Neonatal brachial plexus palsy : impact throughout the lifespan

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CHAPTER TWO

Evaluation of shoulder function after secondary surgery in children with neonatal brachial plexus palsy

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ABSTRACT

Objective

Shoulder function in children with Neonatal Brachial Plexus Palsy (NBPP) can be impaired. Functional gain is possible by an internal contracture release and muscle tendon transfer (ICR+MTT) for external rotation. This study evaluates the functional results of this intervention.

Methods

Assessments were done pre-operatively and 3, 6 and 12 months thereafter and included joint-mobility (ROM), muscle strength, arm function (Assisting Hand Assessment (AHA) and Mallet-score), Quality of Life (QoL) (Pediatric Outcome Data Collecting Instrument (PODCI)) and parental satisfaction. Changes were examined using Wilcoxon's Signed-Rank test and Cohen's effect size.

Results

Ten children (5 boys) aged 3–10 years who underwent a combined ICR+MTT (mm. Latissimus Dorsi/Teres Major) were included. Active and passive external rotation ROM and muscle strength improved ($p < 0.05$). Arm function improved according to the Mallet-score (Hand-to-Head, Hand-to-Mouth, External-Rotation) ($p < 0.05$) and the arm use and pace scales of the AHA ($p < 0.05$). The PODCI Upper Extremity/Physical Functioning and Global Functioning subscales also showed improvements ($p < 0.05$). Parents were highly satisfied concerning daily life activities and sports.

Conclusion

ICR+MTT leads to improvement of ROM, strength, arm function, QoL and high parental satisfaction in this studies' patients and is therefore a good intervention to consider in children with NBPP with limited shoulder function.

INTRODUCTION

Neonatal Brachial Plexus Palsy (NBPP) is the result of an injury to the cervical and/or thoracic nerves (C5-T1), forming the Brachial Plexus, sustained during birth. The incidence of NBPP varies from 0.38 to 5.10 per 1000 live births in various countries.[1-3] Most injuries are mild and spontaneous recovery occurs in about 70% of the children within 4–6 months after birth. The remaining 30% is left with some kind of functional deficit. The clinical manifestations of these injuries depend on the severity of the injury and the roots involved. Children with persisting functional deficits can be either treated conservatively[1,2,4-7] or may undergo micro-neurosurgical intervention.[1,2,4-9] These treatments may not be sufficiently effective in some children[2,4,7,9], resulting in remaining functional deficits and/or anatomical changes which can become permanent or worsen over time.[10]
Limited external rotation of the shoulder is often seen in these children and can be an indication for secondary surgery (contracture-release and/or muscle tendon transfers).[5-7,9,11-23] Observational studies on the outcome of secondary surgical interventions are mainly confined to changes in active or passive range of motion (aROM and pROM) and/or Mallet-scores. Overall, improvements regarding these outcomes were reported in the literature.[11-17,19-23] Two studies described an improvement in quality of life (QoL) using either the Pediatric Data Collecting Instrument (PODCI)[24] or a questionnaire regarding the level of satisfaction with activities of daily living, cosmetics and surgical procedure.[12]
Until now, no study has been conducted which, besides the changes in ROM, Mallet scores and QoL, takes into account other relevant outcomes like muscle strength, bimanual activities and the extent to which parental satisfaction in regard to their treatment expectations are met. Improvements with respect to all of these outcomes are important goals of surgical treatment. The aim of the present study was therefore to comprehensively evaluate the results of a combined internal contracture release and a muscle tendon transfer in the shoulder performed in children with NBPP.

PATIENTS AND METHODS

Study design

This study had an observational design and was conducted between 2008 and 2011 in the Leiden University Medical Center, a tertiary referral center specialized in NBPP in the Netherlands. The institution's medical ethics committee approved the study (Studynr. P08.008). All parents gave written informed consent.

Patients

All children who were seen and physically examined by the orthopedic surgeon (principal investigator), who had an MRI of the affected shoulder and were thereafter, based on the fact that they had limited external shoulder rotation and/or joint deformities, scheduled to undergo a combined internal contracture release and a muscle tendon transfer (mm.

