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Neonatal Brachial Plexus Palsy: the role of diminished sensibility of the hand on functional recovery

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Chapter 6

Gripforce Reduction in Children with an Upper Neonatal Brachial Plexus Palsy

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Abstract

Objective

The aim of this study was to assess gripforce in children with a C5 and C6 neonatal brachial plexus palsy (NBPP) as it may affect hand use. Applying classic innervation patterns, gripforce should not be affected as hand function is not innervated by C5 or C6. Here we compare gripforce in NBPP with a healthy control group and assessed correlations with hand sensibility, bimanual use and external rotation.

Patients and Methods

50 Children with NBPP (mean age 9.8 y) and 25 controls (mean age 9.6 y) were investigated. Nerve surgery had been performed in 30 children and 20 children had been treated conservatively. Gripforce of both hands was assessed with the Jamar dynamometer. Sensibility of the hands was assessed with two-point discrimination and Semmes-Weinstein Monofilaments. External rotation was assessed using the Mallet score. Bimanual use was measured by using one of three dexterity items of the Movement Assessment Battery for Children-2. The affected side of the NBPP group was compared with the non-dominant hand of the control group using one-way ANOVA, chi-squared and Mann-Whitney tests.

Results

The mean gripforce of the affected non-dominant hand of children with NBPP was diminished as compared to healthy controls (95 N and 123 N, respectively, with $p = 0.001$). The mean gripforce of the non-dominant hand in the control group was 92% of that of the dominant hand, while it was only 76% in the NBPP group ($p = 0.04$). There was no relationship between gripforce reduction and sensibility, bimanual use or shoulder external rotation.

Conclusions

The gripforce in NBPP infants with a C5 and C6 lesion is lower than that of healthy controls although classic interpretation of upper limb innervation excludes this finding. The reduction of gripforce in upper NBPP lesions is not widely appreciated as a factor inherently compromising hand use. The reduction of gripforce should be taken into consideration in planning the type of rehabilitation and future activities.

INTRODUCTION

Neonatal brachial plexus palsy (NBPP) is a nerve traction injury that occurs during birth. The most common type involves a lesion of the two upper spinal nerves C5 and C6. In more severe cases, spinal nerves C7, C8 and T1 are involved as well, see Malessy & Pondaag.[1] Classic anatomical innervation schemes indicate that C5 mainly innervates the deltoid, supraspinatus and infraspinatus muscles, whereas the biceps, brachialis and brachioradialis muscles are mainly innervated by C6, see Merle d'Aubigné & Deburge.[2] An upper lesion should, therefore, only affect shoulder functions and elbow flexion. Our clinical observation is, however, that children with an upper NBPP employ their hand less often and the dexterity of the hand seems diminished. Anecdotally parents affirm this observation. A satisfactory explanation for these phenomena are difficult to provide. After all, hand function in itself should not be affected in upper plexus injuries because the muscles of the hand are innervated by the lower nerves of the brachial plexus, C8 and T1.Three factors could theoretically affect hand use: 1) a diminished positioning of the hand in space, 2) a diminished sensibility and 3) gripforce reduction. Research has shown that recovery of glenohumeral external rotation is limited after conservative management or nerve surgery reaching beyond the sagittal plane in only 20% of patients, see Pondaag et al.[3] Limitations in external positioning may affect the development of the preferred hand for writing and playing, see Krumlinde-Sundholm et al.[4] Sensibility of the thumb and index finger in children with an upper plexus lesion (whether treated surgically or conservatively) is also diminished, see Buitenhuis et al[5] and Anguelova et al[6] which is correlated with diminished dexterity see Buitenhuis et al.[5] Some studies have been performed in children with an upper palsy to explore whether the use of the hand is reduced because of a reduction of gripforce. The applied methodologies in these studies, however, leave doubts as to the value of the results. Namely, the affected side was compared to the non-affected side of the individual child, see Krumlinde-Sundholm et al[4], Strombeck et al[7] and Kirjavainen et al[8]. Regardless of the outcome, this type of comparison does not discriminate whether findings are caused by a relative increase of gripforce in the non-affected side, by hand dominance or by an actual reduction of intrinsic gripforce of the affected hand. In addition, the criteria used to define a reduction of gripforce were chosen quite arbitrary and were based on measurements in

