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

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CHAPTER 3

Sensibility of the hand in children with conservatively or surgically treated upper neonatal brachial plexus palsy

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ABSTRACT

Objective

The aim of this study was to assess the sensibility of the hand in children with a neonatal brachial plexus palsy (NBPP) involving the upper nerves C5 and C6, and to correlate the results with dexterity.

Patients and Method

Fifty children with NBPP (mean age 9.8 y) and 25 healthy controls (mean age 9.6 y) were investigated. In the NBPP group, nerve surgery had been performed in 30 children and 20 children had been treated conservatively. Sensibility was assessed with two-point discrimination (2PD) and Semmes-Weinstein (SW) monofilaments. Dexterity was evaluated with a single item from the Movement Assessment Battery for Children-2. We compared the affected side of the NBPP group with the non-dominant hand of the control group.

Results

The sensibility in the first and second fingers was significantly lower in the NBPP group than in the control group for both the 2PD ($p = 0.005$ and $p = 0.014$ respectively) and the SW monofilament test ($p<0.001$). Dexterity was significantly lower in the NBPP group than in controls, corrected for age ($p = 0.023$). There was a significant difference toward decreasing hand function with decreasing sensibility according to the SW test for the thumb (Jonckheere-Terpstra non-parametric trend test, $p = 0.036$)

Conclusions

The sensibility of the thumb and index finger in children with an upper plexus lesion (either surgically or conservatively treated) is diminished. The decreased sensibility has a negative impact on hand function. Appreciation of diminished hand function in patients with NBPP involving C5-C6 is important to optimize treatment.

INTRODUCTION

The neonatal brachial plexus palsy (NBPP) is a traction injury that occurs during birth. The most common type is a lesion of the two upper nerves (C5 and C6) of the brachial plexus. In more severe cases, C7, C8 and T1 can be involved in the lesion as well.[1] Studies have observed that a certain clumsiness of the hand exists in patients with NBPP with a C5-C6 lesion.[2] This is remarkable, as the motor function of the hand is innervated by the lower nerves of the brachial plexus, mostly C8 and T1, which are intact in C5-C6 lesions. One explanation for this phenomenon may be that the diminished hand function is the result of a reduced sensibility in the hand. The sensory innervation area of spinal nerve C6 consists of the lateral ventral lower arm, the radial side of the palm of the hand, and the palmar side of the thumb and index finger. Sensibility in these areas has been found to be reduced in adults with NBPP.[3] Reduced sensory feedback of these two fingers to the brain may have a negative influence on dexterity. Reports on sensibility of the hand in patients with NBPP are limited, and conflicting.[4] Some papers reported normal sensibility, while others reported diminished sensibility.[5, 6, 7]

The current study was performed to assess sensibility in young children with NBPP, with two goals: (1) to see whether we could detect a decreased sensibility and (2) to correlate sensibility with hand function in children with NBPP involving a C5-C6 lesion. We investigated a cohort of children with NBPP that included both conservatively treated children and children treated with nerve surgery in early infancy, and compared their results with those of controls.

PATIENT AND METHODS

The study design was a cross-sectional investigation of patients with NBPP. Fifty children with an upper NBPP and 25 healthy children, aged between 7 and 12 years, were recruited for the study. The children with NBPP were patients who were examined during regular follow-up at our tertiary referral clinic (Nerve Centre of the Leiden University Medical Center, The Netherlands). We diagnosed the children originally with NBPP on obstetrical history, physical examination and additionally with EMG when first presentation was between the age of 4 and 6 weeks.

In 30 children (60%), nerve surgery had been performed in early infancy, while 20 had been treated conservatively. In all children, the clinical diagnosis of NBPP was made with involvement of only the C5 and C6 roots. Children who were conservatively treated, showed recovery of elbow flexion with active biceps muscle at 3 to-6 months of age, all had active elbow extension with active triceps muscle and active wrist extension at least against resistance. All children had normal hand function, as judged by neurological examination. The indication for nerve surgery has been extensively described elsewhere.[1] Children who were treated with nerve surgery, underwent CT myelography or MRI to assess root avulsion injury. During the operation, surgical inspection and direct nerve stimulation were performed to assess the diagnosis. The most commonly performed surgical procedure to restore C6 function was grafting from C6 to the anterior division of the superior trunk. Five infants had undergone a transfer of the pectoral nerve to the musculocutaneous nerve, in one patient just accessory tot suprascapular nerve transfer had been performed and one patient had undergone neurolysis. In these seven children, the axonal pathway from C6 to the anterior division had been left intact; this sub-group was compared to the surgical sub-group in which nerve reconstruction had taken place.

