Outcome of hand surgery in children with spasticity – a 9-year follow-up study

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The aim of this study was to evaluate whether short-term positive effects on bimanual function after surgery of the paretic arm in cerebral palsy are maintained long term. Assisting Hand Assessment (AHA) and active range of motion was tested before surgery and at 7 month and 9-year follow-up (n = 18). AHA improved significantly from 50 to 52 U at 7 months, but was not different from before surgery at the 9-year follow-up, 49 U. Surgery of wrist and elbow flexors significantly improved active extension. Improvement in wrist and elbow extension was maintained at the 9-year follow-up, but usefulness of the hand measured with AHA had returned to the same level as before surgery. *J Pediatr Orthop B*

Introduction

A brain lesion during gestation or in the immature brain may result in cerebral palsy (CP), which is characterized by motor impairments but also includes many other symptoms, depending on the timing, location, and extent of the brain damage [1,2]. Asymmetry of the injury typically results in unilateral CP, with one arm and leg more affected than the other. An acquired brain injury (ABI), occurring after 5 years of age, may also result in spastic hemiplegia [3]. In both groups, contractures are common, with progressive stiffening and shortening of the musculotendinous unit [4]. In a population based study on 771 children with CP, about one third developed contractures which became significant at 4 years of age regarding wrist extension with straight fingers, and at 7 years of age regarding supination and elbow extension [5]. There is one study on the effect of lower limb multilevel surgery with a comparison with a control group, albeit treated with botulinum toxin and physiotherapy. It was found that the operated group showed improved gait and range of motion, whereas the unoperated group showed deterioration [6]. In the upper limb, the muscles most prone to contracture formation are the biceps brachii, the pronator teres (PT), the flexor carpi ulnaris (FCU), the flexor carpi radialis, superficial and deep finger flexors, and the adductor pollicis (AddP) muscles. Cocontraction and poor selective muscle control also contribute toward the typical position with the elbow and wrist flexed and the forearm pronated [7]. When the child runs or is excited, the flexed position of the arm is aggravated

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further, which impairs the function and appearance of the arm even more. Impairment of one hand affects the activity in daily life as we are typically doing most things with two hands. Persons with CP or ABI will naturally use the better hand the most, but the paretic hand can be very useful as a helper hand. The usefulness of the paretic hand can be measured with the Assisting Hand Assessment (AHA), and the score at 18 months can predict the score at 12 years of age [8]. Progressive flexion of the wrist and elbow, adduction of the thumb, and a fixed pronation of the forearm will make it more difficult to approach, hold, and manipulate objects, and body appearance is also affected. This can be treated surgically by individually tailored tendon lengthenings and tendon transfers. Even though tendon transfer for the spastic hand was first described more than half a century ago [9], the evidence for benefits of upper limb surgery in a long-term perspective is low and there are few available studies [10]. Short-term followup of surgery has shown a functional result [11,12] and the effect has even been observed in bimanual activities, measured with the AHA [13].

The aim in this study was to investigate whether short-term improvements in active range of motion and bimanual hand function (AHA) are maintained at a 9-year follow-up by investigating the same participants as in a previous study [13]. In that study, we found that the operated upper limb was more useful in bimanual activities and that the range of motion was improved. We also wanted to know how the appearance of the hand was appreciated by the child.

Patients and methods

This is a case series, performed at the Department of Pediatric Orthopaedic Surgery, Karolinska University Hospital. In a previous study, we examined 18 children

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with CP consecutively planned for hand surgery, and then performed a follow-up at 7 months [13].

For 15 of the 18 children in our previous study, at least 7.5 years had passed after the first surgery, and they were contacted for a follow-up assessment. Two of the remaining 15 patients declined to participate in the follow-up. We included two more patients, no. 7 and 15, with 10 and 8.2 years of follow-up, resulting in a total of up to 15 patients. Twelve of the patients had unilateral CP, and three ABI; one had had a traumatic brain injury at age 10, one was operated for a brain tumor at age 9, and one had had a stroke at 9 years. Details on age at first surgery (11.3 \pm 3.5) and age at long-term follow-up (20.5 \pm 3.5 years) are presented in Table 1. Long-term follow-up was performed at an average of 9 years (106 months, range: 90–126 months) after surgery of the upper extremity.

