Unravelling cossed wires : dysfunction in obstetric brachial plexus lesions in the light of intertwined effects of the peripheral and central nervous system

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Issue Date: 2018-06-26

Chapter 7

Impaired automatic arm movements in obstetric brachial plexus palsy suggest a central disorder

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J Child Neurol. 2016 Mar, 31(8):1005-8.

Abstract

We aimed to find evidence for a central component of the impairment of movement of the affected arm in children with obstetric brachial plexus palsy. We performed a cross-sectional study in 19 children (median age 5 years) with obstetric brachial plexus palsy who were able to voluntarily abduct their affected arm beyond 90 degrees. They were asked to perform four tasks designed to provoke automatic arm movements to maintain balance. We assumed automatic motor programming to be impaired when two of three investigators agreed using video recordings that the affected arm did not abduct beyond 90 degrees while the unaffected arm did. Children abducted the affected arm less often than the healthy one (generalized binary logistic model of all four tasks, p=0.001). The deficit during automatic arm abduction was not observed during voluntary movements and therefore cannot be explained by a peripheral deficit, suggesting a central component.

Introduction

Obstetric brachial plexus palsy is a closed traction injury of the brachial plexus during birth, with an incidence of 0.5 to 2.6 per 1000 live births.[1] A permanent deficit in arm function affects 20 to 30% of cases.[2] Functional recovery following a nerve lesion depends on the number of outgrowing axons that successfully cross the lesion site and on their correct routing.[3-5] Theoretically, recovery of obstetric brachial plexus palsy may additionally be impaired by a disturbed development of central motor programs.

There is neurophysiological evidence supporting defective motor programming in obstetric brachial plexus palsy,[6] and the concept of impaired central motor programs in obstetric brachial plexus palsy is also supported by observations of obstetric brachial plexus palsy infants 'forgetting their arm' during automatic movements: children with obstetric brachial plexus palsy may flex the elbow on the affected side while voluntarily picking up a ball, but the same elbow may not flex during running or other tasks that rely on automatic movements, while the unaffected arm does flex at that time.[3,7] If the observed deficit in the affected arm was wholly due to peripheral nerve, muscle or joint damage, the deficit would not depend on whether a movement is made in a voluntary or an automatic context. The movements of the unaffected arm serve as a control that the task indeed demanded flexion. Accordingly, we reason that arm movements in obstetric brachial plexus palsy that can be performed voluntarily by both arms, but that do not occur in the context of automatic movements of the affected arm, suggest the presence of a central deficit. In other words, we regard the discrepancy between volitional and automatic movements as evidence for a central component. Whether volitional or automatic movements are performed worse does not in fact matter for this reasoning; clinical observation suggested that automatic movements happen to be most impaired.

Motor tasks become consolidated in central motor programs with repetition and practice.[8] Tasks that are highly practiced to the point of demanding few attentional resources are called automatic tasks.[9] Anticipatory postural adjustments of the arms during walking are in part automatic movements.[10,11] To suppress volitional influences that interfere with the automatic component in these arm movements, attention can be diverted by dual motor or cognitive tasks.

The aim of this study was to elucidate whether automatic movements are indeed impaired, suggesting that incomplete recovery is at least partially central in origin.

Methods

Participants

Twenty-three children between three and eight years of age with an obstetric brachial plexus palsy were investigated at the Nerve Centre of the Leiden University Medical Centre between August 17, 2010 and September 21, 2010. Data concerning lesion severity and any surgical intervention were taken from patient records. Parents and children provided verbal informed consent to participate after detailed information was provided. A Mallet grade four for shoulder abduction of the affected side was required for inclusion, equivalent to abduction of at least 90 degrees.[12] Any other relevant disorder affecting movement or sensation served as reason for exclusion. The study was judged by the institutional medical ethics committee to be innocuous and to not warrant a full review, conforming to Dutch law.