Latissimus Dorsi and Teres Major) were eligible for this study. Additional inclusion criteria were: Age 3–10 years, involvement of C5, C6 and/or C7 rootlets ("shoulder affected") and impairment had to be unilateral.
Fourteen children were eligible to participate. One canceled surgery, two children were excluded because one had bilateral NBPP and the second was cognitively impaired and therefore not able to cooperate. Of the 11 remaining children the parents of one child decided not to further participate after baseline due to non-compliance of the child. This participant is not included in the analyses.

Surgical intervention and postoperative rehabilitation
The following surgical procedures were employed:
Anterior internal contracture release: A deltopectoral incision was performed to expose the coracoid. An incision was then made releasing the coraco-humeral ligament at the anterior capsule of the shoulder at a length of 3 mm.
Tendon transfer: Through a curved incision at the posterior axillary border, the mm. Latissimus Dorsi and Teres Major tendons were detached from the humerus. A second incision was made cranial and posterior at the upper arm, followed by a deltoid split, exposing the humeral head. The detached mm. Latissimus Dorsi and Teres Major were transferred underneath the Deltoid muscle to the mm. Infraspinatus/Supraspinatus footprint area. Both tendons were fixed independently with transosseous sutures at the greater tuberosity of the humerus.
Rehabilitation consisted of 6 weeks baycast-plaster in slight shoulder-abduction and external rotation, followed by physical therapy twice a week for at least 3 months. Treatment consisted of maintaining/improving joint mobility and muscle strength and stimulating bimanual activities. After three months, physical therapy was either stopped or continued until no further functional recovery was seen.

Assessments
All children were seen a day prior to surgery and 3, 6 and 12 months thereafter. All outcome measurements were performed at all follow-up time points except for the bimanual activities test which was not performed at 3 months follow up. All assessments were performed by a pediatric physical therapist (first assessor) with over 5 years of experience with NBPP patients.

Sociodemographic and disease characteristics
The following data were retrieved from the medical record: age, gender, involved nerve roots, affected side and previous treatments.

Shoulder range of motion
Active and passive shoulder range of motion in the directions flexion, extension, abduction, and external rotation (in 0° and 90° abduction) were recorded with a goniometer.[25]

Muscle strength

Isometric muscle strength was measured with the MicroFET II handheld dynamometer, Biometrics, Almere, the Netherlands, using the break method.[26,27] Muscle strength of the shoulder external rotators (0° abduction), shoulder abductors (45° abduction) and shoulder flexors (45° flexion) was measured in Newton.

Shoulder movements

Shoulder movements of the affected arm were measured using the modified Mallet-score. This score measures often used arm movements, including overhead movements, with scores ranging from 1 = no function to 5 = normal function.[28-30] The Active Movement Scale (AMS) with M0 = no contraction to M4 = full motion with gravity eliminated and M5 = less than half the motion to M7 = full motion against gravity was administered for external rotation (0° abduction), abduction and forward flexion.[28,29,31]

Bimanual activities

To assess the use of the affected side during bimanual activities, including overhead movements, the Assisting Hand Assessment (AHA) was used. The AHA is a semi-structured, video-recorded, play-session for children (1.5–12 years) in which toys are used that encourage bimanual handling. Scoring is done by reviewing the video with respect to 22 items, subdivided into 6 categories: 'General Use', 'Arm Use', 'Grasp and Release', 'Fine Motor Adjustment', 'Coordination' and 'Pace', using a 4-point criterion referenced rating scale with 4 = Effective to 1 = Does not do. The minimum total raw score is 22 (0%), the maximum raw score is 88 (100%).[32-34] The total score can also be described in logit-based AHA units (0–100).[35] All play-sessions (baseline, 6 and 12 months post-surgery) were recorded and scored by the first assessor. A second assessor scored 10% of the videos. Discrepancies were discussed and by means of consensus a final score was determined. No more than 4 raw points differences were found and therefore no additional videos were assessed.

