess all domains of the WHO ICF. This study provides important information for the treating physician to inform prospective patients that surgical intervention can improve wrist extension and forearm supination as well as pinch strength that enables functional use of the SURGERY IN UPPER-EXTREMITY CEREBRAL PALSY COMPARED WITH BOTULINUM TOXIN INJECTIONS OR THERAPY

limb, which is reflected in improved satisfaction in participation, when compared with three serial botulinum toxin injections or regular, ongoing therapy.

This study had several weaknesses. The randomized study design was an obstacle for many families in consenting to enroll their child²¹. Several other studies have had recruitment difficulties in executing a prospective randomized surgical study; two recent large orthopaedic studies, BRAIST (Bracing in Adolescent Idiopathic Scoliosis Trial) and SPORT (Spine Patient Outcomes Research Trial), also included a patient-preference arm^{28,29}. We did not study the long-term effects of the treatment interventions for this report; the patients were followed for only one year. Even with one year of follow-up, 13% of the patients (five of thirty-nine) did not complete the study.

On the basis of our findings, we no longer recommend botulinum toxin injections as a treatment modality for children who meet indications for tendon transfer surgery. This study did not provide evidence against therapeutic modalities as maintenance treatments, and we continue to recommend them.

Appendix

 $(eA)^{A}_{Gaussian}$ description of the outcome measurement methods used; figures showing a comparison of active ROM supination and pronation, wrist extension and flexion, grip strength, and pinch strength among the groups at baseline and twelve months; and tables presenting a comparison within each treatment group for all outcome measures and a comparison of mean change in secondary outcome measurements among the groups are available with the online version of this article as a data supplement at jbjs.org.

NoTE: The following investigators, therapists (T), coordinators (C), and other contributors (OC) were involved in this study (listed in the order of the number of patients who provided consent, shown in parentheses): *Jon Davids, MD, with *Lisa Wagner and *Laura Peace (T), Shriners Hospitals for Children-Greenville, South Carolina (8); *Philip Gates, MD, with Ann Boyd and Laura Burford (T), Susan Campbell (C), and Virginia Scales (OC), Shriners Hospitals for Children-Shreveport, Louisiana (7); Alfred Hess, MD, and *Cara Novick, MD, with Adrienne Karol, Lynn White, and Carmen Longnecker (T) and Nancy Pisciotto, Jennifer Jenniks, and Ed Quigley (OC), Shriners Hospitals for Children-Tampa, Florida (7); *Douglas Hutchinson, MD, with *Chris Pratt (T) and Bruce MacVilliamson and Barbara Johnson (OC), Shriners Hospitals for Children-Intermountain, Salt Lake City, Utah (5); *Michelle James, MD, and *Anita Bagley, PhD, with *Trang Bui, *Denise Caldwell, and Cheryl Hanley (T), Sherry Middleton and Susan Anderson (C), and Grace McNelis (OC), Shriners Hospitals for Children-Northern California, Sacramento, California (4); *Ann Van Heest, MD, with *Wendy Tomhave (T) and Gabriela Ferski (C), Shriners Hospitals for Children-Twin Cities, Minneapolis, Minnesota (4); and Randip Bindra, MD, with Jasmine Gilliam (T), Shriners Hospitals for Children-Chicago, Illinois (4). Chicago, Illinois (4).

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Indicates members of the working group that planned the study design.

Ann E. Van Heest, MD Department of Orthopaedic Surgery, University of Minnesota, 2450 Riverside Avenue, Suite R200, Minneapolis, MN 55454. E-mail address: vanhe003@umn.edu

Anita Bagley, PhD Fred Molitor, PhD Michelle A. James, MD Shriners Hospitals for Children-Northern California, 2425 Stockton Boulevard, Sacramento, CA 95817

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