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Development of the glenohumeral joint after subscapular release and open relocation in children with brachial plexus birth palsy: long-term results in 61 patients

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Background: We present the long-term results of remodeling of the glenohumeral joint after open subscapularis elongation and relocation of the humeral head in patients with an internal rotation contracture and joint incongruity due to brachial plexus birth palsy.

Methods: In this before-and-after study, 61 patients who underwent open subscapularis elongation and reduction of the glenohumeral joint were evaluated with respect to joint remodeling, with a mean follow-up period of 10.2 years (range, 7-16 years). The mean age at operation was 3.2 years (range, 8 months to 15 years). Measurements of the percentage of the humeral head anterior to the midscapular line (PHHA), glenoid version, and diameter of the humeral head were recorded using magnetic resonance imaging, comparing the affected joints preoperatively vs. postoperatively (n = 31) and comparing the operated vs. unaffected sides postoperatively (n = 61).

Results: The mean increase in PHHA was 27.6 percentage points (95% confidence interval, 22.4-32.7 percentage points; $P < .01$), from 13.2% to 40.8%. The glenoid retroversion changed by 14.8° (95% confidence interval, 11.1°-18.4°; $P < .01$), from 25.4° to 10.6°, approaching a normal value. All patients, even those older than 5 years, showed a clear benefit from surgery.

Conclusions: Our study confirms that open subscapularis lengthening with joint repositioning, up to the age of 5 years, gives consistent remodeling of incongruent shoulders with surprisingly small differences between the operated and unaffected shoulders at long-term follow-up. The findings indicate that open reduction is useful also in adolescents and challenges the notion that older children should be treated with derotational humeral osteotomy.

Level of evidence: Level IV; Case Series; Treatment Study

Keywords: Brachial plexus birth palsy; glenohumeral joint remodeling; subscapularis elongation; glenohumeral dysplasia; internal rotation contracture; glenohumeral joint reduction

Ethical approval was granted by the regional ethical review board of Stockholm under number 2011/790-31/4.

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In the Swedish setting, brachial plexus birth palsy (BPBP) occurs in 2.2 to 2.9 of 1000 live births.[1,18,20] Of those affected, 70% to 80% recover clinically to achieve full function.[18,24] Among those who do not recover fully, the most common sequela is an internal rotation contracture with or without a deforming incongruence of the glenohumeral joint.[6] The contracture limits the ability to position the hand in space and makes it difficult to work in front of the body and to reach the head. Historically, the joint deformity has been described as a consequence of muscular imbalance with a progression from retroversion of the glenoid to posterior subluxation or even dislocation of the humeral head.[6,8,10,14,27,28,30,31] A recent study has shown that the glenohumeral incongruence can occur as early as at 3 months of age,[25] and we have seen complete dislocation at 6 months of age. In our previous studies, roughly 45% of the patients with a rotational contracture also had some degree of glenohumeral incongruity[12,13]; this is slightly less than in comparable studies.[31]

Together with the release of the internal rotation contracture, the treatment of the deformed or incongruent glenohumeral joint has been central to the management of BPBP for over 100 years. Whitman[32] performed closed reduction and casting with the goal of obliterating the old articulation. Fairbank[6] described the incongruity as the only hindrance for recovery in cases with a rapid return of nerve function, proposing tenotomy of the subscapularis tendon and open repositioning of the joint. Carlioz and Brahimi[5] suggested that there were disadvantages to dividing the subscapular tendon and advocated disinsertion of the subscapularis at its origin. Birch and Chen[4] presented the method that we have used since 1997: controlled lengthening of the subscapularis tendon (rather than the tenotomy proposed by Fairbank) and open precise relocation of incongruent glenohumeral joints. Pearl[21] began performing arthroscopic release of the entire subscapularis tendon and overlying capsule with or without latissimus transfer in 1999.[23] Kozin et al[16] focused on releasing the glenohumeral ligaments and aimed to preserve the inferior part of the subscapularis tendon to retain internal rotation in the shoulder. This concept was further developed in publications from Abid et al[2] and Kany et al[15] with the SPARC (subscapularis-preserving arthroscopic release of capsule) procedure, which preserves the entirety of the subscapularis tendon, arthroscopically dividing the glenohumeral and coracohumeral ligaments. Initial publications showed short-term improvement in dysplasia, but a subsequent publication did not advocate preserving the whole subscapular tendon in cases with posterior dislocation.[2,15]

