

Degree of Contracture Related to Residual Muscle Shoulder Strength in Children with Obstetric Brachial Plexus Lesions

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Abstract

Background and Objectives Little is known about the relation between residual muscle strength and joint contracture formation in neuromuscular disorders. This study aimed to investigate the relation between residual muscle strength and shoulder joint contractures in children with sequelae of obstetric brachial plexus lesion (OBPL). In OBPL a shoulder joint contracture is a frequent finding. We hypothesize that residual internal and external rotator strength and their balance are related to the extent of shoulder joint contracture.

Methods Clinical assessment was performed in 34 children (mean 10.0 years) with unilateral OBPL and Narakas classes I–III. External and internal rotation strengths were measured with the shoulder in neutral position using a handheld dynamometer. Strength on the affected side was given as percentage of the normal side. Contracture was assessed by passive internal and external rotations in degrees (in 0° abduction). Mallet classification was used for active shoulder function.

Results External and internal rotation strengths on the affected side were approximately 50% of the normal side and on average both equally affected: 56% (SD 18%) respectively 51% (SD 27%); $r = 0.600$, $p = 0.000$. Residual strengths were not related to passive internal or external rotation ($p > 0.200$). Internal rotation strength ($r = -0.425$, $p < 0.05$) was related to Narakas class. Mallet score was related to external and internal rotation strengths ($r = 0.451$ and $r = 0.515$, respectively; $p < 0.01$).

Conclusion The intuitive notion that imbalances in residual muscle strength influence contracture formation cannot be confirmed in this study. Our results are of interest for the understanding of contracture formation in OBPL.

Keywords

- ▶ obstetric brachial plexus lesion
- ▶ contracture
- ▶ muscle force/strength
- ▶ shoulder function

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Introduction

An obstetric brachial plexus lesion (OBPL) is a lower motor neuron disorder, resulting from traction to the brachial plexus during labor and resulting in a palsy of the upper limb in varying extent. The incidence ranges from 0.42 to 5.1 in 1,000 live births.^{1,2} Although the majority of these children recover, in some children sequelae (paresis, joint contractures, and deformities) persist.³⁻⁵ Contractures of the shoulder, especially internal rotation contractures, are common in OBPL and are related to functional disability.^{2,6}

The pathophysiology of contracture formation in OBPL is incompletely understood. Contracture formation starts with muscle abnormalities. According to an experimental mice study,⁷ denervation after neurectomy leads to reduced length and cross-sectional growth of the muscles. These growth differences were related to the contracture formation. Beside these growth differences qualitative changes as increased fibroses were found, although the relation between fibrosis and contractures was not strong.

Some of these muscle changes have also been found inconsistently in children with shoulder contractures after OBPL; Einarsson et al found reduced sarcomere length and increased fibrosis in the subscapular muscle after OBPL,⁸ but a later study found no abnormalities.⁶

In infants shoulder muscle atrophy was related to shoulder joint subluxation,⁹ but upper arm muscle atrophy was not related to residual muscle function.¹⁰

Data on the residual strength of affected muscles in neuromuscular disorders and their relation to joint contractures are scarce. A recent study found both reduced passive range of motion and muscle strength for internal and external rotations of the shoulder in children with OBPL, but the relation between range of motion and strength was not tested¹¹.

Muscle imbalance is a factor often assumed to be of importance in contracture formation in OBPL.^{3,4} Intuitively the notion that imbalance leads to contracture formation is attractive, but data to support it are scarce. In an experimental mice study,⁷ excision of the external rotators was not able to cause contractures, but in another mice study using temporary paresis¹² with botulinum contractures occurred.

Little is known about residual strength of affected muscles in established joint contractures and whether there is a relation between the degree of contracture and muscle strength. In the Dupuytren hand flexion contracture of the fingers has been related to grip strength,¹³ but findings of this local soft tissue disorder cannot be extrapolated to contractures of the larger muscles in OBPL. In patients with stroke, strength and contracture of the elbow flexors were weakly related at 26 and 39 weeks but not after 1 year.¹⁴ Applying these findings to OBPL is debatable, because stroke is an upper motor neuron lesion generally occurring to full-grown adults. In OBPL the lower motor neuron lesion occurs at birth and contractures develop over time but do not affect all children with OBPL.^{2,3,6}

According to the craniocaudal direction of the injury to the cervical nerve roots, one would expect the external rotators (C4-6) to be weaker than the internal rotators (C5-8), a

situation assumed to lead to the often-described internal rotation contracture.^{3,4} Our hypothesis is that in OBPL internal rotators are stronger than external rotators and that the degree of joint contracture would be related to the balance between the residual strength of internal and external rotators.