Quality of life

QoL was measured with the Pediatric Outcome Data Collecting Instrument (PODCI).[24,29,36-38] The PODCI is a questionnaire designed to assess different aspects of daily living, overall health and pain in children with musculoskeletal disorders. There are 6 scales: 'Upper Extremity and Physical Function', 'Transfer and Basic Mobility', 'Sports and Physical Function', 'Pain and Comfort', 'Happiness' and 'Global Functioning'. The PODCI was translated into Dutch using international guidelines for cross-cultural validation.[39-41] The parent reported version (2– 10 years old) was used.

Parental expectations and post-surgery satisfaction

To identify parental expectations regarding the functional outcome of the surgical intervention a self-developed questionnaire, specifically designed for this study, was used. In this questionnaire parents were asked to list all their expectations at baseline regarding two domains; Activities of daily living (ADL) and sports (including playing activities).

Subsequently they were requested to rank all expectations with respect to their importance, with 1 = the most important expectation and so on (maximum dependant on number of expectations recorded). Twelve months post-surgery, during an interview with the first assessor, parents rated the extent to which their two highest ranked expectations (one in ADL and one in sports) had been met and how satisfied they were overall concerning the functional outcome of the surgical intervention, using a 5-point Likert-scale with 1 = highly unsatisfied to 5 = highly satisfied. A score of 4 indicates an acceptable level of satisfaction.

Statistical analysis

Descriptive statistics were used to describe the clinical characteristics of the patients and the satisfaction regarding treatment expectations. Comparisons of clinical outcomes at the different time points were done by means of Wilcoxon's signed rank tests with statistical significance at $p<0.05$. At 12 months Cohen's effect size compared to baseline was computed (ES: (pre-treatment mean – post-treatment mean)/pre-treatment standard deviation). In general an ES of >0.2 is considered a small effect, >0.5 a moderate effect and >0.8 a large effect.[42] Statistical analyses were executed using SPSS 20.0 software (IBM SPSS Statistics 20.0 for Windows, http://www01.ibm.com/software/ analytics/spss/).

RESULTS

All results of the study are shown in Tables I to IV.

The high number of tests conducted gives an increased chance of a type I error occurring, however correcting for this 'multiple testing' by adjusting the p-levels may lead to an increased chance of a type II error occurring. Therefore, effect sizes, as well as the true observed values (medians and interquartile ranges) are given, whereas correction for multiple testing has not been performed.

Table I shows the characteristics of the 10 patients; 5 boys/5 girls, affected side: 3 left, 7 right, lesion-topography: 4 C5/C6, 6 C5/C6/C7. Primary treatment consisted of neurolysis (n = 1) nerve reconstruction (n = 5) and conservative treatment (n = 4). The primary surgical treatment had been conducted at the age of 4–8 months. All participants had received physical therapy during the first years of their lives.

Table II shows the changes in shoulder aROM, pROM and muscle strength. Active and passive external rotation ROM and muscle strength increased significantly at one or more time points. The differences for active external rotation in 90° abduction and passive external rotation ROM were statistically significant at 3, 6 and 12 months. Overall, a decrease in both active and passive external rotation ROM was seen between 6 and 12 months. Muscle strength in external rotation increased significantly at 12 months. All ES for external rotation including ROM in degrees, the AMS score and muscle strength in Newtons at 12 months were >0.8, indicating a large improvement. Regarding abduction and flexion, active and

Table I Sociodemographic and disease characteristics of 10 children with Neonatal Brachial Plexus Palsy undergoing a combined internal contracture release and muscle tendon transfer.

	Total group (n=10)
Gender (m/f); no.	5/5
Age, years; median (range)	4.5 (3-10)
Lesion topography; no.	
C5/C6	4
C5/C6/C7	6
Affected side; no.	
Left	3
Right	7
Previous treatment(s); no.	
Neurolysis	1
Nerve reconstruction	5
Conservative	4
Physical therapy; no.	
Yes	10
No	0

passive ROM in degrees, AMS-scores and muscle strength in Newtons, no significant changes were seen. Both active and passive shoulder extension ROM decreased significantly at 3 months, but not at 6 or 12 months.

