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## Observational Study

# Humeral retroversion and shoulder muscle changes in infants with internal rotation contractures following brachial plexus birth palsy

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## Abstract

### AIM

To examine humeral retroversion in infants who sus-

tained brachial plexus birth palsy (BPBI) and suffered from an internal rotation contracture. Additionally, the role of the infraspinatus (IS) and subscapularis (SSc) muscles in the genesis of this bony deformation is explored.

## METHODS

Bilateral magnetic resonance imaging (MRI) scans of 35 infants (age range: 2-7 mo old) with BPBI were retrospectively analyzed. Retroversion was measured according to two proximal axes and one distal axis (trans-epicondylar axis). The proximal axes were: (1) the perpendicular line to the borders of the articular surface (humeral centerline); and (2) the longest diameter through the humeral head. Muscle cross-sectional areas of the IS and SSc muscles were measured on the MRI-slides representing the largest muscle belly. The difference in retroversion was correlated with the ratio of muscle-sizes and passive external rotation measurements.

## RESULTS

Retroversion on the involved side was significantly decreased,  $1.0^\circ$  vs  $27.6^\circ$  (1) and  $8.5^\circ$  vs  $27.2^\circ$  (2), ( $P < 0.01$ ), as compared to the uninvolved side. The size of the SSc and IS muscles on the involved side was significantly decreased,  $2.26 \text{ cm}^2$  vs  $2.79 \text{ cm}^2$  and  $1.53 \text{ cm}^2$  vs  $2.19 \text{ cm}^2$ , respectively ( $P < 0.05$ ). Furthermore, the muscle ratio (SSc/IS) at the involved side was significantly smaller compared to the uninvolved side ( $P = 0.007$ ).

## CONCLUSION

Even in our youngest patient population, humeral retroversion has a high likelihood of being decreased. Altered humeral retroversion warrants attention as a structural change in any child being evaluated for the treatment of an internal rotation contracture.

**Key words:** Humeral retroversion; Infants; Brachial plexus; Brachial plexus neuropathies; Shoulder; Humerus

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**Core tip:** This study examines humeral retroversion in infants who sustained neonatal brachial plexus palsy and suffered from an internal rotation contracture. The existing common treatment options all strive for better function of the upper extremity through an improved position of the hand in space. Therefore, a thorough understanding of the development of the pathogenesis of this injury is important. We found a significant reduction of humeral retroversion in our study group (mean difference, 26.8). When treatment becomes warranted and contralateral humeral version measurements greatly differ, a humeral derotational osteotomy may offer the best improvement regarding the hand position.

van de Bunt F, Pearl ML, van Essen T, van der Sluijs JA. Humeral retroversion and shoulder muscle changes in infants with internal rotation contractures following brachial plexus birth palsy. *World J Orthop* 2018; 9(12): 292-299

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## INTRODUCTION

The most common musculoskeletal sequela of neurologic injury of brachial plexus birth palsy (BPBI) is an internal rotation contracture of the shoulder. This contracture is frequently associated with deformity of the glenohumeral joint<sup>[1-5]</sup>. These bony deformities have been thought to be a consequence of abnormal muscular development<sup>[6-8]</sup>.