These hypotheses were tested in a clinical study in children with OBPL, assessing the relation between passive shoulder rotations, residual muscle strength of internal and external rotators, and residual active shoulder function.¹⁵

Methods

Participants

Included were 34 consecutive children (20 boys and 14 girls) with unilateral OBPL with a mean age of 10.0 years, attending the regular, multidisciplinary checkup in the VUMC between December 1, 2009, and August 1, 2011. The extent of OPBL was classified according to Narakas.¹⁶ Excluded were children with Narakas class IV (total paresis of the arm) and patients younger than 4 years, because their cooperation on examination was expected to be poor. The study was in accordance with the rules of the medical ethical commission of the VUMC.

Muscle Strength

Isometric peak muscle strength was measured in Newton with the Microfet 2 dynamometer (Biometrics, Almere, The Netherlands) by the same orthopaedic surgeon on both normal and affected sides. Each measurement was repeated three times and the median value was noted. External and internal rotation strengths were measured standing upright with the shoulder in 0° abduction and rotation as neutral as possible. The elbow was flexed to 90°, the lower arm in neutral degrees of pronation/supination. For external rotation the Microfet 2 was placed on the dorsal side of the distal forearm just proximal of the wrist. For internal rotation it was placed on the palmar side. The patients were requested to keep their upper body still while giving maximal pressure to the dynamometer (► Fig. 1). This method for isometric peak strength measurement has excellent validity.¹⁷ Strength on the affected side was given as percentage of the normal side to correct for age and interindividual differences.

To quantify the balance between residual internal and external rotator strengths, the ratio between the residual internal and external strengths was calculated and related to contracture formation.

Contracture

As a measure of shoulder joint contracture, passive rotations on the affected side were measured with a standard goniometer in standing position with the shoulder in 0° abduction. For the internal rotation contracture, passive external rotation was measured. For external rotation contracture, passive internal rotation was measured.

Clinical Outcome

Active shoulder function was measured using the method of Mallet¹⁵ (► Table 1). This score consists of five dimensions:

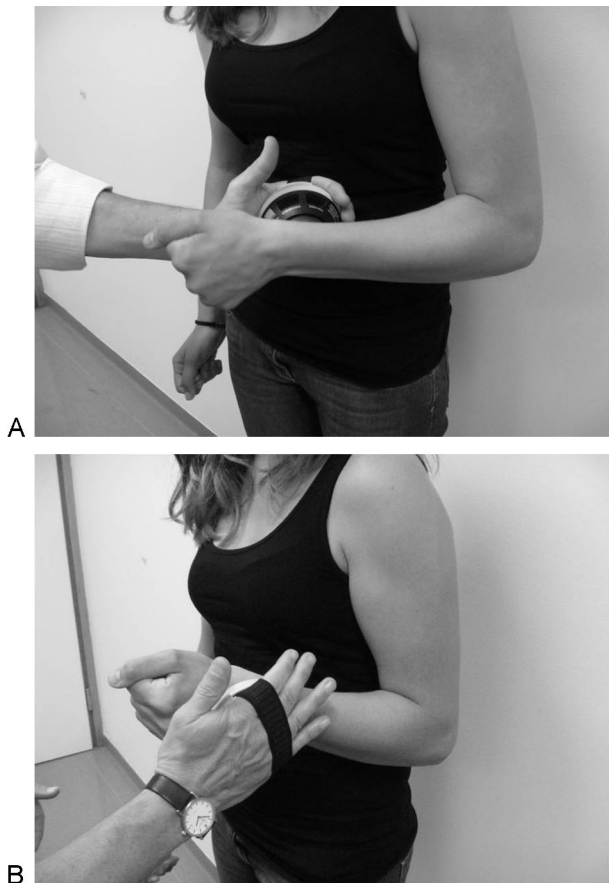


Fig. 1 Measurement of muscle strength. (A) Internal rotation strength is measured with the Microfet 2. (B) External rotation strength is measured.

abduction, external rotation, hand to neck, hand to back, and hand to mouth. We used the total Mallet score and the distinctive scores for external rotation and hand to back (the latter being a dimension for active internal rotation).

Statistics

All data were collected and analyzed in SPSS for Windows (version 15.0). Normal distribution was tested with Kurtosis and Skewness tests. Results are given as mean (standard deviation). Correlations were calculated using Pearson's for interval data or Spearman's for ordinal data test. Differences

were tested using paired *t*-tests. All tests were two tailed and $p < 0.05$ was considered to be significant.