Pearl and Edgerton[22] and Waters et al[31] described the radiologic changes of BPBP-affected joints, building on the works of Gudinchet et al[9] and Friedman et al.[7] In earlier works, the short-term and medium- to long-term results of addressing joint incongruity have been investigated. Hui and Torode[11] found that lengthening of the pectoralis major, division of the subscapularis, and open reduction of the joint in 23 patients gave a 31% decrease in glenoid retroversion at a mean follow-up time of 3 years 7 months (range, 1 year 10 months to 5 years 6 months); they also noted that the improvement in glenoid version increased over time. Waters and Bae[30] found that tendon transfer with concomitant open or arthroscopic reduction of the joint in 23 patients gave a 21° and 25% improvement in glenoid version and percentage of the humeral head anterior to the midscapular line (PHHA), respectively, at a mean follow-up time of 2 years 1 month (range, 11-55 months). Their investigation of the same tendon transfer without open or arthroscopic reduction of the joint in 25 patients did not give clinically relevant improvements in glenoid version and PHHA at a mean follow-up time of 4 years 1 month (range, 2-10 years).[30] Kozin et al[17] similarly found that tendon transfer with or without concomitant pectoralis lengthening and subscapular release yielded no significant changes in retroversion of the glenoid or PHHA in 19 patients examined retrospectively at 3.2 years postoperatively.

The purpose of this before-and-after study was to evaluate the long-term potential for remodeling of the incongruent glenohumeral joint after subscapular elongation and open joint reduction.

Methods

Patients

Between 1997 and 2009, 97 patients with BPBP and incongruent glenohumeral joints were operated on by the senior author (T.H.). The indication for surgery was (1) active external rotation of 0° or less or (2) active external rotation of less than 20° in the presence of a trumpet sign. During the operation, access to the subscapularis tendon is made through an anterior deltopectoral incision and the coracoid tip is reduced; the subscapularis tendon is then divided in a step cut, and the joint capsule is opened to inspect and relocate the joint. The subscapularis tendon is repaired with elongation, allowing for adequate external rotation. Detailed information on the indications and procedures are available in earlier publications.[12,13]

All patients were offered an examination for long-term follow-up at a minimum of 7 years after surgery, including a postoperative magnetic resonance imaging (MRI) scan. We excluded patients who, at the time of follow-up, were too young to be able to lie still for MRI without sedation.

We divided the patients into 3 groups based on age at surgery: younger than 2 years, 2 to 5 years, and older than 5 years. The age categories were initially chosen for our previous publication[12] as reinnervation to the shoulder is usually complete at age 2 years,[24] and clinical follow-up of operatively and nonoperatively treated patients was set to 5 years of age to detect sequelae before the children started school. We strive to operate on patients as soon as they fulfill our criteria,[12] and we can usually offer surgery without long delays. The patients in the oldest age group had all been managed in other clinics initially.