Procedure

The tasks were video recorded. Children were asked to perform four tasks while walking approximately 3 meters on a straight line. We searched for tasks that would lie as far as possible on the 'automatic' side of a scale from 'fully automatic' to 'fully volitional'. We used balance tasks to provoke automatic arm movements, performed to prevent falling. Automatic tasks themselves demand few attentional resources; we added dual motor or cognitive tasks of increasing difficulty to the balance tasks to make the children focus on those tasks, thereby shifting their attention away from volitional control over their arm movements. The children and parents were informed that the investigation was aimed at central motor programming, but our focus on automatic arm movements was not disclosed to avoid voluntary interference. Each task was first demonstrated by one of the authors (GVA): (a) Walking heel-to-toe towards the camera; (b) Walking on the heels with small steps; (c) Walking heel-to-toe with eyes closed; (d) The same as task (c) but with a cognitive task: count out loud or count backwards generally starting from the age of four years or to name five girl or boy names if younger or counting was too difficult. Children were reminded to perform the task until the end of the line was reached. The investigation was stopped when children did not wish to continue.

Video records were reviewed by three authors (GVA, JGvD, MJAM). Blinding for the side of the affected arm was impossible because affected arms were often shorter and always moved differently from the unaffected side. The assessors independently scored whether either arm was abducted to at least 90 degrees in relation to the position of the trunk for each of the four tasks (Figure 1). Videos were repeatedly viewed if requested. Abduction to at least 90 degrees was considered present when at least two assessors judged so, and absent otherwise. We scored automatic movement as impaired if three conditions were simultaneously met: 1. the affected arm could be abducted on request at least 90 degrees with respect to the trunk; 2. the unaffected arm abducted at least 90 degrees during an automated balance task; 3. the affected arm abducted less than 90 degrees during the same task.

Statistical analysis

IBM SPSS Statistics 20.0 (Armonk, NY: IBM Corp.) was used for statistical analysis. A generalized binary logistic model for repeated measurements with an unstructured correlation matrix including the presence of previous brachial plexus surgery as a variable was used to test whether the rates of abduction differed between affected and unaffected arms over all four tasks. A significance threshold of 0.05 was used. The same model without correction for brachial plexus surgery was applied for the analysis of the tasks separately. A Bonferroni corrected significance threshold of 0.01 (0.05/4) was used.

Results

Four of 23 children did not cooperate and were not investigated. The median age of the 19 participants was 5 years (25^{th}-75^{th} percentile: 4-7 years); there were 12 boys. The left arm was affected in 10 cases. Five (26%) had a C5-C6 lesion, nine (47%) a C5-C7 lesion, three (16%) a C5-C8 lesion and two (11%) a C5-Th1 lesion. Fifteen had undergone surgery of the brachial plexus at a median age of 4 months (25^{th}-75^{th} percentile: 3-7) (Table 1).

The task results are presented in Table 2. Abduction over 90 degrees was present significantly less often for affected than unaffected arms of the healthy arms taking all tasks together (-1.38 (95% confidence interval (95%CI) -2.22,-0.53), p=0.001, Figure 1)). The rates did not differ between participants who had undergone brachial plexus surgery and those treated conservatively (0.49 (95%CI -0.59,1.57), p=0.371). Analysis per task showed that abduction over 90 degrees of the affected arms occurred significantly less often during task (b) (-1.57 (95%CI -2.59,-0.55), p=0.003), but not during the other tasks (task (a), -0.47 (95%CI -1.36,0.43), p=0.309; task (c), -1.12 (95%CI -2.35,0.10), p=0.072; task (d), -1.41 (95%CI -2.72,-0.10), p=0.035).