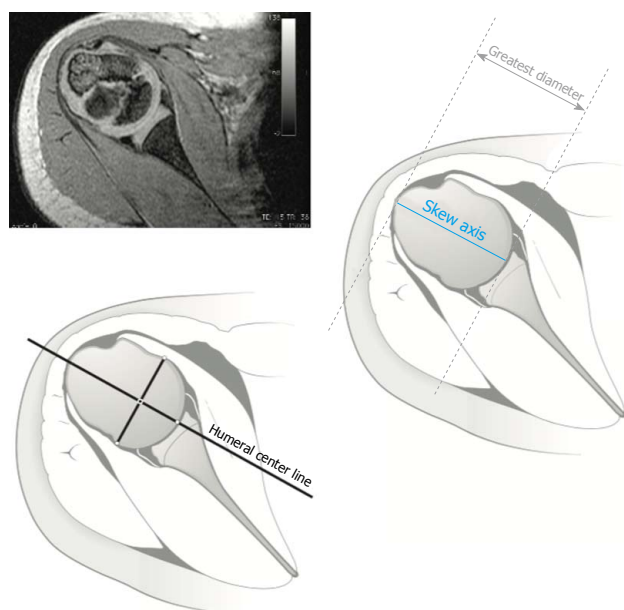
The internal rotation contracture secondary to BPBI has been associated with alterations of humeral retroversion<sup>[9-12]</sup>. Previous studies presented opposite findings, as both older studies reported an increased humeral version angle<sup>[10,11]</sup>, while more recent studies reported a decrease in humeral retroversion<sup>[9,12]</sup>. Normal humeral retroversion is greatest at birth and gradually decreases through adolescence<sup>[13-15]</sup> to adult values averaging between 25-30 with well documented individual variation<sup>[16]</sup>. One well-studied exception is the throwing athlete, for whom retroversion has been shown to be greater on the dominant throwing side, due to repetitive throwing that usually begins in early childhood<sup>[17-21]</sup>.

The existing common treatment options consist of soft tissue procedures (releases and tendon transfers) and bone realignment procedures (rotational osteotomy) with the aim to provide better function of the upper extremity through an improved position of the hand in space<sup>[22-26]</sup>. This position is directly related to the humeral version angle. We studied humeral retroversion in 35 consecutive infants who were under evaluation for treatment of their internal rotation contractures secondary to unilateral BPBI in this retrospective observational study. Our main goal was to further elucidate the timing that these anatomic changes may occur; therefore, we included our youngest patient population. We hypothesized that the retroversion angle (RV-angle) on the involved side would be significantly decreased relative to the uninvolved side and that the difference would increase with age. Since the subscapularis (SSc) and infraspinatus (IS) muscles, are an agonist-antagonist muscle pair regarding humeral rotation, we hypothesized that an imbalance between these muscles would correlate with altered humeral version.

## MATERIALS AND METHODS

In this retrospective observational study, we included 37 Magnetic resonance imaging (MRI) -scans from a consecutive series of infants ( $< 1$  year old) with a unilateral BPBI. All infants were potential candidates for neurosurgical interventions because of the severity of the





**Figure 1** Schematic illustration of measurement parameters applied to a magnetic resonance imaging slice from the proximal part of the normal uninvolved humerus. (Reproduced with modification from: Pearl ML, *et al.* Geometry of the proximal humeral articular surface in young children: a study to define normal and analyze the dysplasia due to brachial plexus birth palsy. *J Shoulder Elbow Surg* 2013; **22**: 1274-84. Reproduced with permission from Elsevier.)

neurological lesion. This study was IRB approved.

MRI studies were performed on a 1.5-T MRI-unit (Magnetom 1.5 T Vision; Siemens, Erlangen, Germany). A FISP three-dimensional pulse acquisition sequence (repetition time, 25 msec; time to echo, 10 msec; flip angle 40°) with ranges from 0.8 to 1.5 mm partitions was used to obtain images from both shoulders and upper arms, representing the full humerus and glenohumeral joint in the axial plane. All children were given pethidine, droperidol and chlorpromazine intramuscularly. During sedation, they were monitored by electrocardiograph, measurement of oxygen saturation, and by video. Children were not moved during the imaging protocol.

From these 37 studies, two were insufficient for completing our detailed measurement protocol, as one study did not capture the entire humerus and motion artifacts compromised the other study.

