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

Buitenhuis, S.M.

Chapter 5

Children with an upper Neonatal Brachial Plexus Palsy: Awareness of Diminished Sensibility and Pain in the Hand

Sonja M. Buitenhuis, Willem Pondaag, Ron Wolterbeek, Martijn J.A. Malessy

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Abstract

Objective

In previous research, we found that objective outcomes such as two-point discrimination, Semmes- Weinstein monofilament testing, and localization ability of the thumb and index finger were diminished in children with a C5-C6 Neonatal Brachial Plexus Palsy (NBPP). We performed the present study to investigate whether these children report a diminished subjective sensation and/or pain in their hand, and to assess whether their parents are aware of these features.

Patients and Methods

During regular outpatient clinic visit 45 children (mean age 10 years) and their parents answered two simple questions concerning altered sensation and pain in the affected hand. The answers of the children were compared with those of the parents. We additionally analyzed the answers of the children in relation to two-point discrimination and Semmes-Weinstein monofilament tests.

Results

We found that 13/45 children (29%) reported a different sensation in the affected hand as compared to the unaffected side. Nine parents of these 13 children (64%) were not aware of the diminished sensation. Pain in the hand was reported by 14/45 children (31%), while the parents of only one child (9%) were aware of the pain. There was no correlation between the responses to the questions and the results of two-point discrimination and Semmes Weinstein testing.

Conclusion

Children with a C5-C6 NBPP have a diminished sensibility of their affected hand, but they are often not aware of this reduced sensation. Altered sensation and pain in the hand are often overlooked aspects of NBPP, but form relevant features.

INTRODUCTION

The neonatal brachial plexus palsy (NBPP) is a nerve traction injury that occurs during birth. Most commonly, the spinal nerves C5 and C6 are affected. In more severe cases, C7, C8 and T1 are involved as well.[1]

Previously, we reported results from an in-depth study of sensation of the hand in a cohort of fifty children with a C5 and C6 NBPP. Both conservatively and nerve surgically treated children were included. We found that two-point discrimination, Semmes- Weinstein monofilament testing, and localization ability of the thumb and index finger were diminished. The impeded sensibility had a negative impact on hand function.[2, 3] Additionally, we showed that both gripforce and dexterity of the affected hand of infants with C5 and C6 NBPP were reduced.[4] The literature on sensibility of the hand after NBPP is conflicting: some papers report findings in line with our previous findings[5, 6], while other papers have reported that sensory recovery after NBPP was good.[7-10]

The subjective experience of touch has been assessed only very rarely. From a cohort of 65 surgically treated children from The Hospital of Sick Children in Toronto, seventy percent described, when asked, that the affected limb "felt different", every day or at least once a week.[11] This was surprising, as during regular follow-up in our outpatient clinic, neither the children nor their parents spontaneously bring up sensory disturbances. We, therefore, chose to include a short and simple questionnaire in our research protocol to systematically assess subjective sensory disturbances and pain in these children. Additionally, we assessed whether parents were aware of a diminished sensation or pain in the hand of their child. This short questionnaire was delivered to children and their parents, just prior to a detailed analysis of hand sensation.

PATIENTS AND METHODS

During their regular follow-up visit at our tertiary referral clinic (Nerve Centre of the Leiden University Medical Center, The Netherlands), fifty children with C5, C6 NBPP between 7 and 12 years of age were included. In 30 children nerve surgery had been performed in early infancy, while 20 had been treated conservatively.

A simple questionnaire on paper was handed out, one to the child and one to the parents. (Table 1).

Table 1 Questionnaire (English translation)

For	Question
Children	1) Do you feel a difference in sensation in your affected hand when you compare it to the unaffected side?
	2) Do you feel pain in your affected hand?
Parents	1) Does your child sometimes indicate that feeling in the affected hand is different from the unaffected side?
	2) Does your child sometimes indicate that he/she has pain in the affected hand?

Both the children and their parents had to answer the questions separately by simply choosing 'yes' or no'' to the questions posed. Occasionally, the child was helped to read the questionnaire, but never helped while giving the answer. The researcher was in the same room to prevent the parents to help or influence the answers of their child. Analysis of the questionnaire was performed only when all questions were completed by both the children and their parents.

After answering the questions, the sensibility of the hand was analyzed in detail according to our research protocol which we previously described in detail. Briefly, two methods were used namely 1) two-point discrimination (2PD) and 2) the Semmes-Weinstein (SW) monofilament test.[2]

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

We used Pearson's or Spearman's correlation coefficients, depending on the type of variable, Pearson chi-square test and the Mc Nemar-test. The error level was set at $p < 0,05$. Data were analyzed with SPSS Statistics for Windows, version 28 (IBM Corp. Armonk, NY).

RESULTS

The questions were completed 45 times out of 50 by both children and parents. The mean age of the children was 10 years (SD 1.83).

Thirteen of 45 children (29%) reported that sensation in the affected hand differed from the unaffected side, 8 of whom had been treated conservatively, 5 were operated. The

remaining 32 children (71%) did not experience a different sensibility (11 conservatively, 21 surgically treated).