adults. To overcome these issues, potential gripforce differences can only be addressed in the setting of a comparison with a healthy control group of the same age.
In the present study we compared the gripforce of the hand in children with an upper NBPP with healthy controls. In addition, we correlated gripforce with hand sensibility, dexterity/bimanual use and glenohumeral external rotation, in order to gain a better understanding of the bimanual use of the hand.

PATIENTS AND METHODS

We used a cross-sectional investigation design of patients with NBPP. Fifty children with an upper NBPP and 25 healthy children were recruited for the study. Ages of both groups ranged between 7 and 12 years. The children with NBPP had been examined on a regular basis from an early age at our tertiary referral clinic (Nerve Center of the Leiden University Medical Center, The Netherlands). The diagnosis of NBPP was based on the obstetrical history and the neurological examination and additional electromyography examination performed between the ages of 4 and 6 weeks, see Malessy et al.[9] Nerve surgery was performed in 30 children (60%) in early infancy, while 20 had been treated conservatively. Based on the neurological examination, all children only had a lesion of the C5 and C6 spinal nerves. At the first visit to our clinic, all participants had a normal hand function, normal elbow extension based on active triceps muscle and active wrist extension at least against resistance. Hence, these children were diagnosed with a C5-C6 lesion, with intact C7-C8-T1 functions. The children who were conservatively treated showed recovery of elbow flexion with active biceps muscles at 3 to 6 months of age.
The indication for nerve surgery has been extensively described by Malessy & Pondaag.[1] Children who were operated upon underwent MRI myelography to assess root avulsion injuries. During the operation, surgical inspection and direct nerve stimulation were performed to confirm the clinical diagnosis. To restore C6 function, grafting from C6 to the anterior division of the superior trunk (ADST) was performed in 23 of the 30 infants. Of the remaining seven patients, five had a medial pectoral nerve to musculocutaneous nerve transfer. In one surgically treated patient, accessory to suprascapular nerve transfer was the sole procedure, and in one other patient surgery was limited to neurolysis. In these seven children, the nerve pathway from C6 to the ADST (containing the sensory fibres of the C6

dermatome) was in continuity. This sub-group of seven patients was additionally compared to the 23 infants in which C6 was grafted to the ADST.

The control group was recruited at the Montessori school at Voorburg, the Netherlands, by announcing the study on the school's message board. All children who participated had a normal cognitive function and attended regular school, see Buitenhuis et al.[10]

Physical examination

The physical examination of all participants was performed by one physical therapist (SB). with a huge experience of physical assessment and treatment of children in all age groups. The gripforce of both hands was assessed with the Jamar dynamometer, according to a standard protocol, see Molenaar et al.[11] The child was sitting with the elbow and forearm resting on a table, with the wrist in a neutral position between pronation and supination. The shoulder was positioned in 0^o anteflexion, 0^o abduction and 0^o external rotation. If this position was not possible due to lack of external rotation, the upper arm was held in a resting position in internal rotation. The dominant hand was tested first. The child was asked to squeeze the handles of the Jamar dynamometer as forcefully as possible. Three attempts at maximum force were recorded, and the mean of the three values was calculated. Before the three measurements were done, we instructed the children very well to do their utmost best to perform as well as they could. Also, during the testing we encouraged them to use the maximum of their abilities. The affected side was compared with the non-affected side within the NBPP group. Additionally, we compared the non-dominant affected side of the NBPP group with the non-dominant hand of the control group.