Table III shows the changes on the Mallet, AHA and PODCI scores. Regarding the Mallet-score, no changes were seen for 'Abduction', whereas significant improvements for 'Hand to Head' and 'Hand to Mouth' were seen at all time points, and for 'External rotation' at 3 months. The 'Hand to Back' item deteriorated at 3 months. At 12 months, large ES were seen for all Mallet sub-scores, except for 'Abduction' and 'Hand to Back'. The AHA 'Arm Use' subscale improved significantly at 6 and 12 months and the 'Pace' subscale at 12 months, with large and moderate ES at 12 months. The AHA total score and all other subscale scores did not change over time. The 'Upper Extremity and Physical Function' and 'Global Functioning' scales of the PODCI also showed significant improvements at 6 months and at both 6 and 12 months respectively. All PODCI scales, except for the 'Happiness scale', showed small ES at 12 months, with the ES for the 'Global Functioning scale' being moderate.

Table IV shows the parental satisfaction regarding pre-operatively highest ranked expectations for the effect on functional improvements, as well as the overall satisfaction with treatment results at 12 months. Eight parents were highly satisfied or satisfied with the results regarding the highest ranked expectations for ADL activities, 6 parents with the results for expectations on sports activities, and 8 parents with the overall treatment outcome.

Table II Shoulder range of motion and muscle strength pre-operatively and at follow up in children with NBPP undergoing a combined internal contracture release and muscle tendon transfer. All results are expressed as median with inter quartile ranges.

Outcome measure	T1 (baseline)	T2 (3 months)	T3 (6 months)	T4 (12 months)	T1-T2 p	T1-T3 p	T1-T4 p	T1-T4 Cohen's d
Active shoulder range of motion; degrees, median (IQR)								
External rotation	-32.5 (-56.3, -18.8)	0 (-12.5, 16.3)	0 (-31.3, 12.5)	-20 (-25, 17.5)	0.018*	0.059	0.052	+1.18
External rotation (90° abduction)	2,5 (-11.3, 22.5)	42.5 (26.3, 62.5)	52.5 (35, 72.5)	45 (30, 61.3)	0.019*	0.008*	0.011*	+1.85
Abduction	150 (110, 161.3)	135 (97.5, 170)	147.5 (118.8, 172.5)	162.5 (140, 172.5)	0.280	0.443	0.103	+0.52
Flexion	147.5 (136.3, 160)	152.5 (107.8, 170)	150 (133.8, 172.5)	152.5 (140, 170)	0.644	0.720	0.592	+0.12
Extension	17,5 (0, 25)	0 (-6.3, 12.5)	10 (0, 12.5)	5 (0, 12.5)	0.049*	0.165	0.178	-0.47
Passive shoulder range of motion; degrees, median (IQR)								
External rotation	-22.5 (-26.3, 0)	30 (22.5, 42.5)	25 (16.3, 45)	20 (20, 37.5)	0.012*	0.008*	0.011*	+1.64
External rotation (90° abduction)	37.5 (15, 62.5)	70 (52.5, 82.5)	77.5 (45, 90)	72.5 (60, 86.3)	0.058	0.032*	0.017*	+1.14
Abduction	160 (128.8, 172.5)	160 (110, 170)	170 (133.8, 180)	170 (154, 180)	0.472	0.483	0.051	+0.52
Flexion	160 (153.8, 167.5)	152.5 (123.8, 166.3)	170 (143.8, 180)	165 (150, 180)	0.075	0.838	0.618	+0.21
Extension	27,5 (17.5, 37.5)	20 (-2.5, 22.5)	25 (20, 26.3)	20 (13.8, 21.3)	0.011*	0.472	0.067	-0.67
Active Movement Scale (M0-M7, worst-best), median (IQR)								
External rotation (0° abduction)	2 (1, 2.8)	5 (2, 5)	5 (4.3, 5)	5 (2, 5)	0.068	0.017*	0.027*	+1.26
Abduction	7 (6, 7)	7 (6,7)	6.5 (5, 7)	7 (6, 7)	1.000	0.317	0.157	+0.28
Flexion	7 (6, 7)	7 (6,7)	6.5 (6, 7)	6.5 (6, 7)	1.000	0.317	0.317	-0.39
Muscle strength ; Newton, median (IQR)								
External rotation (0° abduction)	0 (0, 2.5)	0 (0, 16.4)	0 (0, 23.9)	30.1 (30.1, 46.5)	0.273	0.138	0.017*	+3.81
Abduction	74,9 (23.8, 91.7)	37.5 (18.6, 59.7)	57.2 (45.9, 74.2)	70.7 (50.5, 99.9)	0.116	0.374	0.407	+0.22
Flexion	46,7 (12.8, 75.7)	25.8 (17.3, 54)	53.3 (41.5, 61.9)	56.5 (33.9, 84.4)	0.345	0.678	0.114	+0.50