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 schools.[8]

PHYSICAL EXAMINATION

All assessments in both groups were done by the same physical therapist (SB) to avoid confounders. Before starting with the sensibility tests as described in this paper, we did a try-out of the different test methods on a small group of children, which led to adaptations of the test methodology. The tester had more than 37 years of experience with physical assessment and treatment of children in all age groups.

The sensibility of the hands was assessed using two methods. (1) Two-point discrimination (2PD): the minimal distance between two contact points that children recognized as two separate stimuli was investigated using the Mc Kinnon-Dellon Disk-criminator®. We tested the palmar side of the thumb, the index finger and the little finger. (2) The Semmes-Weinstein Monofilament test (SW): nylon filaments of different diameters (D 2.83 mm; F

3.61 mm; J 4.31 mm; K 4.56 mm) were tested, applied twice to each fingertip. The test started with the thinnest filament. For each finger, we noted the number of stimuli with the thinnest filament that were felt, after which thicker filaments were not applied anymore. During both tests, the sensory stimuli were applied to the volar side of the fingertip, while a screen prevented the children from seeing their own hand. For children who had difficulty to maintain presentation of the palmar surface as a result of fatigue or supination weakness, the tester adjusted her position to ensure that the filaments were put perpendicular to the fingertips.
Dexterity was evaluated with a single item from the Movement Assessment Battery for Children-2 (MABC-2), an internationally accepted and validated test for fine motor skills.[9] For children aged 7, 8, 9 or 10, the specific task to perform was to thread a wire through holes in a board. Children aged 11 or 12 years were asked to make a triangle with bolts and nuts. As these specific tasks are bimanual, both the dominant and the assisting hand are tested. Children were not allowed to put either the wire or the triangle on the table, but were required to keep them in both hands. The time they needed to finish the task was noted and converted to a standard score. This score is known to be age-dependent.[9]

In many children with NBPP, the unaffected hand serves as the dominant hand, as hand dominance may have shifted as a result of their lesion. To exclude a potential confounding effect of hand dominance on sensation, we analyzed children whose dominant hand was the unaffected side. We compared the affected side of the NBPP group with the non-dominant hand of the control group. We defined the dominant hand as the hand in which a child holds a pencil to write. A hand preference shift was assumed to have occurred if a child with a right-sided lesion had left-hand dominance.

The study protocol was approved by the Medical Ethics Committee of the Leiden University Medical Center (ABR number 48977) and informed consent was given by the parents.

STATISTICAL ANALYSIS

For continuous outcome variables, we used analysis of variance (one-way ANOVA). 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 Jonckheere-Terpstra test – a rank-based non-parametric trend test similar to the Kruskal-Wallis test – was applied where indicated.[10] The error level was set at $p < 0.05$. Data were analyzed with SPSS Statistics for Windows, version 22 (IBM Corp.Armonk, NY).

RESULTS

Patient details are presented in Table 1.

Table 1 Patient characteristics

Demographic variables	NBPP	Controls
Total number	50	25
Nerve surgery	30	N/A
Conservatively treated	20	N/A
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	N/A

Legend Table 1
SD: standard deviation.

The analysis of the affected non-dominant hand concerned 28/30 surgically treated children and 14/20 conservatively treated children. (Table 2) In the surgically treated group, an assumed hand preference shift was found in 13/15 children, compared to 3/9 children in the conservative group. This difference is statistically significant ($p = 0.007$).

Table 2 Hand dominance versus affected side

				Dominant hand	
Group				**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

Controls: n = 25; NBPP: infants with an NBPP lesion (n = 50), 20 of whom had been treated conservatively and 30 had undergone nerve surgery. A hand preference shift was assumed to have occurred if a child with a right-sided lesion had left-hand dominance.

* Not included in the analysis; # assumed preference shift.

Sensibility with the 2PD

The mean 2PD for the thumb in the control group was 2.12 mm, compared with 2.69 mm in the NBPP group. Similar differences were found for the index finger. The analysis of the mean 2PD showed significant differences in both the thumb and index finger between the affected hand in children with NBPP and the non-dominant hand in the control group. No statistically significant differences were found when comparing the little finger (Table 3 – top). When we compared the affected side with the unaffected side in all infants with NBPP, statistical differences were found for the thumb and index finger, but not for the little finger (Table 3 – bottom). A comparison of the affected and unaffected sides in the NBPP group showed no differences between nerve surgery and conservative therapy. There was no statistical difference within the surgery group, comparing nerve reconstruction with nerve transfer or neurolysis.