Our Radiology department anonymized the MRI studies before performing our measurement protocol; Digital Imaging and Communications in Medicine files were imported as a numerical database into Osirix (Pixmeo, Geneva, Switzerland). For humeral version measurements, axial plane slides from the involved and uninvolved side that to our best efforts represented the midpoint of the humeral head were selected. For the measurement of muscle dimensions, axial plane slides representing the largest cross-sectional area of the SSc muscle and infraspinatus muscle were selected and exported as TIFF files. The TIFF files were imported into Geometer's Sketchpad version 5.03 (KCP Technologies, Emeryville, CA, United States) for further retroversion

analyses. The region of interest tool available in Osirix was used for muscle cross-sectional area measurements. The Narakas classifications were assigned as described by Narakas<sup>[27]</sup>. Passive external rotation was measured with the arm in the adducted position and the elbow by the side.

### Measure of retroversion

Retroversion was measured with respect to two different methods for the proximal humeral axis and the transepicondylar axis distally, introduced by Pearl *et al.*<sup>[12]</sup>.

The first proximal reference axis was chosen to provide continuity with earlier retroversion analysis performed in this specific patient group<sup>[10,11]</sup>. This axis is conforming to the longest diameter through the humeral head. A line segment was created, which spanned the greatest distance from the periphery of the greater tuberosity to the medial articular surface and is labeled as the skew axis (SA) (Figure 1)<sup>[2]</sup>.

Retroversion was analyzed using the humeral center-line (HCL) as the proximal axis (Figure 1). This is a commonly used axis in various retroversion studies<sup>[19,28-32]</sup>. The HCL represents the perpendicular projection from the margins of the articular surface.

Based on the literature, retroversion of the humeral head is shown as a positive value and anteversion is shown as a negative value. Two investigators performed the humeral version measurements.

### Surface area measurements

Cross-sectional areas of the IS and SSc muscles were measured using the closed region-of-interest polygon tool in Osirix (Pixmeo). The MRI slides depicting the largest muscle bellies were identified for measurement of this cross-sectional area. Muscle size was determined by the muscle cross-sectional area in cm<sup>2</sup> and muscle percentage relative to the corresponding muscle at the uninvolved side. Furthermore, the ratio of the SSc and IS muscle (SSc/IS) was calculated to compare muscle balance between both sides and correlate these with the  $\Delta$ RV-angle.

### Analysis

Statistical analysis was performed using SPSS software (version 22.0; SPSS Inc., Chicago, IL, United States). The distribution analysis showed an approximately normal distribution.

Standard descriptive measures as mean, standard deviation, minimum and maximum values are reported for retroversion of the involved and uninvolved sides, as for the muscle surface area measurements, and their difference ( $\Delta$ ) within the study population. Pearson product-moment or Spearman rank correlation coefficients are estimated between each of these and passive external rotation and Narakas classification, as appropriate, based on the underlying distribution and type of the data. Paired data, such as involved vs uninvolved measurements regarding retroversion and muscle cross-sectional area measurements made on the same