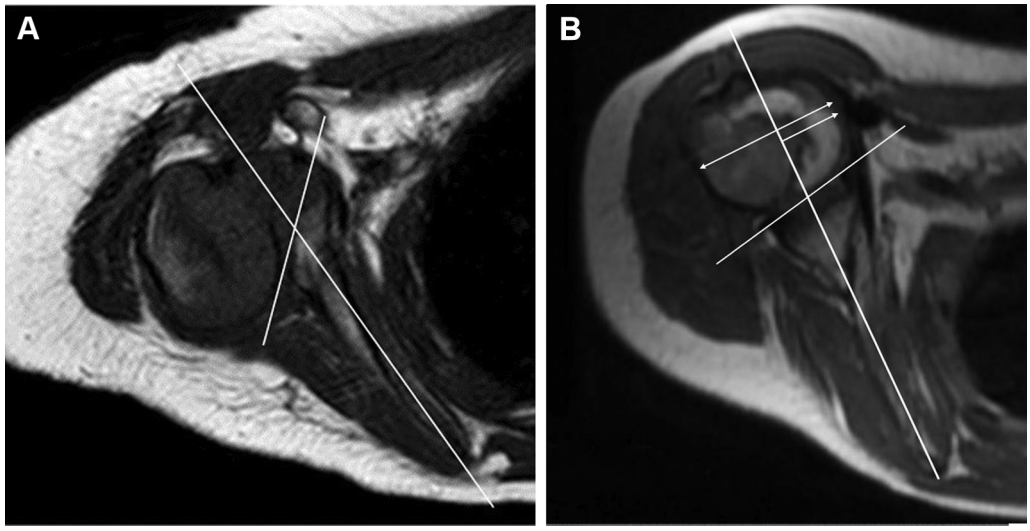

Figure 1 Images of bifaceted glenoid preoperatively with lines representing the glenoid angle measurements (**A**) and at 7 years postoperatively with lines representing the glenoid angle measurements and arrows indicating the PHHA measurements (**B**).

Classification and radiology

Our joint classification was based on direct inspection during surgery rather than on radiology. Three distinct patterns were identified among the incongruent joints: (1) a flattened posterior glenoid in which the humeral head lies in posterior subluxation and smoothly transitions anteriorly in outward rotation once the contracture has been released, (2) a bifaceted glenoid with a distinct ridge between the anterior fossa and posterior fossa in which the humeral head transitions with a snap or click in outward rotation, and (3) a dislocation in which the humeral head has slipped over the posterior edge of the glenoid.

MRI was used to determine the degree of remodeling of the operated shoulder joints (Fig. 1). All images were obtained with the subjects supine and with both arms at their sides. For this investigation, all relevant imaging was scanned into our hospital image viewing system (Sectra IDS7; Sectra, Linköping, Sweden). A senior upper-extremity radiologist (M.W.) evaluated all examination findings preoperatively and postoperatively.

Axial images were used for the measurements. A representative slice through the center of the glenoid was chosen by the radiologist. By use of the radiologic tools available in IDS7, a line was drawn through the long axis of the scapula, from the medial margin to the midpoint of the glenoid, which was then extended laterally. To calculate the percentage of the humeral head anterior to the scapula (ie, PHHA), the maximal diameter of the humeral head was measured, as was the distance from the long-axis scapular line to the anterior aspect of the humeral head. To calculate glenoid version, a second line of neutral version was drawn perpendicular to the long axis and version was calculated from this to a line connecting the anterior and posterior cartilaginous labral surfaces. The method is well established and has shown good intrarater and inter-rater reliability.[7,19,26]

When possible, the preoperative imaging was obtained with the patient under anesthesia directly prior to surgery, but some of the older patients were examined at their local hospital and the images were transferred to our unit ahead of surgery. At the beginning of our series, it was difficult to obtain the scans perioperatively, and as the indication for surgery was clinical and the radiology did not affect the decision to operate or the technique used, some patients lacked preoperative radiology. We now systematically perform preoperative MRI with the patient under anesthesia directly prior to the surgical procedure to document incongruence radiographically. The patient is then transferred from the MRI camera to the operating theater. Long-term follow-up MRI was obtained in conjunction with the clinical follow-up previously reported.[13]

Statistical analysis

All patient data were recorded in SPSS Statistics software (version 23; IBM, Armonk, NY, USA). Population data are presented as means with ranges. The normality assumption was tested using the Shapiro-Wilk test. We used the dependent t test to compare measurements before and after surgery. Analysis of variance (ANOVA) was used to test for significant differences between the different age and joint categories. To compare differences between sexes and the groups with and without preoperative radiology, we used the independent t test. $P \leq .05$ was considered significant.