Table 3 Mean 2PD in dig 1,2,5

Group	Dig 1 (SD)	Dig 2 (SD)	Dig 5 (SD)
Controls (n = 25)	2.12 (0.33)	2.08 (0.28)	2.48 (0.59)
NBPP (n = 42)	2.69 (0.95)	2.45 (0.71)	2.69 (0.68)
Conservative (n = 14)	2.64 (1.22)	2.43 (0.65)	2.93 (0.83)
Nerve surgery (n = 28)	2.71 (0.81)	2.46 (0.74)	2.57 (0.57)
NBPP – affected side (n = 50)	2.70 (0.88)	2.42 (0.67)	2.70 (0.68)
NBPP – unaffected side (n = 50)	2.28 (0.50)	2.16 (0.37)	2.64 (0.66)

Legend Table 3

The mean 2PD is shown in mm, including Standard Deviation (SD).

Top part of table:

Comparison of the non-dominant side in controls (n = 25) versus affected side in NBPP (n = 42). Differences were calculated as follows:

Digit 1: control vs NBPP p = 0.005; control vs conservative p = 0.051, control vs surgery p = 0.008, conservative vs surgery p = 0.782.

Digit 2: control vs NBPP p = 0.014; control vs conservative p = 0.081; control vs surgery p = 0.021; conservative vs surgery p = 0.854.

Digit 5: No statistically significant differences.

Bottom part of table:

Comparison of the affected and unaffected sides in NBPP. Affected versus unaffected, paired T test: Digit 1: p = 0.001; Digit 2: p = 0.004; Digit 5: p = 0.537.

Sensibility with SW

In the control group, 92% of the children were able to feel the thinnest SW filament (D) on their thumb, compared to 43% in the NBPP group (Table 4 – top). This difference was statistically significant whether we compared the controls with either the total NBPP group (p<0.001), the conservatively treated group (p<0.001) or the surgically treated group (p = 0.001). There was no difference between the surgically treated group and the conservatively treated group (p = 0.270 chi-square test). We found similar results for the index finger (data not shown), whereas for digits 3, 4 and 5 we found no statistically significant difference in sensibility. A comparison of the affected and unaffected hands showed a statistically significant difference in the proportion of patients who felt the D filament: 22 versus 42 children (p<0.001, McNemar Test), for both the thumb (Table 4 – bottom) and the index finger (data not shown). There were no statistical differences for digits 3, 4 and 5. There were no statistical differences for the surgical subgroups.

Table 4 Thinnest SW filament that was felt in the thumb.

	Monofilament			
	D	**F**	**J**	**K**
Group	**n (%)**	**n (%)**	**n (%)**	**n (%)**
Controls (n = 25)	23 (92)	2 (8)	0 (0)	0 (0)
NBPP (n = 42)	18 (43)	18 (43)	5 (12)	1 (2)
Conservative (n = 14)	5 (36)	5 (36)	3 (21)	1 (7)
Nerve surgery (n = 28)	13 (46)	13 (46)	2 (7)	0 (0)
NBPP – affected side (n = 50)	22 (44)	22 (44)	5 (10)	1 (2)
NBPP – unaffected side (n = 50)	42 (84)	7 (14)	1 (2)	0 (0)

Legend Table 4

Top part of table:

Comparison of the non-dominant unaffected side in controls versus NBPP. The thinnest filament is D (2.83 mm), with increasing thickness in the F (3.61 mm), J (4.31 mm) and K (4.56 mm) filaments. Statistically significant differences were found for controls (n = 25) vs NBPP (n = 42) p<0.001; controls (n = 25) vs conservative (n = 14) p <0.001; controls (n = 25) vs surgery (n = 28) p = 0.001. Difference between conservative (n = 14) and surgery (n = 28) was not significant: p = 0.270 (chi-square test)

Bottom part of table:

Comparison of the affected (n = 50) versus unaffected (n = 50) side in NBPP. Difference comparing pairs of those who felt SW filament D and those who did not: p<0.001 (McNemar Test).

Hand function according to Movement ABC-2

The children with an upper NBPP had decreased hand function compared with the control group, expressed as the test score (corrected for age) for the single item of the MABC-2 we used (p = 0.023, Mann-Whitney test). The median score in the control group was 11.0 (corresponding to a time of 20-21 seconds in a 9-year-old) and that in the NBPP group was 8.0 (26 seconds for the same age). There was a significant difference toward decreasing hand function with increasing SW filament diameter for the thumb (p = 0.036). (Figure 1) There was no significant difference for SW digit 2 (p = 3.33) or for 2PD (p = 0.829).

