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Early vs. Delayed Surgical Treatment of Complicated Congenital Hand Deformities: A Prospective Study

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Abstract Numerous types of congenital limb differences and anomalies can be surgically corrected. We compared the complexity of surgical interventions and outcomes between early and delayed intervention cases of complex congenital hand deformities. The null hypothesis was that the patient's age at the time of surgery would not affect the quality of surgical interventions and outcomes. Methods. A prospective single-center study with consecutive cases occurred from May 2012 to May 2022. The study cohort comprised patients presenting for surgical management of isolated and syndrome-affiliated unilateral or bilateral complex forms of congenital hand deformities. They were divided into patients younger than five years (Group 1) and older than five (Group 2). Patients with deformities that involve three or more fingers were included in the study. Results. Altogether, 133 patients (Group 1: n=73, M/F 39/34, mean age 2.8 ± 1.5 years; Group 2: n=60, M/F 32/28, 8.5 ± 3.2 years) were operated and completed follow-up. We operated on 732 fingers on 202 hands in 138 surgeries. Most of the patients had complicated syndactyly, symbrachydactyly, and syndrome-related deformities. Young patients restored their motor and sensory nerve activity during the first six months since surgery, while older patients needed from six to 12 months for complete nerve recovery. One reoperation was needed for a young patient because of web creep, and four reoperations were needed for older patients because of graft rejection. No other significant differences between the two age groups were detected. Conclusion. The timing of surgical treatment of congenital hand deformities is not paramount, and good functional outcomes can be achieved in children of any age.

Keywords Congenital hand deformities, Pediatrics, Surgery, Symbrachydactyly, Congenital syndromes

1. Introduction

Congenital hand deformities represent one of the most challenging problems in pediatric orthopedics and cause significant functional, cosmetic, and psychological impairments for patients. In the 1960s-70s, it was agreed that the optimal timing for surgical corrections is between one and two years of age [1]. The issue of choosing the age of patients to start surgical treatment remains controversial, although there are numerous supporters of early interventions. The first two years of life are paramount for developing cerebral cortex patterns of hand use. When surgery is delayed, these patterns will need to be retrained, and the outcome may not be satisfactory [2]. The emerging literature reports that the optimal age for a child to undergo surgical intervention is from three months to two years. The reality is not similarly straightforward [3]. Slevin et al. investigated the timing of surgical release in syndactyly cases in the USA. The mean age of syndactyly release was 3.6 years, but 27.1% of patients underwent release after the age of five years [4]. Other publications reported the age ranges of the operated patients as from 2 months to 3.8 years, 2 to 4 years, 2,5 to 7,3 years, or the average age of 12 years. A review on camptodactyly reported that children of any age from two months to 18 years were operated [5]. Kurebayashi et al. reported that children with syndactyly operated on younger than one year had worse postoperative outcomes than those operated on older than one year. Patients' age under two years old was considered as a contraindication for syndactyly treatment with external fixation frames [6].

The concept of our research was developed in 2012 while observing children of any age admitted to our institution because of congenital hand deformities. The management of commonly encountered conditions, such as clinodactyly, thumb hypoplasia, and simple syndactyly, was scrupulously described. The authors concentrated on more complex anomalies. We aimed to compare the complexity of surgical interventions and the outcomes between early intervention and delayed intervention cases of complicated hand deformities. The null hypothesis was that the age of patients at the time of surgery would not affect the quality of surgical interventions and outcomes.

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2. Materials and Methods

2.1. Study Design and Setting

To address the research purpose, the investigators designed and implemented a prospective single-center study with a consecutive collection of cases following the STROBE guidelines. The study protocol and written informed consent conformed to the ethical guidelines of the 1975/2013 Helsinki Declaration and were approved by the Institutional Review Board. The study period was set from May 2012 to May 2022 for recruitment, exposure, and data collection. It was terminated in June 2023 when study groups reached the calculated sample size, and all participants had at least a 12-month follow-up period. The patients were referred to the specialized Republican Center for Pediatric Orthopedics by secondary care practitioners who diagnosed complex forms of congenital hand deformities. Some patients were referred for reoperations after previously performed unsuccessful interventions or because of recurrence. Two surgeons experienced in pediatric hand surgery operated on these patients at this tertiary referral hospital with fully functioning surgical units and proper imaging facilities.