Surgical procedures and number of surgeries performed are listed for each patient in Table 2. Each child was assessed before surgery and combinations of surgical techniques involving the elbow, wrist, and thumb were tailored individually with the main aim of improving reach, grip, release, and object manipulation.

After surgery, all patients wore a long-arm cast for 6 weeks, maintaining the arm in an overcorrected position. Following cast removal, intense training (2 months, 1–3 times/week, 1–2 h/session) was performed under the supervision of an occupational therapist (OT). Less frequent training (1 time/ week, 1–2 h/session) was continued up until 6 months after surgery. During the initial 6 months, the children were also encouraged to use the hand in their daily activities through an individually designed home-based training program. No supervised training intervention was maintained thereafter.

According to routine, a post-OP AHA and clinical examination was performed about 6 months after surgery. All clinical assessments were performed by the same two OTs (C.L.E. and A.L.). The AHA films were scored by an OT not involved in the treatment or follow-up (B.M.Z.).

The AHA measures and describes how efficiently people with unilateral hand-dysfunction spontaneously use their affected hand in bimanual activities. It is a criterion-referenced test proved to be valid, reliable, and responsive to change for children 18 months to 18 years of age [14]. The assessment is a standardized videotaped 10–15 min long semi-structured

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Patients no.	Elbow extension	Supination	Wrist extension	Finger extension	Thumb ext/abd
1		1	3		
2		1	1	1	
3		1	1		2
4	1	1	1		1
5		1	1		2
6	1	1	1		1
7		1	1		
8	1	1	1	1	1
9	1	1	2	1	2
10	1	1	1		
11		1	2		2
12	1	1	2	1	
13	1	1	2	1	3
14	1	2			
15	1	2	1		4
Patients/goal	9	15	14	5	9

Examples of surgical procedures/goal.

Elbow extension: Biceps tendon lengthening, brachialis aponeurectomy, BR release.

Supination: FCU-> ECRB, ECRL or EDC, PT release, lengthening or rerouting, PQ lengthening/release, release of membrana interossea.

Wrist extension: FCU->ECRB,ECRL or EDC, FCR->ECRL or ECRB, FCR lengthening, ECU->ECRB, BR->ECRB, PT->ECRL, wrist fusion with proximal carpal row ectomy, PL release, intramuscular aponeurotic release of FDS, FDP. Finger extension: intramuscular aponeurotic release of FDS, FDP thumb extension/abduction: EPL rerouting, release of AddP, release of FDI, FPL lengthening, PL->EPL, BR->APL, arthrodesis of MCP I.

AddP, adductor pollicis; Bic, biceps; BR, brachioradialis; Brach, brachialis; ECRB, extensor carpi radialis brevis; ECRL, extensor carpi radialis longus; EDC, extensor digitorum communis; EPL, extensor pollicis longus; FCR, flexor carpi radialis; FCU, flexor carpi ulnaris; FDI, first dorsal interosseus; FDP, flexor digitorum profundus; FDS, flexor digitorum superficialis; FPL, flexor pollicis longus; MCP I, metacarpophalangeal joint of thumb; PL, palmaris longus; PQ, pronator quadratus; PT, pronator teres.