The dominant hand was defined as the hand in which a child holds a pencil to write. A hand preference shift in the NBPP group was presumed to have occurred if a child with a right-sided lesion had left-hand dominance, see Yang et al.[12] When the dominant side was the affected side, children were excluded from analysis.

The sensibility of the hands was assessed with two-point discrimination (2PD) (Bell-Krotoski et al[13] of the index finger and the Semmes-Weinstein Monofilament test (SW) (Weinstein[14]) of the thumb of the non-dominant side in the NBPP group. External rotation was assessed using the relevant Mallet sub-score, see Mallet.[15] A score of Mallet I signifies no active

external rotation. Mallet II indicates < 0° active external rotation. Mallet III represents active external rotation between 0–20°. Mallet IV means >20° active external rotation.
The combined use of both hands was measured by a single item from the three dexterity items of the Movement Assessment Battery for Children-2 (MABC-2), an internationally accepted and validated test for fine motor skills by Schulz et al.[16] For children aged 7, 8, 9 or 10 the specific bimanual task consisted of threading a wire through holes in a board. Children aged 11 or 12 years were instructed to construct a triangle with nuts and bolts in correspondence with MABC-2. We selected this bimanual task because it requires employment of the affected hand. Children were not allowed to put either the wire or the triangle on the table, but were instructed to keep them in both hands. The time needed to finish the task was noted and converted to a standard score, and corrected for age using the MABC-2 manual. We have reported the sensibility and the dexterity results of the NBPP and control groups in a previous paper, see Buitenhuis et al.[5]

The study protocol was approved by the Medical Ethics Committee of the Leiden University Medical Centre (ABR No. 48977) and informed consent was given by the parents.

Statistical analysis

We used analysis of variance (one-way ANOVA) for continuous outcome variables. Categorical outcome variables were compared between groups using chi-squared tests (exact tests if the expected counts were small). Where appropriate, a Mann-Whitney test was used instead of a t-test. The error level was set at $p < 0.05$. Data were analysed with SPSS Statistics for Windows, version 23 (IBM Corp. Armonk, NY).

RESULTS

Patient details are presented in Table 1.
We compared the affected (non-dominant) side in the NBPP group with the non-dominant side of the control group and we compared the difference in gripforce between the dominant and non-dominant hand within the NBPP group, respectively, within the control group. This analysis of the 'affected non-dominant hand' concerned 28/30 surgically treated children and 14/20 conservatively treated children, see Table 2.

Table 1 Patient characteristics

Demographic variables	NBPP group	Control group
Total number	50	25
Nerve surgery	30	NA
Conservatively treated	20	NA
Mean age (years)	9.8 (SD 1.89)	9.5 (SD 1.46)
Boys/girls	22/28	8/17
Affected side left/right	26/24	NA

Table 1 Legend

NBPP: Neonatal Brachial Plexus Palsy; NA: Not Applicable; SD: Standard Deviation.

Table 2 Hand dominance versus affected side

Group				Dominant hand	
				Left	Right
Controls				1	24
NBPP	Conservative	Affected side	left	-	11
			right	3#	6*
	Nerve surgery	Affected side	left	-	15
			right	13#	2*

Legend Table 2

NBPP: Neonatal Brachial Plexus Palsy.

presumed preference shift: a hand preference shift was presumed to have occurred if a child with a right-sided lesion had left-hand dominance; * lesion on dominant side: not included in the analysis.

In the surgically treated group, a hand preference shift was found in 13/15 children (87%), while in the conservatively treated patients, a hand preference shift was found in 3/9 children (33%). This difference was statistically significant (p = 0.007). We cross-checked for gender or age as a confounder, as gripforce was shown to increase with age, and boys are usually stronger than girls, see Molenaar et al.[17] Our NBPP group was 0.3 years older than the control group, and gender did not influence the results, thereby ruling out these confounders.