2.2. Participants and Study Size

The study cohort comprised all patients presenting for evaluation and management of isolated and syndrome-affiliated unilateral or bilateral complex forms of congenital hand deformities. The patients who were admitted and operated on at the age younger than five years comprised Group 1. The 5 to 18-year-old patients were assigned to Group 2. A standard sample size 75 calculation for a single-center study determined that a minimum of 60 subjects per group with a total of 120 subjects were required for the difference between Group 1 and Group 2 values to be statistically significant when $\alpha = 0.05$, the power of the study set at 80% (null hypothesis as equivalence design, $\alpha=0.05$; $\beta=0.20$). The study was terminated when Group 2 reached 60 cases with a proper follow-up period.

Inclusion criterion: patients should have isolated or syndrome-affiliated unilateral or bilateral hand deformities that involve three or more fingers. Single-finger clinodactyly, camptodactyly, thumb hypoplasia, six-digit polydactyly, and simple syndactyly cases were excluded. We also excluded cases with the above hand-and-wrist deformities such as phocomelia.

2.3. Presurgical Evaluation

Proper selection of patients for surgical intervention was the second step after the inclusion/exclusion criteria were met. We considered contraindications for surgical intervention that might include poor health conditions because of a congenital syndrome, within-hand arteriovenous or nerve malformations that might require a multicenter approach, and risk for postsurgical complications and/or recurrence. Patients with multiple syndrome-related malformations were evaluated by a multidisciplinary team.

We documented active and passive range of motion of upper extremity joints, bimanual reach towards an object, the palmar grasp, grip strength, pinch strength, abilities and disabilities in manipulating an object with the fingers and with the palm of the hand according to the "Hand Function in Children with Congenital Disorders" guidelines of 2014. A pincer grasp with the tip of the thumb and index finger was checked if the deformity permits. We also used the Manual Ability Classification System (MACS; from 1 – good to 5 – totally inadequate). ABILHANDS-Kids function scale was not used because some items (e.g., "sharpening pencil") were inapplicable for young patients.

A surface EMG investigation was used to assess the functions of the nerves and extrinsic and intrinsic muscle groups and to rule out or detect compression neuropathies. The Wrinkle test was used to check the nerve integrity of the hand by assessing the nervous response to an environmental stimulus in young children who cannot follow instructions. To investigate vascularization, the possible hand skin temperature asymmetry was checked by infrared thermometry and assessed against the healthy hand or against the normative database for bilateral deformities. Arteriography was used for cases with suspected blood circulation abnormalities to rule out postsurgical risk of jeopardizing distal circulation. X-ray investigation was used to understand positions, aphalangia, and deformities of the hand bones, assess a patient's bone age against his/her chronological age, and detect cases with advanced and delayed skeletal development with the help of the hand-and-wrist radiographic atlas for digital bone age estimation. An evaluation of the amount of available skin helped to choose between graft and graftless surgical techniques.

2.4. Surgical Techniques

We followed well-established techniques for hand, pediatric plastic, and congenital hand defects surgery. They included general anesthesia, a pneumatic tourniquet, drawing of palmar incision pattern, Cronin zigzag incisions, Buck-Gramcko techniques for the thumb and combined finger malformations, skin and subcutaneous tissue flap lobes, and postsurgical analgesics. For multiple-digit malformations, the thumb was released first, followed by the border fingers. A distal revascularization was used to replace the possible arterial deficit if a case so required. In cases when the digital nerve bifurcated distal to the newly constructed web space, microdissection was used to separate it.

We used both graft and graftless surgical techniques. A full-thickness skin graft was harvested from various sites according to the age and condition of a patient. We used autodermal grafts harvested from the thigh, the lateral third of the inguinal crease of the groin area, and the inner shoulder area. In uncircumcised boys, we used foreskin to create proper web spaces. In acrosyndactyly, the index finger was released first. After that, the release of the fifth finger was achieved, which was followed by the release of the central two fingers. We used over compensating the

webspace approach to prevent possible web creep. We used one-stage or two stage surgical corrections individualizing the management of the deformities.

2.5. Follow-up and Outcome Evaluation

We evaluated patients during the dressing change twice a week during the first three to four postsurgical weeks. X-ray, EMG, and infrared thermometry studies were carried out on the third postsurgical day to detect the bone positions and possible nerve and blood supply inadequacies. They were repeated during the six and 12-months follow-ups to assess the recovery process. The subsequent follow-ups were scheduled three, six, and 12 postsurgical months and further in six month periods, checking functional outcomes, physiotherapy and training results, and possible recurrence. For older patients, we examined the grip and in-hand manipulation skills such as writing and drawing or buttoning and tying shoes for two-hand activities. Scaring was evaluated using the modified Vancouver scar scale with 0 (good) to 13 (inadequate) score scale evaluation criteria for scar pigmentation, vascularity, pliability, and height. Web creep was assessed using a 5-point scale system from 0 for normal web space to 4 for total web creep to the proximal interphalangeal joint. We estimated scores 0 to 1 as effective treatment, 2 to 3 as partially inadequate treatment, and 4 as no benefit achieved and reoperation required.