Table 1 Patients with 9.3 years of follow-up of hand surgery in hemiplegia

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Patient no.	Diagnosis	Side	Age at first surgery (years)	Number of surgeries	Years between surgeries	Short-term follow- up (months)	Long-term follow-up (months after surgery)	Age at long-term follow-up (years)
1	Traumatic brain	Left	13	1	0	7	98	21
2	Brain tumor age 9	Left	11	2	2	6	114	20
3	CP hemi	Left	8	2	1	8	116	18
4	CP hemi	Left	11	1	0	6	94	19
5	CP hemi	Right	7	2	3	6	120	17
6	CP hemi	Right	17	1	0	8	97	25
7	CP hemi	Left	8	1	0	7	120	19
8	Stroke age 7	Right	16	1	0	7	108	25
9	CP hemi	Left	8	2	3	6	102	17
10	CP hemi	Left	11	1	0	7	90	19
11	CP hemi	Left	9	4	1, 4, 3	8	91	19
12	CP hemi	Left	17	1	0	6	121	27
13	CP hemi	Left	15	1	0	9	126	26
14	CP hemi	Left	9	1	0	7	99	17
15	CP hemi	Right	9	3	5, 2	6	98	18
Mean		5	11.3			7	106	20.5

CP, cerebral palsy; hemi, hemiplegia.

play session. Specifically chosen toys requiring the use of both hands are used. For adolescents, a semi-structured activity is used, including opening and wrapping a present and writing and placing a note in an envelope. It includes 22 items, describing different actions of the assisting hand, each scored on a four-point rating scale. The raw score range from 22 to 88 (low to high ability). The raw sum score is converted into the logit-based 0–100 AHA-unit scale on the basis of Rasch measurement analysis. The smallest detectable difference for the AHA has been calculated to five AHA-units; this means that on an individual level, a change of 5 U or more can be considered a true measurement change, also clinically and functionally detectible [15]. We based the scoring on the AHA 4.4 version [14,16].

Active range of motion

Active range of motion of the elbow, forearm, and wrist was measured using a goniometer. On the basis of the principle that a neutral position equals 0°, measurements were taken from pronation to supination of the forearm and from flexion to extension of the wrist and elbow. For the elbow, limitation of extension to 0° is stated as an extension deficit (American Academy of Orthopaedic Surgeons, 1965).

Zancolli classification

Simultaneous voluntary wrist and finger extension was classified into four groups according to Zancolli's classification [17]. In type 1, the fingers can extend while the wrist is in less than 20° flexion; in type 2A, the fingers can extend with the wrist in more than 20° of flexion and when the fingers are flexed, the wrist can extend. In type 2B, the fingers can extend when the wrist is flexed more than 20° , but the wrist cannot extend even with the fingers flexed. In type 3, neither the fingers nor the wrist can extend. Klingels *et al.* [18] have shown the classification to have excellent reliability.

House classification of thumb position

The position of the thumb during active grasp was classified into four groups described by House [11]. The evaluation was performed by observing when the patients opened their hand to actively grasp objects. House I=Adduction in the first carpometacarpal joint (CMC I) because of increased tone and/or contracture of AddP, House II = adduction in CMC I and flexion in the first metacarpal-phalangeal joint (MCP I) because of increased tone and/or contracture in AddP and flexor pollicis brevis muscles, House III = adduction in CMC I and hyperextension in MCP I and/or first interphalangeal joint, and House IV = adduction of the CMC I and flexion of MCP I and interphalangeal joint [19].

House Classification of Upper Extremity Functional Use was developed to classify the use of the hemiplegic hand [19]. It is a nine-graded scale, with categories ranging from no use at all (0), to spontaneous use (8). The evaluation was determined by observing how active the affected hand was in selected activities.

Appreciation of appearance of upper extremity

Patients and, in the cases where the patients could not answer, their parents were contacted by telephone. The interview included four questions, presented to each patient in the same order. The patients were asked to rate their satisfaction with the appearance of their hand/ arm from a scale of 1 to 10. The questions were as follows: (a) How pleased are you with the appearance of your hand and arm when you walk? (b) Can you recall how pleased you were before the surgical treatment? (c) How pleased are you with the appearance of your hand and arm when you sit down? (d) Can you recall how pleased you were before the surgical treatment? The telephone calls were made by an MD who had not taken part in the care of the patient at any stage (F.v.W.).