Table 1 Demographics

Subject	Narakas	Age (mo)	External rotation (passive) (°)	Retroversion involved (HCL) (°)	Retroversion involved (SA) (°)	Retroversion uninvolved (HCL) (°)	Retroversion uninvolved (SA) (°)
1	3	2.6	-15	7.665	11.25	26.4	26.315
2	1	3.1	-5	-9.16	11.045	23.18	13.485
3	1	3.2	-40	-17.125	4.04	18.65	17.05
4	3	3.2	-30	14.175	23.395	30.56	31.865
5	3	3.3	0	-7.05	6.67	31.23	32.85
6	3	3.4	-10	-24.74	-19.26	41.72	31.705
7	3	3.4	0	7.67	7.905	30.145	29.165
8	3	3.5	-20	35.595	35.015	27.295	31.23
9	2	3.5	-5	-10.885	1.615	24.17	23.905
10	1	3.5	-20	4.54	2.05	32.21	26.49
11	3	3.6	-20	1.99	5.695	43.55	36.11
12	1	3.6	-20	3.6	22.565	54.905	47.955
13	3	3.8	-25	1.595	9.425	29.355	36.42
14	1	4.0	-5	-6.715	3.44	29.115	28.165
15	2	4.1	-15	-13.53	-1.035	21.59	25.605
16	1	4.1	-15	14.975	9.85	25.575	21.24
17	3	4.5	-25	0.065	-2.075	19.47	19.995
18	3	4.5	-45	24.195	20.195	34.19	34.55
19	1	4.5	-10	-4.115	6.1	24.305	20.175
20	2	4.6	-30	-7.205	11.675	13.465	14.06
21	3	4.6	-10	-3.14	7.29	15.445	12.14
22	1	4.7	-20	4.125	18.195	23.045	30.385
23	1	4.7	-20	-20.83	1.9	21.85	30.085
24	1	4.8	-40	4.875	9.95	15.655	20.11
25	3	4.9	-40	8.935	9.525	18.805	11.765
26	1	5.0	-15	38.24	33.53	20.055	15.945
27	1	5.0	0	-8.86	4.405	24.85	21.6
28	1	5.0	-15	-30.23	-20.135	38.975	23.31
29	1	5.0	-10	24.725	25.535	32.98	39.03
30	1	5.1	-5	8.79	10.05	20.115	-2.295
31	1	5.4	-20	-28.55	-11.965	47.445	39.185
32	3	5.6	-35	3.385	6.45	30.395	27.485
33	3	5.9	-15	-16.805	11.66	18.085	14.5
34	2	5.9	-30	11.89	7.225	28.56	35.075
35	1	6.5	-10	17.315	14.43	31.08	22.5
Mean		4.3	-18.3	0.8	8.5	27.7	25.4
Standard deviation		0.9	12	16.1	11.7	9.2	9.8
Minimum		2.6	-45	-30.23	-20.135	13.465	-2.295
Maximum		6.5	0	38.24	35.015	54.905	47.955

HCL: Humeral center-line; SA: Skew axis.

subject, were compared using paired t- or paired-samples Wilcoxon's signed-rank tests, as appropriate. Inter-rater reliability assessment by Intraclass correlations coefficient (ICC) was performed. A Bland-Altman plot was created to visualize potential differences in retroversion measuring methods<sup>[33]</sup>.

## RESULTS

The 35 children included in our study had a mean age of 4.3 mo (range of 2.1-6.5 mo), and they were classified according to the Narakas classification: Narakas I : 18 cases; Narakas II : 4 cases; Narakas III : 15 cases. Internal rotation contractures varied from -45° to 12°, with a mean of -18°, measured as passive external rotation with the elbow by the side (Table 1).

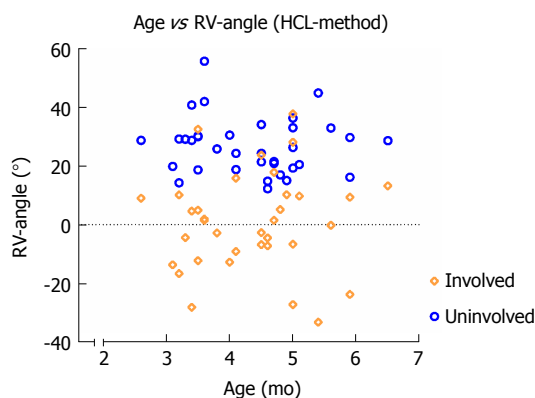
### Humeral retroversion by HCL

Retroversion measured according to the HCL and the

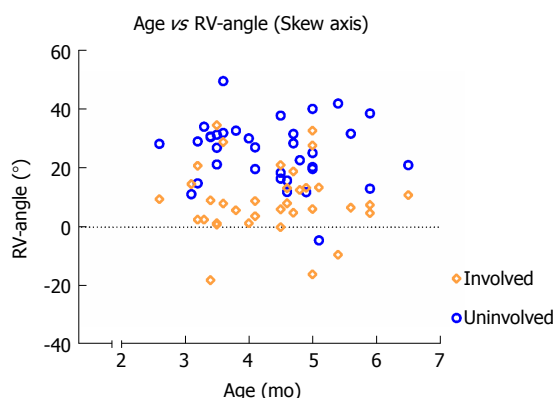
transepicondylar axis was significantly decreased on the involved side as measured by both observers. Mean RV-angles were 0.8° vs 27.7° ( $P < 0.001$ ). Paired differences averaged 26.8°, with a range from -18.4° to 77.8°. Figure 2 shows the distribution of the measurements. In two patients, retroversion increased on the involved side (Table 1). Age did not correlate with a decrease in humeral retroversion ( $r = -0.108$ ,  $P = 0.538$ ).