Results

In this study 34 children with unilateral OBPL were included, with a mean age of 10.0 years (range 4.7–16.0 years, standard deviation [SD] 1.5), 20 boys and 14 girls. The right side was affected in 21 children, the left in 13. Narakas class was I in 19 children, II in 4 children, and III in 11 children, with a mean Narakas class of 1.8 (SD 1.0). In addition, 19 children had prior neurosurgery, 15 did not; 5 children have had secondary surgery around the shoulder. All data were normally distributed. Main descriptives and correlations are shown in ►Tables 2 and 3, respectively.

Muscle Strength

Absolute muscle strength on the affected side was always less than that on the normal side, except for the internal rotation strength in one 11.8-year-old boy (106 and 102 Newton, respectively). There was no significant difference (paired *t*-test $p = 0.26$) between mean residual external (56%) and internal rotation strength (51%) on the affected side (►Table 2). By approximation external and internal rotation strengths were both equally affected as shown in (►Fig. 2) and their correlation was significant (►Table 3).

There was no relation between residual muscle strength and shoulder joint contracture (►Table 3), or between the ratio of residual internal and external strength and joint contractures.

Muscle strength was related to the Mallet score (►Table 3). The Mallet subscore global external rotation was not related to muscle strength, whereas subscore hand to back was related to both muscle strengths (internal rotation $r = 0.52$, external rotation $r = 0.50$; $p < 0.01$).

Shoulder Joint Contracture

Passive external and passive internal rotations were not interrelated (►Table 3). Passive external rotation was related to the total Mallet score, whereas passive internal rotation was not. Passive external rotation was related to the Mallet subscore global external rotation ($r = 0.78$, $p = 0.000$), whereas passive internal rotation was related to subscore hand to back ($r = 0.35$, $p = 0.04$).

Table 1 Measurement of active shoulder function according to Mallet

Functional parameter	Class 1	Class 2	Class 3	Class 4	Class 5
Abduction	None	<30°	30-90°	>90°	Normal
External rotation	None	<0°	0–20°	>20°	Normal
Hand to neck	None	Not possible	Difficult	Easy	Normal
Hand to back	None	Not possible	S1	Th12	Normal
Hand to mouth	None	Marked trumpet sign ^a	Partial trumpet sign ^a	<40-degree abduction	Normal

Notes: Normal function in every dimension is 5 points, absent function is 1 point. Total Mallet score is the sum of the five sub-scores.

^aTrumpet sign is abduction of the shoulder with simultaneous flexion of the elbow.

Table 2 Descriptive of output values in mean, SD, range, and 95% CI

	Descriptives	Mean	SD	Range	95% CI
	Age (month)	120	32	57–191	108–132
	Narakas class	1.8	1.0	1–3	1.4–2.1
Normal side	Internal rotation strength	84 N	26 N	35–145 N	75–94 N
	External rotation strength	64 N	21 N	27–130 N	56–72 N
Affected side	Internal rotation strength	42 N	26 N	1–106 N	32–51 N
	External rotation strength	35 N	18 N	15–76 N	29–42 N
Ratio affected/normal	Internal rotation strength	51%	27%	0.01–1.04%	41–60%
	External rotation strength	56%	18%	20–86%	49–62%
Contracture	Passive external rotation	27°	34°	–40–90°	15–39°
	Passive internal rotation	67°	18°	30–90°	61–74°
Mallet score	Total	14.8	2.5	11–19	14–16
	External rotation	2.6	0.8	2–4	2.3–2.9
	Hand to back	3.0	0.9	2–4	2.7–3.3

Abbreviations: CI, confidence interval; N, Newton; SD, standard deviation.

Table 3 Correlations between residual strength (% of normal) and contractures. Shown are *r* values, with *p* value subscribed

	External rotation strength	Internal rotation strength	Passive external rotation	Passive internal rotation
Internal rotation strength	0.60 <i>p</i> = 0.000			
Passive external rotation	0.22 0.21	0.07 0.71		
Passive internal rotation	0.21 0.24	0.02 0.91	–0.23 0.18	
Total Mallet	0.45 <i>p</i> = 0.007	0.42 <i>p</i> = 0.014	0.52 <i>p</i> = 0.002	0.24 0.17

Narakas Class and Age

Age was related to all absolute muscle strengths ($p < 0.001$), not to any of the other parameters. Narakas class was only related to internal rotation strength, not to any of the other parameters. In the higher and more severe Narakas classes, this internal rotation strength was weaker ($r = -0.43$, $p = 0.01$). In the subgroup of children with Narakas class 1, no relation was found between muscle strength and contracture (►Table 4), neither a relation was found in the other classes.