Four parents of the 13 children who reported a difference in hand sensation indicated that their child had mentioned it. The remaining 9 parents answered that they were unaware of any sensation differences. Of the 32 children who did not report a different sensibility, 2 of the parents stated that their child had mentioned a difference in sensation (see. Table 2).

Table 2. Sensibility of the hand: comparison of answers to the questions regarding sensation in the hand provided by both the children with a C5, C6 neonatal brachial plexus injury and their parents (n = 45)

		Different according to children		Total
	Answers	**Yes**	**No**	
Different according to parents	**Yes**	4	2	6
	No	9	30	39
Total		13*	32**	45

Table 2 Legend

* 8 conservative, 5 nerve surgery

** 11 conservative, 21 nerve surgery

The sensibility of the hand as analyzed with two-point discrimination (2PD) and Semmes Weinstein (SW) monofilaments was compared with the unaffected side.[2] The differences of the 2PD were statistically significant for the thumb (paired t-test $p = 0.001$) and index ($p = 0.004$). The thinnest SW filament was often not recognized in the affected thumb and index finger ($p < 0.001$; McNemar Test).[2]

We analyzed the relationship between the answers of the children to the questionnaire with the results of the 2PD for thumb and index. The Pearson correlation coefficient was -0.11 ($p = 0,48$) and -0.14 ($p = 0.36$) respectively. Additionally, we analyzed the relationship of the responses to the questionnaire with SW testing, which resulted in a Spearman correlation of 0.01 for the thumb ($p = 0.93$) and -0.09 for the index ($p = 0.56$), see Table 3. We repeated these analyses in two age groups: children younger than 10 years ($n = 23$) and older than 10 years ($n = 22$). The results were similar: no correlations were found (data not shown).

There was no correlation between subjective sensation and whether primary nerve surgery had been performed or conservative therapy was provided (Pearson Chi-Square Test: 0.037, p = 0.848).

Table 3 Correlation coefficients between the answers from the children regarding the questions about differences in sensation in the affected and unaffected hand (yes/no) and the sensibility tests 2PD, SW

	2PD		SW	
	Dig 1	**Dig 2**	**Dig 1**	**Dig 2**
Statistical test	Pearson	Pearson	Spearman	Spearman
Correlation coefficient	-0.11	-0,14	0.01	-0.09
p	0.48	0.36	0.93	0.56

Table 3 Legend

2PD: Two-point discrimination test; SW :Semmens Weinstein test.
Pearson: Pearson Chi-Square Test; Spearman: Spearman correlation

The question about pain was completed by both children and parents 45 out 50 times. The mean age of these children was 9.9 years (SD 1.86).

Thirty-one of the 45 children (69%) reported that they did not experience pain (12 conservatively, 19 surgically treated). The parents of these children consistently reported that their child had no pain. The remaining 14 children (31%) reported that they experienced pain in the affected hand (5 were conservatively treated, 9 surgically were treated). Only one parent of these 14 children was aware of the pain of their child (see Table 4).

Table 4 Answers to the questions about pain in the hand of both the children with a C5, C6 neonatal brachial plexus injury and their parents (n = 45)

		Pain according to children		**Total**
		Yes	**No**	
Pain according to parents	**Yes**	1	0	1
	No	13	31	44
Total		14*	31**	45

Table 4 Legend

* 5 conservative, 9 nerve surgery
** 12 conservative, 19 nerve surgery

There was no correlation between the presence / absence of pain and whether primary surgery had been performed (Pearson Chi-Square Test: 2.461, p = 0.117). We tested the correlation between the presence or absence of pain and the outcome of sensory testing, in the same way as we analyzed subjective sensory differences described above, and found no correlations (data not shown).

DISCUSSION

In a previous study we showed that children with a C5, C6 NBPP lesion have a decreased sensibility in the thumb and index finger, which was assessed with 2PD and SW filaments testing. In the current paper, we analyzed whether children and their parents were aware of the impaired sensation. We found that 13/45 children (29%) of children with an NBPP expressed, when asked, that they had a different sensation in the affected hand as compared to the unaffected side. The vast majority of the parents was not aware of these differences. As regard to pain, 14/45 (31%)of the infants mentioned that they experienced pain in the hand, while the parents of only one child (9%) were aware of the pain.

The major finding of this study is that we did not find a correlation between the responses of the children to the questions and the outcome of two-point discrimination and Semmes Weinstein testing (Table 3), regardless of their age group. While the objective testing results indicated that the children had a different sensation in the affected hand as compared to the un-affected hand, less than one third also indicated to experience a different sensation. From these results we conclude that in this series the majority of children with an upper NBPP have a diminished sensibility of their affected hand, but that these children are not aware of this reduced sensation.