The mean gripforce of the non-dominant affected hand was statistically significant reduced in the NBPP group as compared to the non-dominant hand of the controls (95 N and 123 N, respectively, with p = 0.001). The mean gripforce of the non-dominant hand in the control group was 92% of that of the dominant hand. The gripforce of the non-dominant hand of NBPP group was 76% of that of the dominant hand (p = 0.04). (Figure 1 and Table 3I)
Gripforce did not differ statistically between the conservatively and surgically treated subgroups. The mean gripforce was 75% of the unaffected side after nerve grafting (n = 22); 81% after nerve transfer (n = 5), and 78% after conservative treatment (n = 14).
The gripforce of the non-injured dominant hand was diminished 10% in children who had undergone nerve surgery compared to controls (121 N versus 134 N; p = 0.20), and 15% in children who shifted dominance (114 N versus 134 N; p = 0.13), see Table 3.

Table 3 Gripforce of the hand (Newton)

		Mean* (SD)	
Group		**Dominant**	**Non-dominant**
Controls	(n = 25)	134 (42)	123 (42)
NBPP	All (n = 41)	125 (45)	95 (38)
	Conservative (n = 14)	132 (41)	100 (30)
	Nerve surgery (n = 27)	121 (48)	92 (42)
	Presumed preference shift (n = 16)	114 (42)	92 (45)
	No presumed preference shift (n = 25)	128 (47)	94 (33)

Legend Table 3

NBPP: Neonatal Brachial Plexus Palsy; SD: Standard Deviation; n = sample size.
* The mean gripforce of the non-dominant affected hand in the NBPP group differs significantly from the non-dominant hand of the control group (p = 0.001, t-test); there was no significant difference between conservative treatment and nerve surgery in the NBPP group for both the non-dominant affected hand (p = 0.34) and for the dominant non-affected hand (p = 0.42). The difference between the children with and without a presumed preference shift was not significant for the non-dominant affected hand (p = 0.92) and for the dominant non-affected hand (p = 0.32). The difference between the dominant hand after a presumed preference shift and the dominant hand of the control was not significant (p = 0.14).