Discussion

Our hypothesis that in OBPL internal rotators in the shoulder are stronger than external rotators and the hypothesis that the balance between residual muscle strength of internal and external rotators is related to the internal rotation contracture were not confirmed.

There are several limitations of our study. The first limitation is that it is not clear which muscles were measured. Several muscles contribute to rotation of the shoulder, but their individual contribution is not fully known. Based on

electromyographic (EMG) studies in healthy patients, subscapularis and, to a lesser extent, pectoralis major are predominantly active during internal rotation while supra- and infraspinatus, trapezius, and serratus muscles are active during external rotation.^{18,19} It is unknown whether the relative contributions of the muscles in the OBPL child are the same. Changes in lateral myofascial strength transmission might possibly lead to changes in the relative contribution of the various muscles to rotation strengths.^{20,21}

Another limitation is the measurement of muscle strength. Measurement using a handheld dynamometer have been found to be reliable.¹⁷ In our study we used the median of three consecutive isometric measurements. However, isometric peak strength is among others dependent on muscle length.²¹ Though this was standardized by keeping the arm in neutral external rotation if possible, differences in muscle resting length in this position between the children might have influenced the results. Given the presence of contractures, the neutral position probably does not standardize the muscle length of the internal rotators in these children and in some they are stretched beyond their resting length, thus influencing peak muscle strength. The duration of the peak is

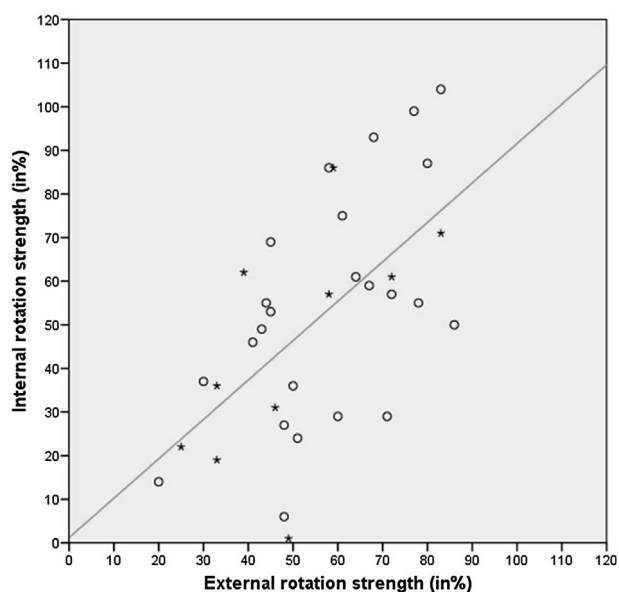


Fig. 2 Significant relation between external and internal rotation strengths ($r = 0.60, p = 0.00$). The distribution of children with severe internal rotation contractures (*, passive external rotation $\leq 0^\circ$) and children with mild contractures (O, passive external rotation $> 0^\circ$) is shown. Children with severe contractures are evenly distributed, and there is no relation between contracture and strength.

neither considered by the handheld dynamometer with differences in endurance not being measured. Peak values might be possibly subservient contributors to the function of these partially denervated muscles.

Another limitation is that the original lesion that predominantly affects the upper trunk (the external rotators) and that was the basis of our hypothesis on residual strength was influenced by the neurosurgery that was performed in the majority of patients (19 of 34). Finally, the contralateral side may have hypertrophied to compensate for the paretic arm, thus increasing the difference between affected and unaffected.

In spite of these caveats, peak strength measurement seems to measure a valid aspect of OBPL as it was related with Narakas class, a measure of the neurologic trauma, and with clinical outcome of OBPL as assessed by total Mallet score. We found that the Narakas class correlated with internal rotation strength but not with external rotator strength. Children with higher Narakas classes had weaker internal

rotation strength than those with lower classes. This makes sense as in Narakas class I only C5 and 6 are affected, and the subscapular muscle (internal rotator) is innervated by the subscapular nerves from C5 to 8. In higher Narakas classes in which C7 and C8 too are affected, internal rotation strength is weaker. As the external rotators (mostly C5 and 6) are affected in all Narakas classes, this explains why there was no relation between Narakas class and external rotation strength.