Results

The mean follow-up time was 10.2 years (range, 7-16 years), and 40 female and 21 male patients were included (Fig. 2). The mean age at operation was 3.2 years (range, 8 months to 15 years). Of the patients, 20 were younger than 2 years, 33 were aged 2 to 5 years, and 8 were older than 5 years (range, 6-15 years) at the time of surgery. In this series, 14 patients had a flattened glenoid, 41 had a bifaceted glenoid, and 6 had a dislocated joint at the time of surgery. Six patients underwent latissimus dorsi transfer. Of the patients, 59 had Erb-type palsy and 2 had a C5-T1 injury. Five patients underwent nerve reconstruction at an early age. The average gain in external rotation for this group of patients was 45° to 60° at long-term follow-up; the

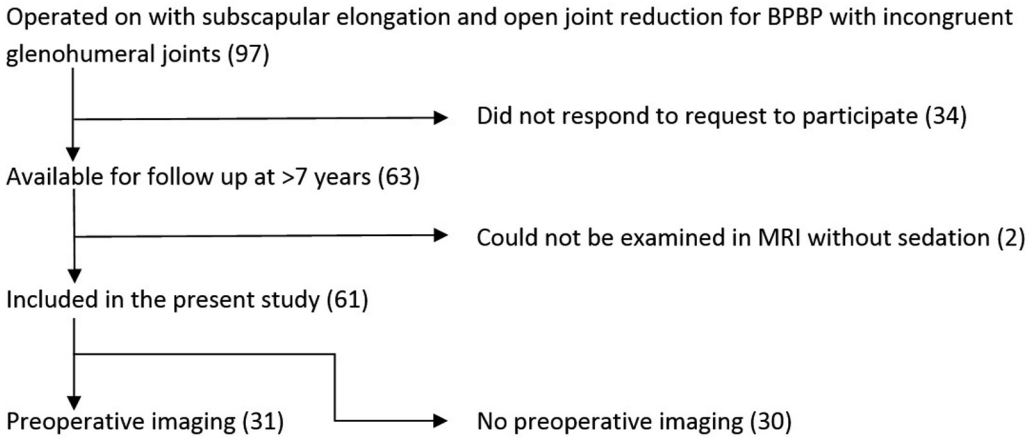

Figure 2 Flowchart of patient inclusion. *BPBP*, brachial plexus birth palsy; *MRI*, magnetic resonance imaging.

average loss of internal rotation was around 22°. The average 3-grade Mallet score increased by 3.1 units. Detailed information on clinical outcomes in the short and long term are available in earlier publications.[12,13]

All patients showed positive remodeling between preoperative and postoperative evaluations as measured by our 3 chosen outcomes. On average, the increase in PHHA was 27.6% (95% confidence interval [CI], 22.4%-32.7%; $P < .01$). The average change in glenoid retroversion was 14.8° (95% CI, 11.1°-18.4°; $P < .01$). The average increase in diameter was 16.5 mm (95% CI, 14.0-19.1 mm; $P < .01$).

Figures 3 and 4 show the average PHHA preoperatively and postoperatively in the age and joint category subgroups. The increase in PHHA postoperatively for the youngest age group (aged < 2 years) was 27.1% (95% CI, 16.5%-37.8%; $P < .01$); for patients in the middle age group (aged 2-5 years), 25.8% (95% CI, 17.9%-33.7%; $P < .01$); and for patients in the oldest age group (aged > 5 years), 29.5% (95% CI, 9.1%-49.9%; $P = .019$). The increase in PHHA postoperatively for patients with a flattened glenoid was 19.0% (95% CI, 10.1%-28.0%; $P = .01$); for patients with a bifaceted glenoid, 27.4% (95% CI, 21.0%-33.8%; $P < .01$); and for patients with a dislocated joint, a nonsignificant increase of 49.0% (95% CI, −81.5% to 179.6%; $P = .132$).