We hypothesize that this lack of awareness results from the early lack of sensory input to the brain resulting in habituation: they simply don't know otherwise. Only occasionally, children spontaneously mentioned difficulties they experienced in daily life which we could link to a reduced sensibility of the affected hand, whereas the children themselves did not connect their limitations to the underlying sensory loss. To provide insight into the significance of our findings for daily practice, we give some examples. One child mentioned difficulties to switch gears of his race bike which were to diminished sensitivity of his thumb. Another child described how she tested whether a pan is hot or not. She touched the pan with the thumb and index finger of the affected side, because as it would be hot, it would be too painful to

touch it with the non-affected hand. Lastly, we found inflammation of the cuticle due to biting the thumb and index finger of the affected side, which could be tolerated only because of a diminished pain sensation. Although the sensation loss in children with upper NBPP is not complete, it may still effects daily life activities.

Another aspect that needs to be addressed is the current lack of awareness amongst treating occupational or physical therapists of any sensibility disturbances in the hand of children with NBPP. As sensibility provides input for the brain to execute fine motor tasks and complex hand function, these disturbances should be taken into account while developing and executing training programs. Previously, we showed that children with an upper NBPP do not only have an impaired sensibility, but a reduction of gripforce[4] and dexterity[2] underlying a diminished hand function.

Parents were often surprised to learn that the sensory testing of their child showed a diminished sensibility in the affected hand. They apparently assumed that the sensibility was not affected because their child never mentioned it spontaneously.

Besides the subjective awareness of diminished sensibility, we posed a question on pain. In the present series around 30% of the children reported to have pain in the hand of the affected arm. This percentage is much lower as compared to the 70% previously reported by the Toronto group.[11] The reason for this discrepancy might be that in the current series only patients with NBPP lesions limited to the C5, C6 spinal nerves were studied, whereas more extended lesions affecting the C7, C8 and /or T1 roots were included in the Toronto series as well. In addition, the Toronto series comprised only children who were operated, while in the present series also conservative treated children were included. Another difference was the way the pain was assessed, namely with the Faces Pain scale and / or the Adolescent Pediatric Pain Tool and not a dichotomous questionnaire.

Another paper reported pain prevalence around 78% (29 children out of 37). The pain had been spread out throughout the whole arm, irrespective of severity and was episodic in nature. The type of pain was described as musculoskeletal and nociceptive.[12]

Surprisingly, only one of the parents was aware of the pain in the hand of their child. The parents of those children who mentioned to have pain supposed that their child mistakenly answered the questions the wrong way around, because they had never heard them complain. Another factor that might also have played a role is a difference in phrasing or

interpretation of pain. Some children, for instance, said: “It does not feel nice when you touch the top of my finger”. Children may not only experience pain in a different way, but they may also use other words to describe pain.[11] For children, pain can be expressed as ‘pain’, but also as an ‘unpleasant feeling’.

The questions were given to the parents and children separately before starting the sensibility testing. We did not find a correlation between the answers of the children and the sensibility tests, signifying that most children were not aware of their sensory deficit. Regularly during testing, both the child and the parents were surprised when the sensation appeared to be diminished. The following striking reaction of a child was noted: “Now it became clear to me why, during typing or playing the piano, I always need to visually control my affected hand”.

The number of reports dealing with sensibility of the hand in children with NBPP is limited, and conclusions are conflicting.[13] Some papers reported normal sensory findings[7-10], whereas others reported that sensibility was affected.[5, 6] These conflicting data may be the result of different methods how sensation was tested and difference in the interpretation of the results.[14] Anand and Birch, for example, tested the sensibility with six sensory modalities: monofilaments, cotton wool, pinprick, warm/cool sensation, joint position sense and vibration. Their definition of ‘excellent restoration’ of sensory function was recovery to normal limits in all dermatomes ‘for at least one modality’. In their series only 6 / 20 operated patients recovered to normal SW testing results, so we feel that the often-cited excellent recovery in their paper may be partly due to their optimistic definition of good recovery.[7]

The strength of the present study is that we posed our questions prior to examining hand sensation. A second strength is that we also asked the parents. A weakness of the study is that we did not assess whether the children are so used to the impaired sensory input that they, therefore, do not mention it. Additionally, we did not assess whether parents may have misinterpreted complaints that were actually expressed by their child or even have unconsciously denied them. It should thereby be considered that an important part of the daily focus of the parents in relation to the NBPP is whether tasks can be executed which involve motor function and sensory perception might thereby be pushed to the background. Another drawback of this study is that the questions we used were not validated. Unfortunately, a validated questionnaire was not available. The youngest children in our

cohort were 7 years old. Especially the young ones asked support from their parents to help them understand the questions. Although we strived to minimize such interaction, we cannot completely exclude whether parents exerted some influence on how their child answered the questions. We feel, however, that this cannot have been to such an extent that this may have affected the outcome of the study.

CONCLUSION

Children with a C5, C6 NBPP lesion have sensory loss in the hand, next to shoulder and elbow motor function limitations. Both children and parents are frequently not aware of the sensory impairment of the hand. One third of the children expressed pain in the hand, which was hardly recognized by their parents. Both loss of sensation in the hand and pain in children with C5, C6 NBPP are factors that deserve attention, not only of the parents, but also of therapists to optimize treatment.

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