IQR: inter quartile range
* Significant difference p<0.05
Effect size Cohen's d; >0.2 = small change, >0.5 = moderate change, >0.8 = large change.

Table III Mallet, Assisting Hand Assessment and Pediatric Outcome Data Collecting Instrument scores pre-operatively and at follow up in children with NBPP undergoing a combined internal contracture release and muscle tendon transfer. All results are expressed as median with inter quartile ranges.

Outcome measure	T1 (baseline)	T2 (3 months)	T3 (6 months)	T4 (12 months)	T1-T2 p	T1-T3 p	T1-T4 p	T1-T4 Cohen's d
Malletscore (I-V, worst-best), median (IQR)								
Abduction	4 (4, 4)	4 (3.8, 4)	4 (4, 4)	4 (4, 4)	0.317	0.317	0.317	+0.32
Exorotation	1 (1, 1.3)	2 (1, 3)	2 (1, 3)	1 (1, 3)	0.040*	0.064	0.167	+1.42
Hand to Head	3 (2.8, 3)	4 (3, 4)	4 (4, 4)	4 (3, 4)	0.023*	0.008*	0.008*	+1.23
Hand to Back	4 (2, 4)	2.5 (2, 3.3)	3 (2, 4)	4 (2, 4)	0.038*	0.102	0.317	-0.21
Hand to Mouth	2 (2, 2.3)	4 (3, 4)	4 (3, 4)	4 (3, 4)	0.004*	0.006*	0.006*	+3.55
Assisting Hand Assessment; median (IQR)								
Total score (0-100 logit based AHA units)	82 (76, 83)	x	85 (72.8, 86.3)	86 (73.8, 86.3)	x	0.095	0.095	+0.20
General Use items (%)	100 (83.4, 100)	x	100 (75, 100)	100 (91.7, 100)	x	1.000	0.102	+0.24
Arm Use items (%)	62.5 (56.8, 66.7)	x	75 (56.2, 75)	75 (58.3, 83.3)	x	0.016*	0.007*	+0.95
Grasp-Release items (%)	100 (84.5, 100)	x	100 (75, 100)	100 (67.8, 100)	x	0.854	0.197	-0.16
Fine Motor Adjustment items (%)	100 (69.5, 100)	x	100 (72.3, 100)	100 (75, 100)	x	0.257	0.705	+0.04
Coordination items (%)	100 (95.8, 100)	x	100 (95.8, 100)	100 (95.8, 100)	x	0.317	0.317	+0.15
Pace items (%)	83.4 (66.7, 88.9)	x	88.9 (77.8, 88.9)	88.9 (88.9, 88.9)	x	0.074	0.041*	+0.50
Pediatric Outcome Data Collecting Instrument (PODCI); median (IQR)								
Upper Extremity scale (range 0-100)	71 (50, 84.5)	66.5 (54, 94.3)	73 (57.8, 89)	80 (61.8, 84.8)	0.866	0.043*	0.313	+0.22
Transfer and Basic Mobility scale (0-100)	97 (93.3, 100)	98.5 (94.5, 100)	98.5 (94, 100)	100 (96.3, 100)	0.343	0.197	0.072	+0.42
Sports and Physical Functioning scale (0-100)	88.5 (78.5, 96.3)	88 (80.5, 90.3)	94 (89, 96.5)	95 (87.8, 98)	0.213	1.000	0.068	+0.46
Pain and Comfort scale (0-100)	100 (76.3, 100)	89 (70, 100)	100 (98.3, 100)	100 (93.5, 100)	0.131	0.655	0.109	+0.24
Happiness scale (0-100)	100 (85, 100)	90 (75, 100)	95 (82.3, 100)	100 (93.8, 100)	0.068	0.655	0.109	+0.05
Global Functioning scale (0-100)	88 (75.5, 90.5)	85 (78, 92.3)	90 (86.3, 95.8)	90.5 (80.8, 95.5)	0.735	0.041*	0.024*	+0.57