Based on the craniocaudal direction of the injury to the cervical nerve roots, we expected the external rotation strength (C4–6) to be weaker than internal rotation (C5–8). However, we found on average no difference between residual internal rotation strength (51% of normal side) and residual external rotation strength (56% of normal). In a substantial number of patients internal rotators were weaker than external rotators. Our results are not confirmed by the scarce literature; the Kirjavainen et al's study¹¹ found less residual strength in external rotator (28% of the contralateral side) than in internal rotators (44% of the contralateral side). Possibly the fact that 50% of their population had had secondary shoulder surgery (either soft tissues or humeral osteotomy) may have influenced their results although this would lead to a decrease in internal rotator strength, which is not what they found. Their measurement technique was different, but their shoulder position was identical. Part of the different results could be explained because they used the best of three values, in our study the median.

The lack of difference between internal and external rotators despite the fact that the neurologic lesion to the external rotators is larger is intriguing. Possibly the muscles were not measured on identical lengths in their strength length curves. In children with an internal rotation contracture, the neutral position lengthens the internal rotators and shortens the external rotators. The measurement points on the strength length curves would differ: internal is shifted to the right, external to the left. This bias would, however, lead to higher values for the internal rotators, which is not what we found.

The strength length curves of the affected muscles are probably different and the weakest muscles may reorganize their architecture to optimize their peak strength. The internal rotation contracture itself might be a symptom of this process. It stretches the external rotators and shortens the internal rotators, thus shifting their strength length curves in a way that increases external rotation strength and reduces

Table 4 Correlations in subgroup of children with Narakas class 1 ($n = 19$)

	External rotation strength	Internal rotation strength	Passive external rotation
Internal rotation strength	0.67 0.002		
Passive external rotation	0.27 0.27	-0.003 0.99	
Passive internal rotation	0.26 0.27	0.28 0.25	-0.34 0.16

Note: Correlations are shown in r value, with p value subscribed.

internal rotation strength. In this context the usual adaptation of the number of sarcomeres after shortening^{22,23} possibly does not seem to occur as muscle strength of the internal rotators is permanently reduced compared with normal.

Possibly increased fiber pennation, more parallel fibers, increased lateral strength transmission, from other muscles, and reduced tendon elasticity all contribute to maximization of peak strength in the external rotators.²¹ Alternatively, the internal rotators might possibly adapt to a reduced function of their antagonist by reduced growth because of inadequate stretching by the antagonist. This reduced growth might reduce their function. Both mechanisms might be responsible for the described balancing of the residual strengths around the shoulder.

As far the role of strength imbalance and contractures: in shoulders of children with OBPL, muscle imbalance is often mentioned as (or thought to be) a cause of secondary shoulder deformities.^{24–26} Muscle imbalance after experimental neurectomy in mice causes contractures.⁷ Hogenboom et al²⁴ suggest that muscle imbalance in less affected OBPL children contributes to shoulder deformity and that in more severe OBPL cases the absent imbalance protects these shoulders from severe deformity. However, we did not find a muscle imbalance in our study; even in our subgroup of children with Narakas class I, internal and external rotation strengths were equally affected and related.

The relation between strength imbalance and contractures is weak. Although on average there is no imbalance in peak muscle strength between internal and external rotations, in most children an imbalance is present, as shown in (→ Fig. 2), but this imbalance could not predict extent of contracture. The role of strength in contracture formation is further questioned by an experimental study⁷ where strength imbalance created by resection of the external rotator was not a dominant factor in contracture formation.

Based on experimental and clinical studies, differences in muscle growth seem important in the causation of contractures. In children with OBPL muscle volume imbalance has been described with the subscapularis muscle (internal rotator) being more affected than the infraspinatus (external rotator).^{5,9,25,27} In the cited neurectomy study⁷ subscapularis and elbow flexors had reduced longitudinal growth that correlated with contracture formation. Qualitative muscle changes also play a role. In the cited experimental study, muscle fibrosis was present, but the correlation with contractures not consistent. In OBPL children Einarsson et al found reduced sarcomere length and increased fibrosis in the subscapular muscle⁸ although, given the nature of this controls (other upper arm muscles), the changes in sarcomere length have to be interpreted with care and a later study from the same group found that muscle biopsies overall showed normal morphology.⁶ Structural muscle size abnormalities on magnetic resonance imaging (MRI) level in subscapularis and infraspinatus correlated with glenoid deformity and humeral head subluxation.²⁴

Reduced length growth of partially denervated muscles is an important factor in the pathophysiology of contracture

formation, while our results do not confirm that imbalances in residual muscle strength seem to be relevant.

Conclusion

In our study there was no relation found between shoulder joint contracture and residual muscle strength of internal and external rotators in OBPL. Muscle strengths of internal and external rotation were on average equally affected. The intuitive notion that imbalances in residual muscle strength lead to an internal rotation contracture of the shoulder cannot be confirmed in this study. Reductions in growth rate in partially denervated muscles are more likely causes of contractures.

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