Discussion

We found that children with obstetric brachial plexus palsy abducted the affected arm over 90 degrees less often than the unaffected arm in automated balance tasks even though they were able to abduct the affected arm over 90 degrees on request. The discrepancy can therefore not be explained by incomplete peripheral nerve regeneration or joint problems. We propose that disturbed central motor programming underlies this phenomenon, at least partially.
Involvement of the basal ganglia, supplementary motor, premotor and motor cortex and the brainstem has been shown in the generation of anticipatory arm movements.[11] At the onset of learning a new motor skill in healthy subjects as well as in patients following upper extremity injury and reconstruction, there is an expansion of motor cortical representation.[13] Once a skill is mastered the degrees of cortical representation and excitability decrease again.[13] A decreased contralateral cortical activation has been found in the primary motor cortex during attempted movement in paraplegics compared to healthy controls studied with motor imagery fMRI, explained by an increased need for attention allocation.[14] Accordingly, a similar pattern of cortical deactivation in obstetric brachial plexus palsy may be the basis of our current findings.

There may be four explanations why automatic movements are impaired in obstetric brachial plexus palsy. The first concerns sensory deprivation: in children with obstetric brachial plexus palsy the connection between the brain and the affected arm is disrupted at birth, leading to muscle weakness

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and diminished sensory feedback.[6] Recovery of the peripheral pathways, if present, takes weeks to many months,[15] during a period when automatic motor programs develop.[6] These programs may remain disrupted even if sensory feedback is repaired afterwards. The relevance of a critical window during motor development in obstetric brachial plexus palsy was previously suggested by Brown et al.[6] They supported their hypothesis by previous observations of poor functional recovery in visually deprived newborn kittens or human infants, or after sciatic nerve crushes in rabbit hind limbs.[6] Obstetric brachial plexus palsy likewise may concerns sensory deprivation during a critical period for the formation of automatic motor control. If so, the effects might be less severe than in the examples provided. It is possible that the degree to which movements become automated also depends on the severity of the lesion. In the present study that severity was limited because recovery had to be sufficient to allow abduction of at least 90 degrees and the central deficit may be explained by the initial afferent deficit. It is possible that a central deficit may play a larger role in obstetric brachial plexus palsy patients with less functional recovery.

The second explanation may be that automatic movement programs are formed later than normal in obstetric brachial plexus palsy, simply because the affected arm is not used often or well enough for movement automation to occur. Corresponding to the 'dual mode principle of motor skill learning', supported by experimental data,[9] tasks can become automatic when they are practiced often enough, resulting in performance that does not require direct full attentional control. If so, automatic movements in obstetric brachial plexus palsy might improve with practice and rehabilitation.

The third explanation holds that the observed decreased use of the affected arm in obstetric brachial plexus palsy during walking represents a compensatory strategy to counter any balance disrupting effects of abnormal arm movements of the affected arm. However, we feel this is unlikely for several reasons: arm swinging is useful as it decreases energy consumption,[16,17] increases stability,[18] and contributes to balance recovery after a perturbation.[19] In cerebral palsy an increased swing of the unaffected arm compensates for the increased angular momentum produced by the legs.[20]

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A fourth possible explanation for decreased automatic arm movement might be that the movements are related to the mass of the arm, which is reduced in obstetric brachial plexus palsy. Again, we feel this is unlikely based on the following: adding mass to one arm has been shown to decrease movement amplitude in that arm and increase the amplitude of movements of the other arm,[20] suggesting that the opposite should hold if the mass of an arm is abnormally low. So, if the low mass of the arm would cause abnormal automatic movements, increased movements would be expected rather than decreased ones. This reasoning implies that the decreased movements impair balance causing further functional impairment.

A potential limitation of this study is that most children had undergone surgery. The lack of a difference in abduction rates between those who had and had not undergone surgery suggests that this factor is not critical. Another limitation is that three tasks appeared not specific enough to evoke abduction in healthy arms in the majority of participants. This may explain the lack of a significant difference for these tasks. The task that showed a clear difference between the affected and unaffected arms consisted of walking on the heels with small steps without additional cognitive tasks. This task may simply represent a more difficult balance act than the other ones. Alternately, our attempts to increase balance difficulty by adding cognitive tasks may not have done so as well as intended: according to the multiple resources theory, the cognitive tasks did not interfere enough with the motor acts because the motor and cognitive tasks share few resources and so cause little interference with one another.[21]

In summary, differences in automatic movements between the affected and unaffected side are present in obstetric brachial plexus palsy. These are likely caused by incomplete central program development and may contribute to incomplete arm function recovery following obstetric brachial plexus palsy.