IQR: inter quartile range
x: The assisting hand assessment was not recorded at T2.
* Significant difference p<0.05
Effect size Cohen's d; >0.2 = small change, >0.5 = moderate change, >0.8 = large change.

Table IV Parental expectations and satisfaction pre-operatively and at follow up in children with Neonatal Brachial Plexus Palsy undergoing a combined internal contracture release and muscle tendon transfer.

Patient no. (age at baseline in years)	No. of expectations at T1 (baseline)	Primary expectation ADL at T1; improvement in:	Satisfaction at T4 (12 months) range 1-5*	Primary expectation Sports at T1; improvement in:	Satisfaction at T4 (12 months) range 1-5*	Overall satisfaction at T4 (12 months) range 1-5*
1 (5)	6	Bringing something to the mouth	3	Swimming	5	5
2 (4)	2	Cycling	5	Swimming	5	5
3 (7)	3	Running	2	Cycling	2	2
4 (3)	4	Eating	4	School gymnastics	5	5
5 (3)	5	Dressing	5	None	3	4
6 (8)	5	Cycling	4	Swimming	4	4
7 (4)	6	Dressing	4	Swimming	4	4
8 (10)	8	Personal hygiene	5	School gymnastics	2	3
9 (3)	2	Placing hand on and above head	5	None	3	5
10 (6)	4	Dressing	5	Swimming	5	5

* Post surgery satisfaction range 1; highly unsatisfied – 5; highly satisfied.

DISCUSSION

In case of persistent external rotation limitations in children with NBPP, secondary surgery consisting of a combined internal contracture release and a muscle tendon transfer (mm. Latissimus Dorsi and Teres Major) can be considered. The current study in 10 children found that shoulder external rotation ROM and strength, bringing the hand to the head and to the mouth, the use of the affected arm in bimanual activities, and overall (arm) function improved significantly in the year following this intervention. A negative effect on shoulder extension ROM and bringing the hand to the back was seen. The majority of parents were satisfied with the result after 12 months.

The results are generally in line with the literature. Concerning shoulder external rotation ROM, positive effects were also reported in 13 other studies.[11-13,15-17,19-24,43] With respect to shoulder abduction, previous studies reported an increase[11-13,15-17,19-24,43], whereas in this

study no significant effect on abduction was seen. This could be due to the relative good shoulder abduction and flexion ROM, AMS scores, strength and Mallet scores before surgery. Improvements of functional movements of the arm, including bringing the hand to the head and mouth have been reported earlier as well.[20,22,23] The same holds for the negative effect on bringing the arm to the back.[15,17]

In general, the effect on shoulder external rotation in ROM and on Mallet scores decreased between 6 and 12 months. This decline might be related to the observation that for most daily activities only a limited range of shoulder external rotation, especially in 0° abduction, is needed. External shoulder rotation in daily activities is usually combined with some shoulder abduction/flexion. It might also be related to the fact that external rotation exercises were only performed during the physical therapy period (first 3 months after baycast-plaster removal). The question remains to what extent external rotation in 0° abduction is of clinical importance regarding the performance of daily activities. Still, loss of external rotation in 0° abduction is one of the parameters indicating the need for secondary surgery.[44,45] Ultimately, active ROM is a composite that will determine overall functionality and thus quality of life of the child.