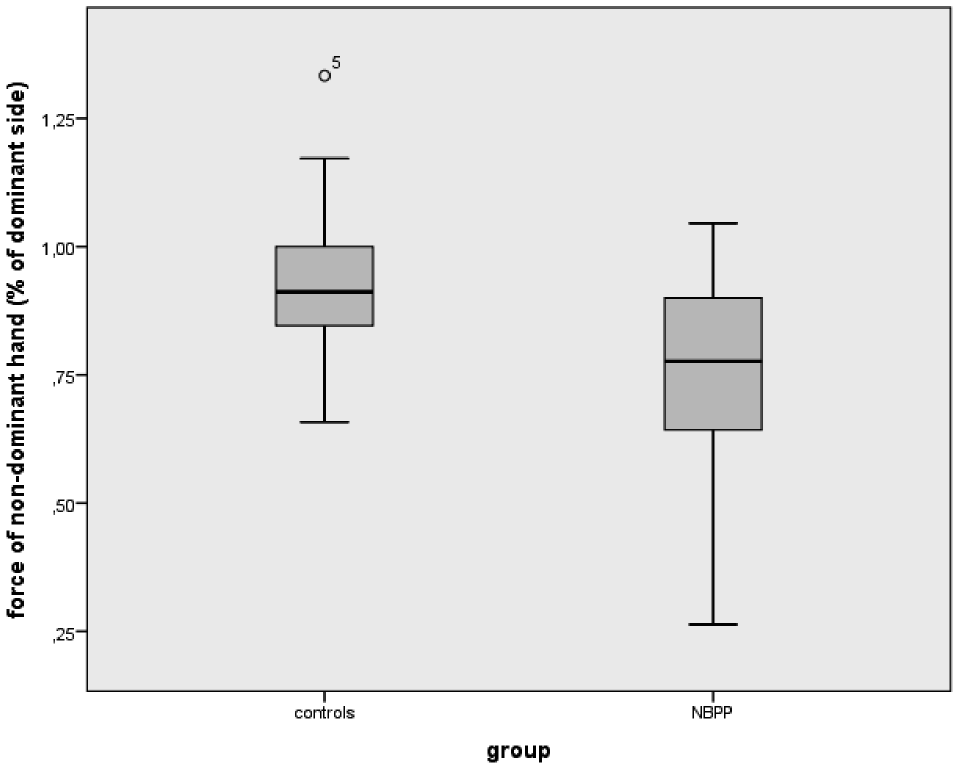

Figure 1 *Gripforce of the non-dominant hand as a percentage of the dominant side*

Legend Figure 1

Controls: mean gripforce of the non-dominant hand was 92% of that of the dominant hand; NBPP (neonatal brachial plexus palsy)-group: mean gripforce of the non-dominant hand was 76% of that of the dominant hand.

In the MABC2 bimanual use test, the children in the control group (n = 22) had a mean test score of 11.0, compared with a mean test score of 8.0 in the NBPP group (n = 50). The higher test score signifies that the controls perform the bimanual use test faster than the children with NBPP, which was statistically significant (p = 0.036). Due to a slight age difference between patients and controls, 31/50 (62%) of the children with NBPP performed the wire thread test compared with 20/22 (91%) of the controls. We additionally analysed the wire thread test only to rule out different outcomes due to the difference between the wire thread test and triangle construction test. The time to thread the wire through holes in a board, was 29.9 seconds in the NBPP group (mean age 10.1 year) compared with 23.9

seconds in the control group (mean age 9.1 year). This difference in time was also significant (p = 0.01).

Shoulder external rotation scores were Mallet grade I in 16 children with NBPP; Mallet II in 18; Mallet III in 12; and Mallet IV in 4.
We found no correlation between sensibility and gripforce. Nor did we find a correlation between gripforce and the bimanual use test (Pearson Correlation coefficient: 0.092, p = 0.47). We found no correlation either between gripforce (corrected for age) and the Mallet subscore for external rotation (p = 0.57).

DISCUSSION

This study was performed to analyse whether gripforce is reduced in children with an upper NBPP lesion following the clinical observation that the affected hand is used less. This research question was supported by the observation of others that a shift of hand preference occurs in many children with NBPP, see Yang et al.[12]
In the present study, we found a reduced gripforce of the affected side which was 76% of the unaffected side. In the healthy control group, the mean gripforce of the non-dominant side was 92% of that of the dominant hand. The findings in the control group matches those of a previous report in which a 94% ratio was found by Molenaar et al.[11]
Our findings take away the doubts that still existed as regard to gripforce levels in upper NBPP lesions. Previously, it was stated that 50% of children with C5–C6 lesions have a reduced gripforce, see Krumlinde-Sundholm et al[4] and Strombeck et al[7]. A Martin Vigori meter consisting of a rubber bulb connected to a manometer was used. The bulb had to be squeezed 3 times and the highest value for each hand was recorded. Gripforce was regarded as reduced when it was 20% less than the strength in the unaffected hand. The cut-off point of 20% was chosen based on gripforce measurements with a dynamometer in adults, see Petersen at al[18], Strombeck et al[19] and Bechtol[20], which is, in the setting of children, quite arbitrary. In another study another cut-off point was applied, namely more than 89% of the unaffected hand. It was found that only 18% of children with NBPP with a C5-C6 injury had a normal gripforce ratio, see Kirjavainen et al.[8] The discrepancies between the studies of Kirjavainen et al[8], Petersen at al[18] and Bechtol[20], illustrate the effect of choosing different criteria on outcome, and create doubts as to its value.

The factors that cause a decrease of hand gripforce in NBPP with C5-C6 lesions need still to be determined.