Acknowledgements

We thank all members of the Leiden University Medical Centre brachial plexus team.

Author contribution

GVA, MJAM, SMB and JGvD substantially contributed to conception or design. GVA, MJAM, SMB, EWvZ and JGvD contributed to acquisition, analysis, or interpretation of data. GVA, MJAM and JGvD drafted the manuscript. All authors were involved in critical revision of the manuscript for important intellectual content.

Declaration of conflicting interests

The Authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Ethical approval

The study was judged by the institutional medical ethics committee Leiden University Medical Centre to be innocuous and to not warrant a full review, conforming to Dutch law.

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Figure 1. Task example
Example of maximal abduction range during task (b), walking on the heels; the right arm is the affected one.

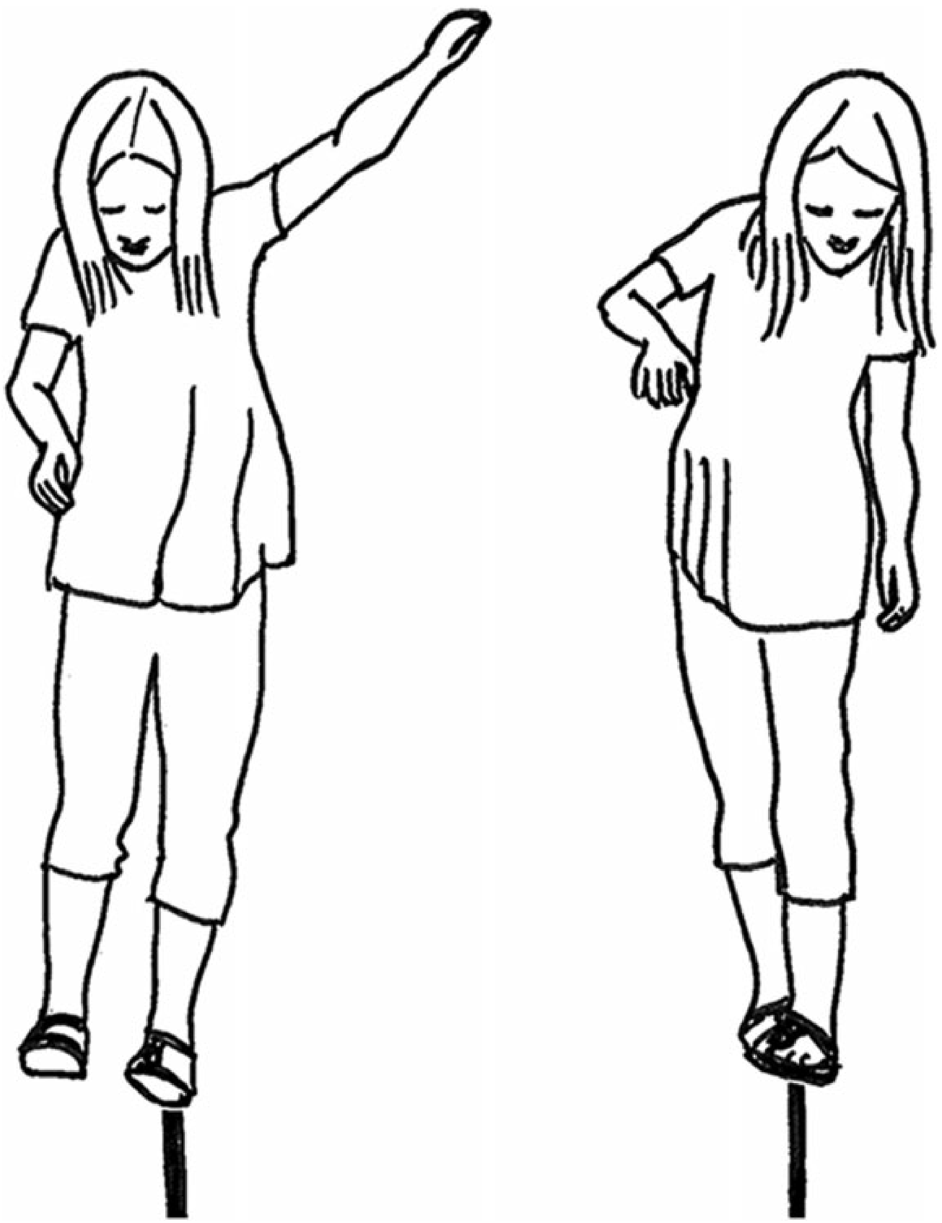

Table 1. Demographic and surgical details

Subject	Age (y)	Gender	Affected arm	Lesion	Treatment
1	5	M	L	C5 – C7	No surgery
2	4	M	R	C5 – C6	Nerve grafting ST
3	4	M	R	C5 – Th1	Transfer accessory nerve - SSN Nerve grafting C5 - PDST, C6 - ADST, anterior filaments C8, Th1
4	5	F	L	C5 – C6	Transfer medial pectoral nerve-musculocutaneous nerve
5	7	F	L	C5 – C7	Nerve grafting C5 - SSN, C5 – PDST, C6 - ADST
6	7	M	L	C5 – C7	Nerve grafting C5 - C5 and intraplexal transfer C5 - ventral filaments
7	7	M	R	C5 – C7	Nerve grafting C5 - SSN, PDST, C6 - PDST, ADST
8	8	F	R	C5 – C6	Nerve grafting C5 - SSN, C5 - PDST, C6 – ADST, neurolysis C7 - MT