No previous study included a measurement of muscle strength so far. Although it is difficult to perform muscle strength measures in young children using a handheld dynamometer, it is a well-known and usable assessment instrument for children. Reference values of maximum isometric muscle force obtained with a handheld dynamometer are available for children between the age of 4 and 16.[26] It remains to be established though to what extent the gain in muscle strength seen in the present study contributes to the overall increase in arm function.

Few studies have so far focused on daily activities and quality of life. Regarding bimanual activities, the AHA was never used before in patients undergoing the described intervention. In this study a small, yet positive change in the 'Arm Use' items was seen. The overall AHA score however did not change significantly. This may be due to the relatively good hand function most of our patients had before surgery.

Concerning quality of life, a positive effect for the PODCI scales 'Upper Extremity and Physical Function', 'Sports and Physical Function' and 'Global Functioning' was seen in previous studies[24], where as the present study showed no effect on 'Sports and Physical Function'. In line with the previous studies, no improvements were seen for the other three scales.[24] This lack of effect may be related to the fact that the PODCI is not specifically designed for upper extremity conditions nor for NBPP.[29] For the sake of efficiency in future research in patients with NBPP it could be considered to use only the PODCI 'Upper Extremity and Physical Function' scale.

In concordance with the results of the present study, a relatively large proportion of parents were satisfied with the intervention in previous research as well.[12] Measuring function in children with NBPP is difficult, because most of the time they are fully functional in their own way by employing compensational strategies. Moreover, no consensus exists on how to measure this function in these children.[46] Nevertheless, it is still important to evaluate the effectiveness of surgical interventions in children with NBPP. Children reported functional problems due to NBPP in a recently conducted focus group study[47] and the parents of the children in the current study reported functional problems as well. These findings underline the importance of an evaluation on a functional level.

This study had a number of limitations. First, a relatively small group of patients was used. This is due to the fact that the prevalence of NBPP is fortunately low these days. Therefore, the present study and some previous papers reporting on this surgical procedure included moderate to small groups of patients.[11-13,15-17,20,23] In previous studies, the range of follow-up and age was even wider than in the present study. Moreover, the characteristics of the patients at baseline varied. Although this might be the case, all patients have a limited external shoulder rotation and limited arm function in regard to the Mallet score and the AHA in common. Because of the relatively large number of tests and time points in a small sample so called 'multiple testing' has occurred. Because of this a type I error cannot be ruled out. Correcting for Multiple Testing can be done by adjusting the p-levels, however this method increases the chance of a type II error.[48] To counteract this problem, effect sizes were calculated with predefined cut off points for their interpretation. Overall, the use of effect sizes and the fact that the observed scores and the magnitude of their changes are in line with those reported in the literature, suggest that the changes seen in this group of patients are real and not a result of a type I error.[48] It remains to be established though whether the results are generalizable to other patients with NBPP and external rotation limitations. Another limitation was the observational design, with no control group. However, it is questionable whether using a control group is ethical in this study population. In contrast with most of the previous studies however[11-13,17,20,21], data was gathered prospectively with standardized timing of assessments, using well defined outcome measures allowing an in-depth analysis including multiple components of the ICF-CY (Children and Youth) as well as parental satisfaction.[49]

CONCLUSION

This study showed that a combined internal contracture release and a muscle tendon transfer (mm. Latissimus Dorsi and Teres Major) for external shoulder rotation is a good intervention to restore arm function in children with NBPP. External rotation mobility and muscle strength, hand to head and hand to mouth Mallet-score items, AHA 'Arm Use' items and general functioning increased. Parents were overall highly satisfied with their expectations concerning both daily life activities and sports being met. The results of this

study are important for parents and children as more detailed information on the expected treatment outcomes can contribute to the quality of the decision-making process.

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