Figures 5 and 6 show the average glenoid angle preoperatively and postoperatively in the age and joint category subgroups. The change in glenoid angle postoperatively for the youngest age group (aged < 2 years) was 12.8° (95% CI, 4.6°-21.0°; $P = .007$); for patients in the middle age group (aged 2-5 years), 16.8° (95% CI, 11.9°-21.8°; $P < .01$); and for patients in the oldest age group (aged > 5 years), a nonsignificant increase of 6.7° (95% CI, −13.5° to 27.0°; $P = .367$). The change in glenoid angle preoperatively vs. postoperatively for patients with a flattened glenoid was 12.7° (95% CI, 7.2°-18.3°; $P = .001$); for patients with a bifaceted glenoid,

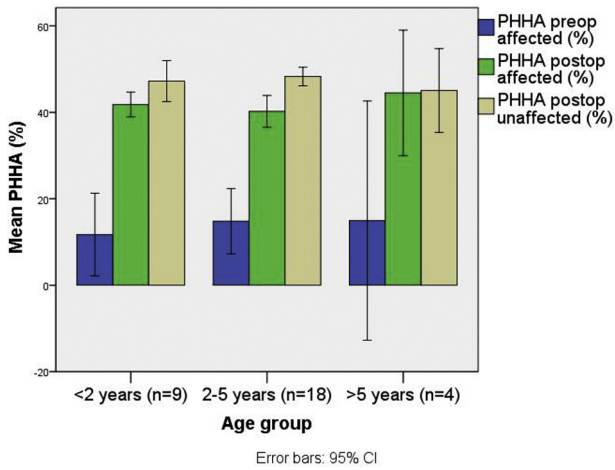

Figure 3 Bar chart showing average percentage of humeral head anterior to midscapular line (*PHHA*) in different age groups preoperatively (*preop*) for affected side and at mean 10-year follow-up for both affected and unaffected sides. *CI*, confidence interval.

13.1° (95% CI, 8.2°-18.1°; $P < .01$); and for patients with a dislocated joint, a nonsignificant change of 33.2° (95% CI, −27.1° to 93.6°; $P = .090$).

No significant difference in the increase in PHHA was found between age groups (27.7%-30.7%; $P = .804$, ANOVA), nor was the change in glenoid angle significantly different between age groups (0.8°-16.4°; $P = .051$, ANOVA). We found no significant differences between age groups (39.6%-41.1%; $P = .849$, ANOVA) or between joint categories (40.2%-43.9%; $P = .371$, ANOVA) in achieved PHHA for affected shoulders at long-term follow-up. We also found no significant differences between age groups (9.3°-16.4°; $P = .075$, ANOVA) or between joint categories (6.0°-11.4°; $P = .225$, ANOVA) in achieved glenoid angle for affected shoulders at long-term follow-up (ie, all groups benefitted equally from surgery).

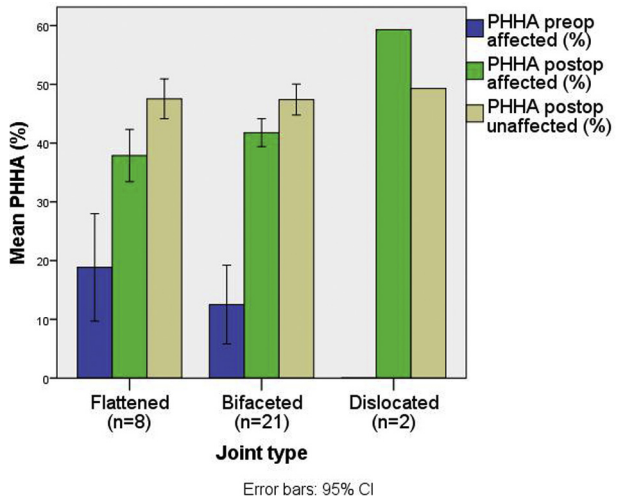

Figure 4 Bar chart showing average percentage of humeral head anterior to midscapular line (*PHHA*) for different joint types preoperatively (*preop*) for affected side and at mean 10-year follow-up for both affected and unaffected sides. *CI*, confidence interval.