### Humeral retroversion by SA

Retroversion measured according to the SA and the transepicondylar axis was also significantly decreased on the involved side, as measured by both observers. Mean RV-angles were 8.5° vs 25.4° ( $P < 0.001$ ). Paired differences averaged 17.5°, with a range from -22.2° to 53.3°. Figure 3 shows the distribution of measurements. In five patients, retroversion was increased on the involved side (Table 1). Age was again not correlated with a decrease in humeral retroversion ( $r = -0.120$ ,  $P =$



**Figure 2** The distribution among measurements using the humeral center line as a proximal axis. HCL: Humeral center line; RV-angle: Retroversion angle.



**Figure 3** The distribution among measurements using the skew axis as a proximal axis. In the deformed humeral head, the skew axis yields systematically higher values compared to the humeral center line. RV-angle: Retroversion angle.

0.492).

### Muscle surface area

Both muscles were significantly smaller on the involved side. The IS muscle measured a mean surface area of 2.35 cm<sup>2</sup> vs 2.84 cm<sup>2</sup> (83%) ( $P < 0.001$ ), and the SSC muscle was 1.56 cm<sup>2</sup> vs 2.20 cm<sup>2</sup> (70%) ( $P < 0.001$ ).

Furthermore, the muscle ratio (SSc/IS) on the involved side was significantly smaller compared to the uninvolved side ( $P = 0.007$ ). In Table 2, the results of the muscle cross-sectional area measurements are summarized.

### Correlations

Pearson's product correlation tests were performed for the retroversion measurements, the  $\Delta$ RV-angle and the muscle area ratios and muscle surface area measurements, however no significant correlations were found on the involved side. When correlating age with decrease of retroversion, the Spearman Rho test was performed for retroversion measurement and Narakas' score and passive external rotation, no significant correlations were found ( $P > 0.05$ ).

### HCL method vs SA

For retroversion measured by HCL, the ICC for interrater reliability on the involved side was 0.934 (95%CI: 0.863-0.967;  $P < 0.001$ ). The ICC for interrater reliability on the uninvolved side was 0.889 (95%CI: 0.747-0.948;  $P < 0.001$ ). For retroversion measured using the SA, the ICC for interrater reliability on the involved side was 0.934 (95%CI: 0.897-0.970;  $P < 0.001$ ). The ICC for interrater reliability on the uninvolved side was 0.923 (95%CI: 0.853-0.960;  $P < 0.001$ ).

The distribution of measurements was larger on the involved side (Figure 4). Both measurement methods yielded comparable results in the uninvolved shoulder. However, the SA yielded systematically higher values in the deformed humeral head compared to the HCL.

## DISCUSSION

We found a significant reduction of humeral retroversion on the involved side compared to the uninvolved side in a consecutive series of patients with internal rotation contractures secondary to BPBI. Additionally, the size of the SSc and IS muscles on the involved side was significantly decreased, as was the muscle ratio (SSc/IS) on the involved side compared to the uninvolved side.

Considering the RV-angles measured, our results are similar to those reported by Pearl *et al.*, which were: 1.8° and 5.8° compared to 20.2° and 18.9°, respectively, depending on the method of measurement. However, the mean age of the study groups differed considerably, 3.2 years old vs 4.3 mo old. Our results suggest that declined humeral version is not something these children slowly grow into. The altered humeral version angle may already develop within the first weeks after birth, when the humerus is probably most prone to altered development caused by altered muscle forces gripping the humeral head. This is supported by the lack of significant correlation found between age and decreased retroversion on the involved side in both studies.