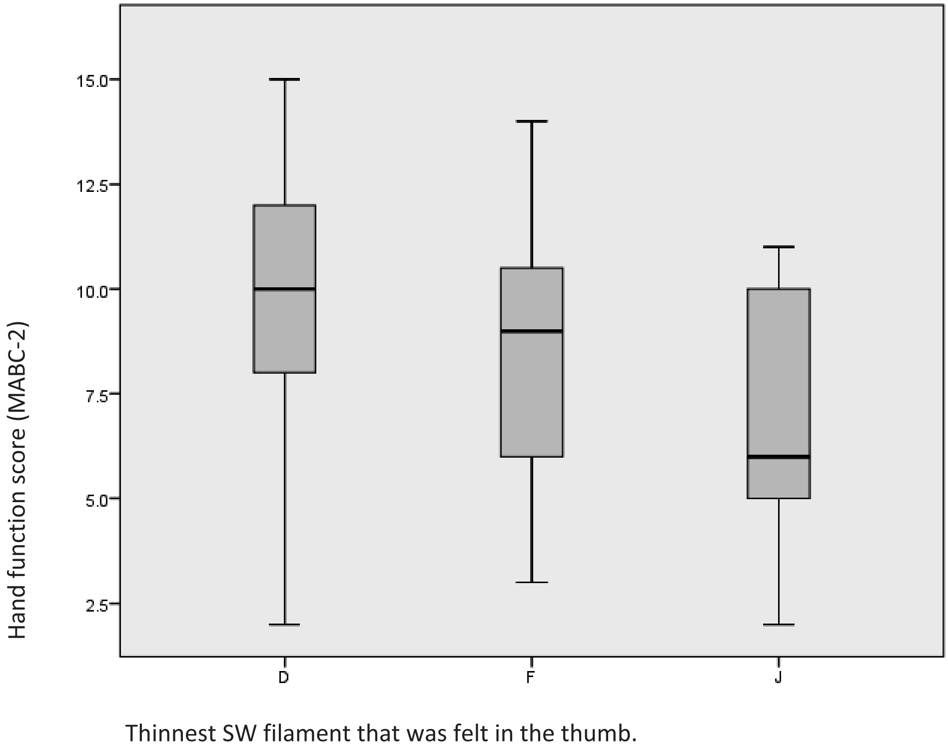

Figure 1 Correlation between SW digit 1 and hand function

Legend Figure 1

Hand function score: standardized value derived from the time it took to complete a single item from the MABC-2. D, F, J: Nylon filaments of different diameters: D = 2.83 mm.; F = 3.61 mm.; J = 4.31 mm. Plot shows median value, 25-75% range (box), 95% range. There was a significant difference toward decreasing hand function with increasing SW filament diameter for the thumb (Jonckheere-Terpstra non-parametric trend test: p = 0.036)

DISCUSSION

In the present study, we found reduced sensibility of the thumb and the index finger in a cohort of 50 children with NBPP with an upper brachial plexus lesion, who had been treated either conservatively or surgically. Scores for the thumb on both the 2PD and SW tests were found to be reduced when comparing infants with NBPP with healthy controls, and also when comparing the affected and unaffected sides in infants with NBPP. Hand function also appeared to be diminished in the NBPP group, based on a single item of the Movement

ABC-2. Together, these findings suggest that decreased sensibility may have a negative impact on hand function.
Our findings contradict previous reports stating that sensory recovery in children with NBPP is generally good,[6, 7, 5] and support others which also found a diminished sensibility.[11, 12, 2] These conflicting data may have resulted from differences in sensory testing methods. Our results support previous results from our group showing that sensibility was affected in adults whose NBPP was conservatively treated.[3]

Decreased hand sensation may contribute to decreased hand function, as normal sensory input to the somatosensory cortex in early life is essential for the development of motor skills.[2, 13, 14] Additionally, the threshold for afferent input to the sensory cortex depends on stored memory and experience, which may be altered in children with NBPP. Another factor that might be related to the clumsiness of the hand may be a reduced proprioceptive sensation. This has been found in the elbow joint,[12] but has not been studied for the hand. Diminished sensibility in one hand may require additional visual control during two-handed activities. Motor control of the thumb and index finger already requires more visual guidance in complex tasks, which may become tiring.[15] The need for additional visual control to properly perform a task should be explained to the caretakers of children with NBPP, and incorporated in physical therapy.