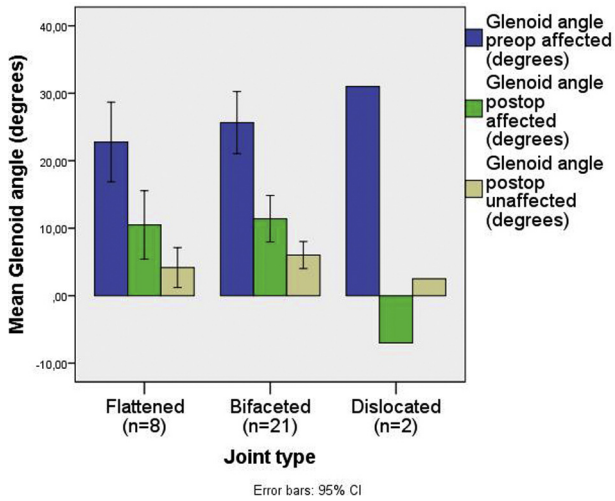

Figure 6 Bar chart showing average glenoid angle for different joint types preoperatively (*preop*) for affected side and at mean 10-year follow-up for both affected and unaffected sides. *CI*, confidence interval.

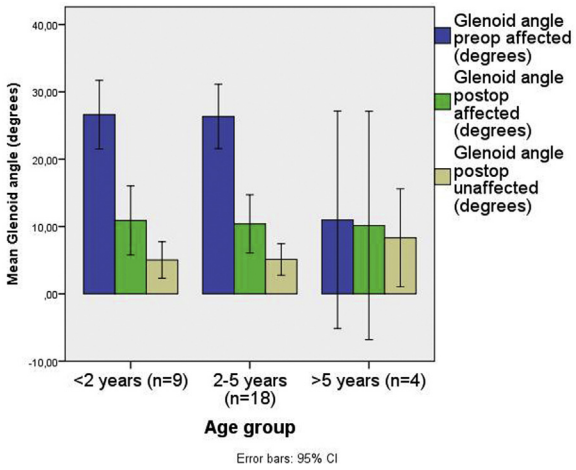

Figure 5 Bar chart showing average glenoid angle in different age groups preoperatively (*preop*) for affected side and at mean 10-year follow-up for both affected and unaffected sides. *CI*, confidence interval.

Figures 7-10 show the average differences between the operated and unaffected shoulders in the age and joint category subgroups. We found very small differences between the sides. The average difference in PHHA was 6.7% (95% CI, 5.0%-8.4%; $P < .01$). The average difference in glenoid angle was 5.3° (95% CI, 3.4°-7.2°; $P < .01$). The average difference in size was nonsignificant, at 0.8 mm (95% CI, −0.2 to 1.9 mm; $P = .116$).

No significant differences in PHHA (95% CI, −2.8% to 3.3%; $P = .885$), glenoid angle (95% CI, −2.3° to 5.4°; $P = .431$), or diameter (95% CI, −2.6 to 1.3 mm; $P = .531$) of the affected shoulders at long-term follow-up were found between patients with and patients without preoperative radiology. In addition, no significant differences in PHHA (95% CI, −4.8% to 1.7%; $P = .340$), glenoid angle (95% CI, −5.4° to 3.0°; $P = .558$), or diameter (95% CI, −0.6 to 3.5 mm; $P = .171$) of the affected shoulders at long-term follow-up were noted between male and female patients.

Discussion

The radiologic evaluation confirmed that subscapular elongation and open joint reduction allow for a very high degree of positive remodeling of incongruent shoulder joints toward normal. The results were consistently good in children who were operated on up to the age of 5 years. The differences in PHHA and glenoid version between the operated and unaffected shoulders were small, with values of the affected side approaching those of the unaffected side. The results indicated that it is also possible to achieve useful remodeling in children older than 5 years (the oldest patient in this material was aged 15 years at the time of operation).

This study showed a consistent improvement in joint configuration over time, after relocation. This is in line with the findings by Waters and Bae that open repositioning improves remodeling[28] compared with tendon transfer alone.[30] The relatively lower incidence of dysplasia in the cohort that provided patients for the present study compared with others[10,22,31] may be accounted for by differences in categorization or in referral patterns between centers.