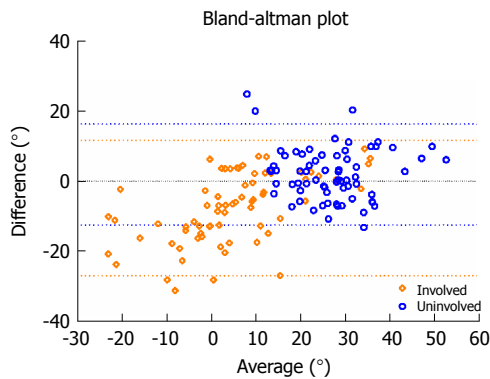
Of further note, the earliest reports by Scaglietti<sup>[11]</sup> and van der Sluijs *et al.*<sup>[10]</sup> found an increase in retroversion. Scaglietti's study was in a very different era of imaging technology and presented his observations with little quantitative data. van der Sluijs *et al.*<sup>[10]</sup> utilized MRI, but nearly two decades ago in a somewhat older age group, when current software tools were not available for image analysis, and the lesser image quality might have influenced measurements. Perhaps these methodological differences explain these contradictory findings.

Consistent with the literature, we observed a significant decrease in muscle size on the involved side compared to the uninvolved side, with the SSc muscle being more affected than the IS muscle<sup>[6,34-36]</sup>. However, no significant correlation between the muscle ratio (SSc/IS) and the humeral RV-angle was observed. Nonetheless, the reduction in muscle ratio does not support the theory that the internal rotators overpower the injured (paralyzed) external rotators, but suggests



**Table 2** Main results of the muscle cross-sectional area measurements

Muscle area, cm <sup>2</sup>	Mean - involved	Mean - uninvolved	P value
Subscapularis muscle	1.56 ± 0.315	2.20 ± 0.372	< 0.001
Infraspinatus muscle	2.35 ± 0.520	2.84 ± 0.495	< 0.001
Ratio	68.51 ± 16.90	78.88 ± 15.45	0.007



**Figure 4** The distribution of measurement in the involved shoulder is larger than on the uninvolved side, indicating measurement differences between the skew axis and humeral center line are larger on the involved side. The blue and orange dotted lines represent the 95% limits of agreement.

that failure of the SSc to grow or develop may result in a contracted SSc, which restricts external rotation.

Another theory could be that the changes in humeral retroversion are partially related to injured muscles outside of the rotator cuff, perhaps those with at least some innervation outside of the original zone of injury. Further study of other muscles is warranted, looking for evidence as to whether they were also injured resulting in impaired growth<sup>[7,37]</sup>, or whether they recovered so strongly that they overwhelmed their antagonists or are used differently in children with varying levels of recovery.

In addition, animal studies have shown that impaired longitudinal muscle growth and strength imbalance mechanisms are capable of producing shoulder deformities and impaired growth to a somewhat greater extent than muscle imbalance<sup>[8,38-41]</sup>. However, this has not yet been related to altered humeral version. For example, impaired growth and increased stiffness of the SSc muscle fibers may have a significant effect on humeral version development. In combination with other internal rotator muscles such as the pectoralis muscle, mechanical stiffness of these muscle fibers may not be directly related to cross-sectional muscle area measurements.

Further research is necessary to elucidate a causal relationship between those mechanisms and shoulder deformities, concerning both the humerus and glenoid, which will help guide clinical treatment decisions for BPBI.

This study has several limitations. The measurements made were based on axial slices of the humerus; measurements made from a 3D-reconstruction, as those performed by Sheehan and others, would have the potential for minimizing errors related to patient

positioning and inconsistent image acquisition. In our studied age group, the humeral head and epicondylar axis are mostly cartilaginous, making 3D-reconstruction of the humeral anatomy much more challenging than in a skeletally mature subject. While the software tools currently exist, they are labor intensive and extremely difficult to implement in clinical practice. Therefore, we chose to utilize methods often used in our clinic setting and shown in a prior publication<sup>[12]</sup>.