A neuroanatomical explanation for the innervation of gripforce seems unlikely, as the long flexors of the fingers are innervated by C8 and T1, which should be normal in children with an upper trunk lesion. Indirectly, the innervation of wrist extension might play a role, as stable wrist extension is essential for a strong hand grip. Electrophysiological studies have shown that the nerve fibers innervating the extensor carpi radialis muscle arise from C5 and C6, see Zhang et al.[21] A reduced innervation of wrist extension could therefore contribute to a decrease of gripforce. In our experience, however, in lesions limited to the C5-C6 spinal nerves we never observe prominent reduction of wrist extension when we resect a neuroma of the superior trunk followed by nerve grafting. This implies that there is sufficient innervation from C7 and C8 to maintain a proper wrist extension with the extensor carpi radialis and ulnaris muscles, see Zhang et al.[21] All in all, it seems therefore unlikely that neuroanatomical factors are involved in the reduction of gripforce in C5-C6 NBPP lesions.

An indirect explanation of the reduced gripforce of the hand on the affected side in children with NBPP might be the reduced use the affected side less often, and this in turn may lead to a decreased force. The factors causing less use of the hand caused by impaired spatial positioning due to a limited shoulder function, and diminished dexterity due to reduced sensibility. These factors have been investigated and it has been shown that the ability to incorporate the affected arm and hand in a co-ordinated movement pattern correlated with sensation and prehension of the hand, but not with shoulder and elbow function, see Dumont et al.[22]

We found no statistical correlation between gripforce and sensibility, between gripforce and external rotation, or between gripforce and bimanual use. Because of the absence of such correlations, a direct relationship seems unlikely. One might cautiously conclude that other factors, which have so far not been defined or measured, play a causative role. One of these might be cerebral control which is potentially disturbed in the development of central motor programs. In clinical observations and fMRI data, we found evidence of changes in central control see Anguelova et al.[23] [24] It was previously hypothesized that a diminished tactile input to the brain could explain diminished embedding of movement of the affected arm, which was coined ‘developmental apraxia’, see Brown et al.[25] Strombeck et al[26] concluded considerable EMG changes observed in NBPP, even within fully recovered children. We previously assessed sensibility with 2PD and SW filaments, and found that the index finger is the most sensitive finger tested with the 2PD test, and the thumb with the SW, see Buitenhuis et al.[10] Sensibility of thumb and index finger are essential to perform fine motor tasks, and it proved to be these fingers that showed diminished sensibility in the children with an upper trunk lesion. The absence of a correlation between gripforce and sensation in this study, does not exclude a role of sensation. Disruption of proprioception might be of more relevance, rather than the tactile sensation we tested, see Figure 2.

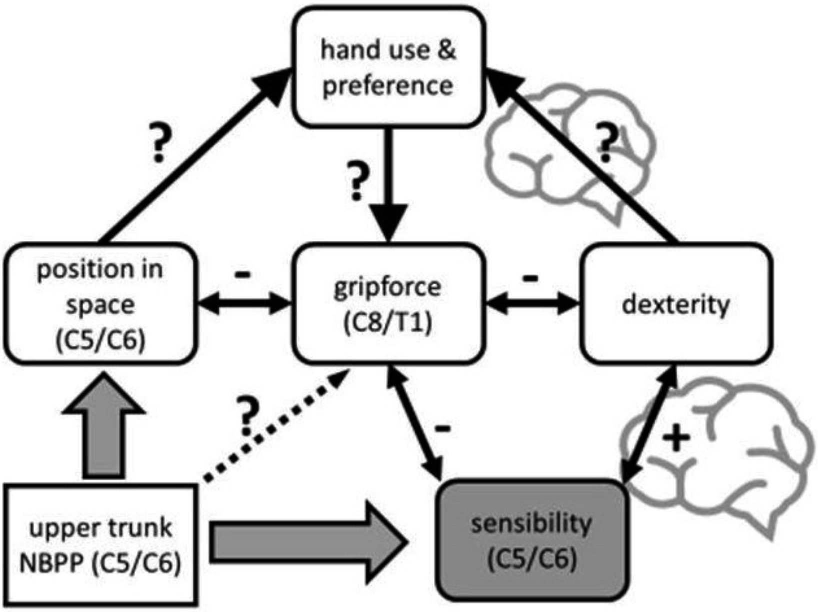

Figure 2 Potential correlations between sensibility, gripforce and dexterity to explain diminished hand use in children with NBPP