2.6. Bias

To avoid selection bias, both study groups consisted of consecutive cases. While we had fewer late-release cases than early-release cases, the data collection period ended when Group 2 reached the desirable sample size. Therefore, Groups 1 and 2 had an unequal number of cases, but both reached the calculated sample size. To avoid report bias, all cases, starting from 2012, were reported in the charts and a study database form in a similar way indicating all variables to be used for further analysis.

2.7. Analysis

The groups of patients were formed according to their age, which served as a primary predictor variable. A Chi-Square test was used to analyze demographic data and time-related variables. We used one-way ANOVA to compare the differences between the groups in the follow-up outcomes and the postsurgical complication rate (Fisher's exact test) (SPSS ver. 19.0). Statistical significance was considered at the $p \leq 0.05$ level.

3. Results

A total of 536 potentially eligible patients were examined for eligibility; 133 patients (Group 1: n=73, M 39, F 34, mean age 2.8 ± 1.5 years; Group 2: n=60, M 32, F 28, mean age 8.5 ± 3.2 years) were confirmed eligible and included in the study, were operated, completed follow-up, and were analyzed (Table 1). The remaining 403 patients were excluded following the exclusion criteria, surgery contraindication, or lost in follow-up. Most of the patients had complicated syndactyly, symbrachydactyly, and syndrome-related deformities.

Table 1. A comparative analysis of demographics and hand characteristics between Group 1 (n=73) and Group 2 (n=60)

Variables	Group 1	Group 2	P value
Age at surgery (yr), mean + SD	2.8 ± 1.5	8.7 ± 3.4	
Sex (M/F)	39/34	32/28	32/28
Affected hand (no.)			
Left	13	9	0.67
Right	22	20	0.90
Bilateral	38	31	0.36
Poland syndrome	15	12	0.95
Apert syndrome	8	5	-
Constriction ring syndrome	3	2	-
MACS score unilateral lesion (n=64)	3.2 ± 0.5	2.8 ± 0.6	0.34
MACS score bilateral lesion (n=69)	4.1 ± 0.7	3.6 ± 0.8	3.6 ± 0.8
Skin temperature asymmetry	14	11	0.42
Prior hand surgery/recurrence	21	18	0.88

Of the 133 patients, 37 had unsuccessful prior hand surgery or recurrent deformities, 96 patients underwent initial surgical corrections, 48 patients had genetic multiple malformation syndromes, including 13 with Apert syndrome and 27 with Poland syndrome. We operated fingers on 202 hands in 157 surgeries. Table 1 shows no statistically significant differences between the groups except for age. No significant differences between the groups were detected in the speed and quality of their postsurgical recovery and hand function improvement (Table 2).

The 3-day after-surgery EMG check did not detect fibrillation potentials in the hand muscles in all cases and ruled out possible muscle denervation secondary to axonal dropout. Further follow-up EMG investigations revealed a statistically significant difference between the groups in the speed of nerve recovery. Group 1 patients restored their motor and sensory nerve activity during the first six postsurgical months, while Group 2 patients needed six to 12 months for complete nerve recovery.

The number of complications was small (11/133, 8.3%) (Table 3). However, five reoperations were needed. Only one reoperation was needed for a Group 1 patient because of score 4 web creep, and four reoperations were needed for Group 2 patients because of graft rejection. A lack of a single case with skin necrosis demonstrated that no nerve or arterial injury occurred, and vascularization of the skin flaps was not decreased. Web creep was rare (1.5%) and occurred in two patients who were operated before 18 months of age.

Table 2. A comparative analysis of surgery-related, follow-up, and functional outcomes results between Group 1 (n=73, 111 hands) and Group 2 (n=60, 91 hands). Surgery duration, number of involved fingers, number of graft and graftless techniques, and Vancouver scar scale scores are given for the operated 202