Statistical analysis

Nonparametric methods were used to define the outcome of the surgery. Results were compared using Friedman's test for repeated measures, with Dunn's post-hoc test for multiple comparisons between groups. The short-term AHA was missing for patient no. 7 and 15. As their AHA at other time-points during their childhood and adolescence were very stable, we estimated the short-term AHA as the mean between pre-OP and long-term follow-up. GraphPad Prism 7 was used for calculations (GraphPad Software, La Jolla, California, USA). For all statistical tests, a probability of P value of less than 0.05 was adopted as the criteria for determining significant differences. Values in the text are expressed as the median, the minimum, and the maximum, apart from age, which is expressed as mean. Median and interquartile range were used for all figures. No statistics were performed on the categorical data, Zancolli classification, House classification for hand function, House thumb classification, or how the child appreciated the appearance of the affected hand.

Results

There was a wide range of hand function in the children included, evidenced by a large span in the AHA units at pre-OP, ranging from 7 to 64, with a median of 50 U. At the post-OP follow-up at about 7 months, AHA had improved to 52 U, range 24–67 (main effect of time P=0.0243), but declined to 49 U, range 12–67 at the 9-year follow-up (Fig. 1a). However, different items of the AHA did not change uniformly (Fig. 1b). The AHA test items 'calibrates', 'stabilizes by grip', 'flow in performance', 'orients objects', and 'approaches objects' were still improved compared with before surgery, whereas 'manipulates', 'reaches', 'coordinates', 'initiates use,' and 'changes strategy' had become worse.

Active elbow extension deficit was present in 12 out of 15 children before surgery, median 18°, range 0° -35°(Fig. 2a, left). Biceps tendon lengthening was performed in nine of the children and resulted in a significant improvement of the active extension of the elbow at 7 months after OP (Fig. 2a, middle), which was largely maintained up until 9 years of





(a) Assisting Hand Assessment (AHA) score (logits converted into a 0-100 scale) pre, post (6 months), and follow-up (9 years).*Significantly different pre to post, P < 0.05. Data are presented as median and interquartile range. (b) Changes in raw scores between the assessments before surgery and at the 9-year follow-up, distributed by the separate Assisting Hand Assessment test items with mean values for the group. The items are ordered and numbered in accordance with the hierarchy of difficulty established by the Rasch analysis with the easiest item at the bottom.



(a) Extension defect (deg.) of the elbow joint in left: all included patients (n = 15) middle: all patients undergoing biceps lengthening (n = 9) right: all nonsurgery cases (n = 6). (b) Active supination (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist in all included patients (n = 15). (c) Active extension (deg.) of the wrist (n = 15). (c) Acti

follow-up (pre-OP extension deficit 30° (range: 5°-35°), 7-month post 15° (range: 0°-25°), and 9-year follow-up 15° (range: 0°-30°), pre-OP to 7-month post, and post-to 9-year follow-up P = 0.0267. However, in the children who did not undergo surgery of the elbow flexors, there was a significant reduction of the active elbow extension at the 9-year longterm follow-up (pre-OP extension deficit 5° (range: 0°-25°), post-OP 7 months 0° (range: 0°-25°) and follow-up 9 years 16.5° (range: 0°-30°), and 7-month-post-OP to 9-year-followup P = 0.0424 (Fig. 2a, right). As shown in Table 2, all children underwent surgical procedures aiming to increase active supination. Active supination increased significantly following surgery, pre-OP to 7 months post-OP P = 0.0140, and seemed to remain improved at the 9-year follow-up, but was not statistically significant [pre 0° (range: -80° to 90°), post-OP 60° (range: -50° to 90°), and 9-year follow-up 55° (range: -90° to 90°)] (Fig. 2b). All children, except one, were operated on with the aim of improving wrist extension using a variety of

Table 3 Active range of motion

surgical techniques (Table 2). Surgery improved active wrist extension more than 50° when evaluated 7 months post-OP (P=0.0185) and was still significantly improved at the 9-year follow-up (P=0.0078) compared with pre-OP [pre -10° (range: -80° to 60°), post 50° (range: -10° to 65°), and follow-up 55° (range: -70° to 70°)] (Table 3).