Analyses of the IS and SSc muscles are based on cross-sectional area measurements from the MRI-slice, depicting the largest muscle belly as used in multiple previous studies<sup>[6,35,36]</sup>. Capturing the full volume of both muscles would likely have been more informative; however, such software tools were not available to us. Furthermore, muscle thickness was only assessed for the IS and SSc muscles, and the measurement of other external and internal rotator muscles may offer additional insight into muscle behavior and its effect on humeral retroversion in this population.

The most common sequel and focus of surgical intervention in children with BPBI is an internal rotation contracture at the shoulder. These surgical interventions all aim for better function through an improved position of the hand in space. Humeral version undeniably affects hand functionality because with all other factors being equal, decreased humeral version results in an increase of the severity of the clinical presentation of an internal rotation contracture. A large reduction in humeral retroversion at a very young age could be a predictor (or an argument when apparent at an older age) for the necessity of a humeral derotational osteotomy to provide adequate improvement of hand and possibly elbow function. Furthermore, this study shows that secondary osseous changes can occur within several months in this patient population. A prospective study analyzing possible changes in humeral version in this patient population over time would be of interest, as it seems through these results and results from recent studies that changes in humeral version occur early, but that they may not change much after that.

In conclusion, humeral retroversion has a high likelihood of being significantly decreased in this patient population. These findings are relevant for any child under consideration for surgical intervention aiming to improve external rotation, since all other factors being equal, decreased humeral retroversion results in an increased severity of the clinical presentation of an internal rotation contracture. We measured these changes in infants 2-7 mo old and found that altered humeral development

can occur very early in life in a population where internal rotation contractures are apparent.

## ARTICLE HIGHLIGHTS

### Research background

The existing common treatment options for children suffering from brachial plexus birth palsy all strive for better function of the upper extremity through an improved position of the hand in space. This position is directly related to the humeral version angle.

### Research motivation

Since earlier studies did not reveal a correlation between age and decreased retroversion on the involved side, the question remained at what age this anatomic change may occur.

### Research objectives

Our objective was to elucidate the timing that decreased retroversion may occur; therefore, we included our youngest patient population (2-7 mo old).

### Research methods

We measured humeral version relative to two proximal axes and one distal axis (transepicondylar axis). The proximal axes were: (1) the perpendicular line to the borders of the articular surface (humeral centerline), and (2) the longest diameter through the humeral head. Additionally, cross-sectional areas of the infraspinatus (IS) and subscapularis (SSc) muscles were measured. The difference in retroversion was correlated with the ratio of muscle sizes.

### Research results

Retroversion on the involved side was significantly decreased,  $1.0^\circ$  vs  $27.6^\circ$  (1) and  $8.5^\circ$  vs  $27.2^\circ$  (2), ( $P < 0.01$ ), as compared to the uninvolved side. SSc and IS muscle size on the involved side was significantly decreased,  $2.26 \text{ cm}^2$  vs  $2.79 \text{ cm}^2$  and  $1.53 \text{ cm}^2$  vs  $2.19 \text{ cm}^2$ , respectively ( $P < 0.05$ ). Additionally, muscle ratio (SSc/IS) on the involved side was significantly smaller compared to the uninvolved side ( $P = 0.007$ ), but was not related to alterations in humeral version.

### Research conclusions

Our results show that altered humeral development can occur very early in life in a population where internal rotation contractures are apparent.

### Research perspectives

A large reduction in humeral retroversion at a very young age could be a predictor (or an argument when apparent at an older age), for the necessity of a humeral derotational osteotomy, to provide adequate improvement of hand and possibly elbow function. A prospective study analyzing changes in humeral version over time would be of interest to assess the predictive value of decreased retroversion at such a young age, concerning various treatment options (soft-tissue and bony).

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