Variables	Group 1	Group 2	P value	P*
Surgery duration per hand (min)	110 ± 35	95 ± 40	0.35	
Three fingers involved (n)	21	25		
Four fingers involved (n)	36	29		
Five fingers involved (n)	47	44		
Skin grafts (n)	39	43		
Two-stage surgical intervention (n)	11	8	0.25	
Follow-up 1 month**				
MACS score unilateral lesion (n=64)	3.1 ± 0.5	2.8 ± 0.6	0.87	0.96
MACS score bilateral lesion (n=69)	4.1 ± 0.7	3.4 ± 0.8	0.16	0.96
Vancouver scar scale score	0.9 ± 0.3	1.1 ± 0.4	0.90	
Skin temperature asymmetry (n)	1	0		0.001
Follow-up 3 months				
MACS score unilateral lesion	2.7 ± 0.5	2.5 ± 0.6	0.84	0.35
MACS score bilateral lesion	3.9 ± 0.7	3.1 ± 0.8	0.14	0.70
Vancouver scar scale	0.45 ± 0.3	0.6 ± 0.4	0.73	
Web creep score***	0.2 ± 0.2	0.1 ± 0.1		
Follow-up 6 months				
MACS score unilateral lesion	2.1 ± 0.7	2.1 ± 0.8	1.00	0.05
MACS score bilateral lesion	2.3 ± 0.8	2.1 ± 0.9	0.95	0.03
Vancouver scar scale	0.2 ± 0.2	0		
Follow-up 12 months				
MACS score unilateral lesion	1.4 ± 0.4	1.4 ± 0.4	1.00	0.01
MACS score bilateral lesion	1.7 ± 0.7	1.3 ± 0.3	0.79	0.01

Table 3. The rates of postsurgical complications in young (Group 1, n=73) and older (Group 2, n=60) patients. The statistical comparison was not performed because of the small number of complications

Complication	Group 1	Group 2
Perioperative complication	1	1
Contracture	0	0
Web creep scores 2-3	1	0
Web creep score 4	1	0
Hematoma	1	0
Skin necrosis	0	0
Infection	0	0
Persistent pain	0	0
Finger joint stiffness	1	1
Hypertrophic scarring	0	0
Donor site infection*	0	0
Flap maceration	0	0
Graft rejection*	0	4
TOTAL	5	6
Revision surgery	1	4

* Donor site infection and graft rejection were assessed among the patients who were operated with graft techniques.

The average follow-up period was 3.6 years for Group 1 and 4.7 years for Group 2 patients. While initial functional improvements were observed during the 3-month follow-up meeting, 6 to 12 months were needed to achieve stable improvements (Tables 2, 3). Rehabilitation, physiotherapy, and training activities during the follow-up period contributed to postsurgical results. Besides MACS scores improvement and positive EMG changes, we noticed improvements in the palmar grasp, pincer grasp, and in-hand manipulation skills.

4. Discussion

We compared the complexity of surgical interventions and the outcomes between early intervention and delayed intervention cases of various isolated or syndrome-affiliated unilateral or bilateral complicated congenital hand deformities. Besides a speedy nerve recovery in young children and graft rejection cases in the group of older patients, all other variables did not show statistically significant differences. Therefore, our null hypothesis that the age of patients at the time of surgery does not affect the quality of surgical interventions and outcomes is mainly confirmed.

Complicated hand deformities have a functional, cosmetic, and psychosocial impact. A constant improvement in surgical approaches, including microsurgical techniques, within this

area of pediatric surgery has been traced during the last 20 years. It made the question of the timing of surgery less acute. Surgeons operate on children with hand deformities of any age with comparable results. The question of why some parents do not apply for surgical treatment of their children at an early stage remains. In some our cases, physicians evaluated an infant's condition as "untreatable" and reevaluated as "treatable" in ten or more years. Some parents self-evaluated the child's condition as untreatable until convinced otherwise. Some delayed cases were operated on when the general health conditions of patients improved. We noted a fear of complicated surgical intervention, religious sentiments, and financial considerations. Our observations support results of Corder et al., who mentioned parents' fear of surgical correction and congenital cardiac anomalies as factors which delayed surgical interventions.

It is beyond the aim of this research, but we would like to share some other observations. While we performed a presurgical assessment of vascularization in the affected hand and used arteriography in some cases, we confirm Wong et al. observation that presurgical assumptions and trans-surgical findings in hand vascularization do not always correlate. We support Solia's et al. conclusion that the nerve supply of the hand is characterized by numerous variations. Web creep remains a challenging postsurgical complication. We found that the foreskin is perhaps the best graft for proper web formation for uncircumcised boys. The bone age estimation as delayed, normal, or advanced against the chronological age was helpful for surgery planning, decision -making, and assessing functional outcomes. The hand-wrist film method was helpful to estimate skeletal maturity. The nature of interventions necessary to correct certain specific hand deformities remains debatable. Customized methods remain the surgical choice because all variations of hand differences are possible.

5. Conclusions

The timing of surgical treatment of congenital hand deformities is not paramount. Good functional outcomes can be achieved in children of any age, assuming that the surgical intervention will be followed by proper rehabilitation, physiotherapy, and training activities.

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