The method of categorizing the joint via direct inspection will not detect a small degree of retroversion of

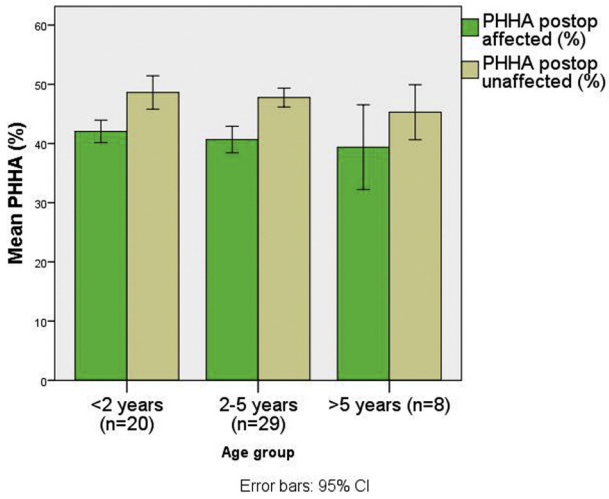

Figure 7 Bar chart showing average difference in percentage of humeral head anterior to midscapular line (*PHHA*) in different age groups between operated and unaffected joints at mean 10-years follow-up. *CI*, confidence interval.

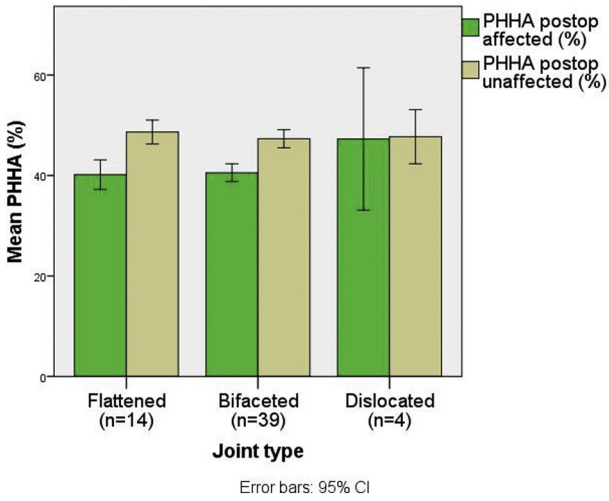

Figure 8 Bar chart showing average difference in percentage of humeral head anterior to midscapular line (*PHHA*) for different joint types between operated and unaffected joints at mean 10-years follow-up. *CI*, confidence interval.

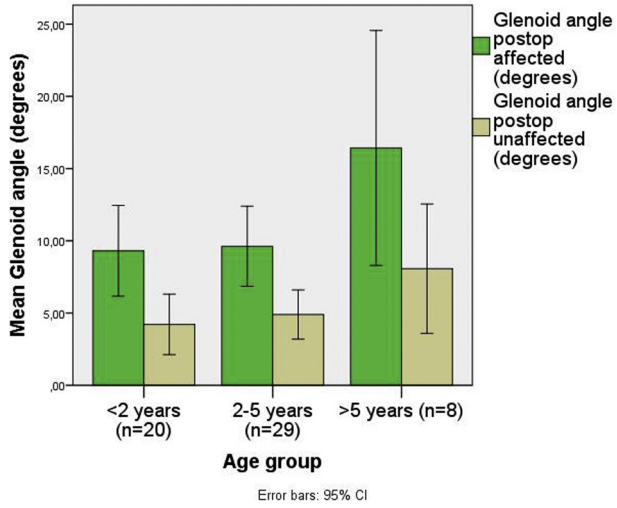

Figure 9 Bar chart showing average difference in glenoid angle in different age groups between operated and unaffected joints at mean 10-years follow-up. *CI*, confidence interval.