Results of the Zancolli's classification of active wrist and finger extension are shown in Table 4. Compared with pre-OP, at the 9-year follow-up, for four it was more difficult to extend the wrist and the fingers, for another four it was easier to extend wrist and fingers, while there was no change in seven patients. Patient 11 was operated on with a proximal row carpectomy and wrist fusion 4 years after the first surgery. He, for obvious reasons, could not extend the wrist dorsally, but with the wrist fixed at 0°, he could extend straight fingers fully, and was therefore classified as Zancolli 1.

Patients no.	Elbow active extension deficit pre-OP	Elbow active extension deficit post- OP	Elbow active extension deficit long- term follow-up	Supination active pre-OP	Supinaton active post- OP	Supination active long- term follow-up	Wrist extension active pre- OP	Wrist extension active post- OP	Wrist extension active long- term follow-up
1	0	0	20	25	45	90	-50	5	-5
2	10	5	18	40	65	55	-50	45	35
3	0	0	15	-40	60	0	50	50	55
4	30	20	20	60	60	60	30	65	70
5	10	0	15	0	90	85	40	50	60
6	20	20	10	50	80	70	45	65	55
7	25	25	30	90	0	0	- 55	50	50
8	5	0	0	- 65	-50	75	-80	40	60
9	15	5	20	-80	-40	-90	- 70	25	- 70
10	30	25	30	0	35	70	- 10	60	65
11	0	0	0	30	90	80	- 10	-10	0
12	35	20	15	- 55	-10	-90	40	40	10
13	30	15	10	60	35	38	-50	65	55
14	18	15	10	0	60	45	60	60	60
15	30	0	28	0	80	25	0	0	40
Mean	17	10	16	8	40	34	-7	41	36

Patient 11 underwent a proximal row carpectomy and wrist fusion 2 years before the long-term follow-up.

Table 4 House functional class, House classification of thumb and Zancolli classification

Patient	House 0–8 pre-OP	House 0–8 post-OP	House 0–8 long-term follow-up	House Thumb pre-OP	House Thumb post-OP	House Thumb long-term follow-up	Zancolli pre-OP	Zancolli post-OP	Zancolli long- term follow-up
1	1	1	2	4	4	4	3	3	3
2	5	5	5	2	2	0	2b	2	2b
3	6	6	5	2	2	1	2	2	2
4	5	6	5	а	а	1	2	2	1
5	6	6	5	4	3	2	2	1	2b
6	5	6	7	2	2	4	2b	2	2b
7	1	а	1	а	а	2	а	а	3
8	1	2	1	2	2	0	3	2	3
9	2	2	1	4	3	4	2b	2	3
10	3	3	3	3	2	3	2b	2	3
11	5	5	4	4	2	2	2b	2	а
12	3	3	5	3	3	4	2b	2	2a
13	4	3	5	2	2	3	2b	2b	2a
14	7	7	8	1	1	0	1	1	1
15	6	а	4	a	1	2	1	1	1

^aMissing value.

The operated hand was also classified according to the House Classification of Upper Extremity Functional Use (Table 4). Compared with pre-OP, at the 9-year follow-up, five patients still had a passive grip (House 1–3), seven still had an active grip (House 4–6), and one patient still used the hand spontaneously. In one patient, passive function of the hand had become active (House 3 to > 5), and in one patient, from active to spontaneous use (House 5 to > 7).

The patients who were treated for thumb in palm by different tendon transfers and releases are reported in Table 2. On comparing the House classification of thumb position pre-OP with the 9-year follow-up, six had a more functional thumb position, three patients had a more dysfunctional position, and there was no change in 3 (three patients had missing values at pre-OP (Table 4). Some of the patients had required more than one operation for the thumb to have an acceptable position at the initiation of a grip. Twelve of the 15 patients took part in the telephone interview. Ten reported that they were now more satisfied, at the 9-year follow-up, with the appearance of the hand and arm when walking (Fig. 3a). Similarly, 9/12 patients were more satisfied with the appearance of the hand and arm when sitting down now compared with before the surgery (Fig. 3b).