Legend Figure 2

– no correlation (current paper); **+** correlation according to Buitenhuis et al[5]

An attempt to explain diminished hand use in children with an upper trunk NBPP lesion. Diminished upper trunk function results in diminished sensibility of the hand and diminished shoulder function, but cannot anatomically explain diminished gripforce. Cerebral control may play a role to connect sensibility and hand use.

For the execution of a fine coordinative movement, such as threading a wire through holes in a board or constructing a triangle with nuts and bolts, both proprioceptive and tactile sensibility are more important than force.

In this series, the non-injured dominant hand had a 10-15% decreased gripforce compared to the dominant hand in controls both in children who had undergone nerve surgery as well as in children who had a presumed dominance shift. These differences were not statistically significant, and may be a result of chance. However, we feel that this observation deserves further study as the absence of statistical significance can also be caused by the relatively limited size of our cohort. Not surprisingly, a dominance shift occurred frequently in surgically treated children who suffered a more severe nerve lesion than conservatively treated children. We hypothesize that a shift of hand dominance affects the dominant non-injured side at the cerebral levels of movement control, causing an additional disadvantage for learning bimanual activities. A cortical dominance shift has been described earlier in relation to speech development, see Auer et al.[27]

Finally, our findings may be relevant for strategic choices in brachial plexus repair. Fascicles from the ulnar and median nerves are used as donors in transfers to the biceps and brachialis motor branch to reanimate elbow flexion in case of root avulsion in NBPP. Although the use of these fascicles might jeopardize hand function development, hand dysfunction was not found following the use of either the ulnar or median fascicle in a small series of 8 patients, see Siqueira et al.[28] In this series, however, only the affected hand was examined, and findings were not related to the non-affected side or healthy controls. Since we show that gripforce is reduced even in upper lesions, it might actually be the case that more hand function is lost due to the application of this technique than is currently appreciated.

One limitation of our study is that the participating children were followed at our tertiary referral clinic, and as a result, surgically treated children were overrepresented in our sample. Surgical procedures were diverse, but gripforce did not statistically differ in different surgical groups. Additionally, children with a good clinical recovery after surgery or conservative treatment usually do not have a long follow-up and are, therefore, underrepresented in the current study. Another limitation of our study is that we did not systematically document whether the use of the hand was diminished, but rather documented it anecdotally when parents reported it during visits of our clinic. Future studies should include patient reported outcome measures for example the Hand-Use-at-Home questionnaire to document the frequency of hand employment, see Van der Holst et al.[29]

In summary, various explanations have been offered for the diminished hand use in children with NBPP with an upper palsy. (Figure 2) In the current study, children appeared to have a diminished gripforce. This finding was not directly correlated to diminished sensibility or other factors.

More research is needed to fully understand the diminished hand usage and gripforce in upper trunk NBPP. It is advised to assess the dominant non-injured hand in future cohorts, and its role in dexterity and bimanual activities. This may ultimately provide clues for designing tailored physical or occupational therapy to improve hand usage.

CONCLUSION

We found a reduced gripforce of the hand in children with an upper neonatal brachial plexus lesion, which we hypothesize to be caused by diminished use of the hand, and diminished cerebral control. Additionally, the non-injured hand had diminished grip force, especially in children with a presumed dominance shift, which may further impair their bimanual ability.

We did not find a relationship between gripforce and sensibility, bimanual use or shoulder external rotation function.

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