An alternative explanation is that the development of hand function in NBPP infants is impaired due to the diminished range of motion of their shoulder and elbow. Many children are particularly limited in external rotation and supination.[16] This suboptimal positioning of the hand may lead to decreased development of fine motor skills. During the test protocol we choose the described items of the MABC (thread a wire / compose a triangle), as these tests concern a specific bimanual test for which the non-affected hand can adapt to a suboptimal position of the affected hand. Alternative options (e.g. a pegboard task) was considered, but this test requires normal active external rotation, which is usually limited in an upper brachial plexus lesion.
Another factor that influences assessment of motor function is the normal neurological development. In this cohort of children, we did not formally assess possible developmental delays, but in a previous paper we found that developmental delays, including cerebral

palsy, developmental coordination disorder , mental retardation, but also behavioral problems, such as attention deficit hyperactivity disorder, may be present more often in children with NBPP than in the normal population.[17]

Finally, the observed clumsiness could potentially be explained by the fact that lesions that are supposedly limited to the upper trunk may involve subclinical damage to the middle and lower trunks as well. In fact, some authors found electromyographic (EMG) abnormalities in hand muscles of children with normal neurological examination of the hand.[14] We did not examine the intrinsic hand muscles of our children with EMG, and can therefore not exclude this possibility. However, our findings in the sensory examination do not seem to support additional damage apart from C5 and C6. The results of the 2PD test showed a significant difference with healthy controls for the thumb and index finger, but not for the little finger. The same pattern was found in the SW test: there were differences for the first and second fingers, but not for the third to fifth fingers. These findings confirm the anatomical distribution of dermatomes for the thumb and index finger, which are innervated by C6, as well as the involvement of C6 in upper trunk NBPP. The third to fifth fingers were not affected in our patients with NBPP, indicating only C5 and C6 involvement.

A strong point of our study is that we compared the sensibility of infants with NBPP with the sensibility of healthy controls, which has not been done previously. We were therefore able to make a direct correlation with hand function. In order to facilitate the interpretation of our results in the context of those reported in the literature, we also performed a paired comparison of the affected and unaffected sides, as is usually done.

We compared the affected NBPP hand with the non-dominant hand of controls. The reason for this is our assumption that sensation in the unaffected side of the children with NBPP may be better than normal, as a result of more intensive use. The use of the unaffected side as a control may therefore yield differences which cannot be explained only by reduced sensation in the affected hand. A study in our group of healthy children found no difference in sensation between the dominant and non-dominant hands.[8]

A limitation of our study is that no validated and generally accepted measurement tool for sensibility of the hand is available for children. We used existing measurement tools to develop a method that is applicable in children, by adapting this set of methods or tools to the intended population, and making them suitable for use in practice, in view of the smaller size of children's fingers, their level of understanding and their concentration span. These modifications were tested in a pilot study among healthy children, which led to further adaptations. During the pilot, we found that children below the age of 7 years could not manage sufficient concentration to complete longer sensibility tests. We hope that our protocol may be of value to others in developing a universally accepted way to examine sensibility in children.

In the present analysis, we assessed the non-dominant hand of the control group. In many patients with NBPP, the dominant hand is the unaffected side. A previous paper showed that only 17% of children with a right-sided NBPP were reported to be right-handed.[18] In the general population, it is expected that 90% of children has a right hand preference, so it was hypothesized that in the majority of children with a right sided lesion their hand preference had shifted to the left side. In the present, albeit smaller, series, we found that the left hand was dominant in two-thirds of the patients with right-sided lesions, and hand dominance may have shifted in many of these children. We found a difference between conservatively treated children and those who had undergone nerve surgery. The surgically treated children showed a 'hand preference shift' in 13/15 cases, compared to 3/9 in the conservatively treated group. This finding may reflect the more severe nerve lesions in the surgically treated group as compared to the conservatively treated ones. The groups were too small to compare different surgical strategies, such as proximal reconstruction by nerve grafting and distal nerve transfers. In the latter treatment option, there is no reconstruction of sensory axons. The current study cannot answer the question which of the two is preferred from the perspective of sensibility.

CONCLUSIONS

In conclusion, we found that diminished sensibility in the thumb and index finger correlates with diminished hand function. These findings should contribute to a better understanding and appreciation of the observed clumsiness of the hand in patients with NBPP upper trunk lesions. Giving specific attention to hand function when treating these children might reduce the deleterious effects of diminished sensation. Finally, our test protocol may serve as a step toward a validated, universally accepted test protocol for sensibility in infants with NBPP and other peripheral nerve lesions.

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