Discussion

This long-term follow-up of hand surgery in children with spastic hemiplegia shows that a progressive malpositioning of the hand can be halted with tendon lengthenings and transfers. The short-term advances in bimanual function return to pre-OP status after some years of self-selected use of the hand.

The usefulness of the assisting hand, tested with the AHA, was first improved at 7 months, and then returned to the pre-OP level at 9 years, similar to both CP and ABI. In our previously published study with short-term follow-up 6 months after hand surgery, we found that the AHA test items 'grasps', 'holds', 'stabilizes by grip', 'readjusts grip',

Fig. 3



(a) Patient-rated appreciation (0–10 scale) of the upper extremity before surgery and at the 9-year follow-up during walking. (b) Patient-rated appreciation (0–10 scale) of the upper extremity before surgery and at the 9-year follow-up while sitting down. n = 12 for both graphs.

'calibrates', 'manipulates,' and 'flow in performance' improved the most, probably because of diminished wrist flexion and pronation, which is a desired result of, for example, a Green transfer (FCU to ECRB or EDC) (Fig. 1a) [13]. A previous study has shown a correlation between AHA scores and active supination and dorsal extension of the wrist [20]. As FCU to ECRB (green) transfer and a PT release or rerouting improves dorsal extension of the wrist and supination, there seems to be a potential for improvement of the AHA post-OP. However, other studies have not been able to show an effect of hand surgery on AHA at follow-up. A multicenter study comparing (a) FCU with ECRB tendon transfer + PT release + release of AddP+rerouting of EPL, (b) standardized botulinum toxin treatment of FCU, PT, AddP, and (c) standard therapy regimen found no significant change in AHA in any group after 12 months [21]. In our study, the procedures were not standardized, but individually tailored for each child after analyzing hand and arm activity, and after assessing true contractures during the surgery. In this way, the surgery could be optimized for each individual.

Another contributing factor for our short-term AHA improvement could be that during the first 6 months after the surgery, training of the operated assisting hand was prioritized. In our regimen, the training is home based, with visits to the OT once or twice a week after the casting period, which sums up to about 20 sessions. At home and in school, the children do their normal activities, but with a special focus on the operated hand. The aim is that training of everyday tasks is repeated enough so that functional goals can withstand distractions, which probably has an impact on the short-term AHA improvement.

At our 9-year follow-up after the first surgery, the mean total AHA score had returned to the pre-OP status, proposing that in the long run, the amount of daily automatic use of the most affected hand is more important for bimanual function than is the muscle balance around the wrist. When the study started, the patients were children or adolescents who trained at the initiative of their parents or teachers, but at the long-term follow-up they had become adults, and could decide themselves whether they wanted to train their affected hand or not. Most had little motivational drive to train their hand and arm specifically. Automation of trained tasks declined, increasing the risk for dual-task interference [22-25]. The AHA test items that had deteriorated the most at long term were 'manipulates', 'coordinates', 'initiates use', and 'changes strategy', which are items that reflect inherent motor control and attentional demands the most. The item 'reaches' had also deteriorated, either because the arm is longer when children had become adults and they therefore do not have to reach as much, or that the contracture formation of the elbow flexors had progressed. However, the items 'stabilizes by grip', 'calibrates', and 'flow in performance' were still improved compared with pre-OP, implying that the improved position of the wrist makes gripping easier (Fig. 1b). However, as yet, there is no study on the natural course of the AHA into adulthood in patients with CP or ABI, only till 12 years of age, before many severe contractures have developed [8]. Progressive contractures of the wrist, thumb, and pronators will put the hand in a dysfunctional position, which may affect the AHA negatively [20]. A small decline in the AHA in absolute numbers 9 years after surgery may thus then instead be better than an assumed natural course of deterioration of the AHA score.