9	3	M	L	C5 – Th1	Nerve grafting C5 - PDST, C6 - ADST, C7 - (PD)MT, C8, Th1, accessory nerve - SSN
10	9	F	R	C5 – C7	Nerve grafting C5 - PDST, C5 - ADST Transfer accessory nerve - SSN
11	6	M	L	C5 – C7	Nerve grafting C6 - ADST
12	7	F	R	C5 – C8	Nerve grafting C5 - motor fascicle C7, C6 - ADST, C6 - PDST
13	4	M	R	C5 – C8	Nerve grafting C5 - SSN, C5 - PDST, C6 - ADST
14	3	M	L	C5 – C7	Nerve grafting C5 - SSN, C5 - PDST, C6 - ADST
15	4	M	R	C5 – C6	No surgery
16	3	M	L	C5 – C8	Nerve grafting C5 - SSN, C5/C6 - PDST, C6 - ADST
17	5	M	L	C5 – C7	No surgery
18	4	F	L	C5 – C6	Intraplexal transfer medial pectoral nerve - musculocutaneus nerve
19	8	F	R	C5 – C7	No surgery

y: years, M: male, F: female, R: right, L: left, SSN: suprascapular nerve, ST: superior trunk, ADST: anterior division of the superior trunk, PDST: posterior division of the superior trunk, (PD)MT: (posterior division of the) middle trunk

Table 2. Arm scores for the four tasks
The tasks were: (a) Walking heel-to-toe in a straight line; (b) Walking on the heels with small steps; (c) Walking heel-to-toe with eyes closed; (d) The same as the third task but with a cognitive task, suitable for the child's age. (For the healthy arm numbers may be less than 19 because some children did not perform all tasks) * $p<0.01$

Task (a)		**Arm affected**		
		No	*Yes*	
Abduction >90°	*No*	16	17	33
	Yes	3	2	5
		19	19	38

Task (b)*		**Arm affected**		
		No	*Yes*	
Abduction >90°	*No*	7	14	21
	Yes	12	5	17
		19	19	38

Task (c)		**Arm affected**		
		No	*Yes*	
Abduction >90°	*No*	13	16	29
	Yes	5	2	7
		18	18	36

Task (d)		**Arm affected**		
		No	*Yes*	
Abduction >90°	*No*	11	15	26
	Yes	6	2	8
		17	17	34