We know that the contracture formation starts early in the lower limb, long before evident contractures can be seen, and also in muscles with a minimal increase in tone [4,26]. A similar phenomenon occurs in the upper extremity, with progressive adduction of the shoulder, flexion of the elbow, and flexion and pronation of the wrist. Wrist flexors have been shown to be stiffer and thinner compared with typically developed patients [27], and the biceps brachii muscle has been shown to have more ECM around the fiber bundles, probably increasing stiffness, and fewer satellite cells and less ribosomal RNA as a sign of reduced growth [28].

We used the FCU muscle, as a transfer in 13/15 patients as it takes away the flexing and ulnarly deviating force that it has on the wrist, whereas it enables both supination and wrist extension when it is transferred around the ulna to finger/wrist extensors. However, long-term follow-up shows that it is common for the transfer to deteriorate. It is a muscle prone to progressive shortening, myopathic changes, and stiffening [29–32]. A transfer of the FCU therefore has the potential to become tighter over time, causing an extension contracture of the wrist, sometimes called a reversed contracture, and perhaps a static supination [27]. Six of the 15 patients in this study needed repeated surgeries because of progressive contractures, and follow-up till adulthood is therefore recommended.

The transfer could also rupture or become too loose or weak, leading to a flexed wrist again [33]. A strong side-to-side tendon transfer technique adopted from tetraplegia surgery could be a better choice [34]. One example is patient no. 9, who, at the long-term follow-up, was again unable to extend the wrist, making it more difficult grip and hold and to see what is in the hand. A person with a relapse like this could benefit from a surgical repair of the transfer or taking another muscle as a donor to the ECRB, or a proximal row carpectomy and a wrist fusion would also lead to a relative lengthening of the contracted finger flexors and a stable wrist. With an effortlessly stable wrist, more focus can be placed on flexing and extending the fingers during grip and manipulation of objects.

Even though the AHA had returned to the pre-OP level, many patients appreciated that the surgery had been performed as they liked the appearance of the hand. Less flexion of the arm during gait has been shown to correlate to better self-esteem and sense of coherence [35], and most patients in this study appreciated the look of the arm when walking. General cosmesis of the hand when sitting is also a concern [36] and has been shown to be improved after hand surgery in CP [37,38]. While interviewing these young adults, many stated that the function of their afflicted hand was still quite poor, but the surgery made it easier to hold the cell-phone and, for example, text with better hand, which is important in communicating with others. The hand looked better while resting in the lap, and the arm did not flex as much while walking or running, making the patients feel less different.

Limitations

This is a clinical study of a small number of patients with diverse impairments treated with individually tailored surgical procedures. Therefore, a prospective randomization design of different tendon transfers, etc. was not possible. The treatment methods and surgical techniques have been chosen according to the literature and clinical experience. By comparing the clinical status before surgery with both 7-month short-term and 9-year long-term follow-up, we believe that some conclusions can be made. Missing data are a common problem in clinical studies. For two patients, we replaced missing 7-month short-term AHA with the mean between pre-OP and long-term post-OP AHA (+2 and -1.5 U from pre-OP AHA, respectively), which was less than the mean increase of 6 U in the whole cohort.

Our estimations of missing values did thus not exaggerate our results or conclusions. However, ideally, a multicenter randomized-controlled study should be carried out in the future. Large-scale longitudinal studies of the development of contractures and of the AHA through adolescence would also help to interpret the effect of treatment.

Conclusion

Brain lesions in children may result in deficient motor control and progressive contracture formation in the arm, leading to flexed wrist, elbow and fingers, and diminished supination. In our study, tendon transfer surgery and tendon lengthenings/releases improved bimanual function in a short-term perspective, and returned to pre-OP function in the long term. If elbow flexors had been lengthened, active elbow extension was maintained long term, but if no surgery had been performed, contracture formation progressed. The hope is that in the future, muscles prone to growth disturbance and stiffening can be identified early by, for example, ultrasound or MRI. Prophylactic treatment could then be individually tailored, and if surgery is still needed, the optimal tendon transfer for that specific patient could be planned.

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Conflicts of interest

There are no conflicts of interest.

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