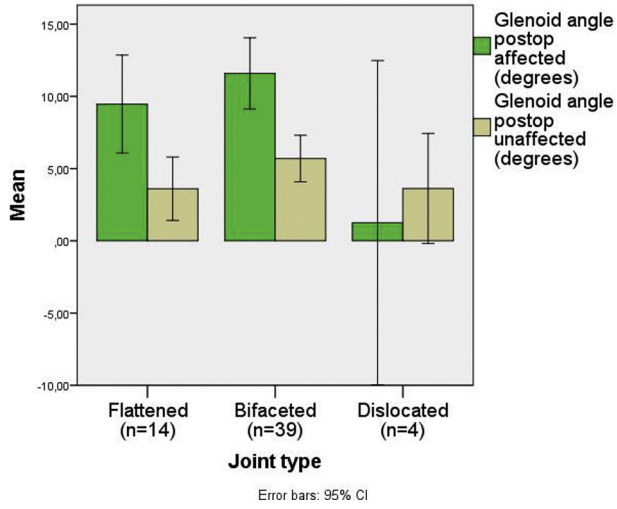

Figure 10 Bar chart showing average difference in glenoid angle for different joint types between operated and unaffected joints at mean 10-years follow-up. *CI*, confidence interval.

the glenoid, but it has other benefits over the radiologic categorizations. Instability in younger children may be dynamic,[27] and open inspection not only provides a direct view of the joint surfaces but also allows for dynamic examination of the joint's behavior between its position in internal rotation and its position in outward rotation. The dynamic examination reveals whether the humeral head slides forward during outward rotation in an abnormally flat glenoid or whether the glenoid is divided into 2 separate facets. Visualization of the joint at surgery also allows the surgeon to confirm that the joint head is seated in the anatomic (anterior) facet of the glenoid. We have noted that there is sometimes a discrepancy between the preoperative MRI scan and what is seen during surgery. The static view provided on MRI may thus give false categorizations of joint incongruity. Therefore, even though MRI is valuable for providing measurements that can be documented, compared, and evaluated over time, we prefer the dynamic categorization of joint types that is possible during surgery.

Measuring the glenoid angle is a well-documented method,[7,19,26] but it has some limitations. Most of our patients had a bifaceted glenoid, making accurate measurements more difficult. In this study, we chose to always use the labra most anterior and posterior, rather than measuring the individual facets. One could argue that

measuring across the entire glenoid underestimates the degree of angulation compared with measuring only the posterior facet, which in fact supports the head. In a convex glenoid, it becomes very difficult to ascertain where to measure the glenoid angle. We find that it is easier to identify consistent reference points when calculating PHHA, and we believe that PHHA is a more robust measure.

Among patients operated on early in the series, a large proportion lacked preoperative radiology. However, the indications and the method of operation remained unchanged throughout the series. No significant differences in PHHA, glenoid version, or diameter were found between the group that underwent preoperative radiology and the group that did not. Thus, we consider the group without preoperative radiology to be representative of the whole material. The substantially longer follow-up time and comparatively large group of patients examined, compared with previous publications, are the main strengths of this study.

There is a widespread opinion that the incongruence of the glenohumeral joint in BPBP develops gradually over time, as a consequence of an internal rotation contracture and muscular imbalance, and that there is increasing posterior displacement of the humeral head with subluxation, then development of a bifaceted glenoid, and finally, in some cases, a complete posterior dislocation.[6,8,10,14,27,28,30,31] We are not convinced that the posterior displacement develops gradually in most cases. We do not find support for a general correlation between age and severity of the joint deformity as previously described,[22] and indeed, the majority of children with joint incongruence who we identify today receive their diagnoses at a very young age. We sometimes see complete dislocation in children as young as 6 months. It could well be that severe instability and dislocation sometimes occur very early after birth due to the nerve injury with paralysis of the cuff muscles, possibly in combination with a distension of the joint capsule or ligaments, both caused by birth trauma. As the cuff muscles recover active function, they introduce forces that can lead to deformation of the surfaces of an already displaced and unstable joint—with development of a bifaceted glenoid in many cases, whereas in other patients, a milder deformity develops with flattening of the posterior glenoid, and in some, complete dislocation remains.

Why is it important to relocate the incongruent glenohumeral joints and to strive for optimal positive remodeling? There can be no doubt that a joint needs to maintain its normal configuration as far as possible to function properly. A dorsal displacement of the humeral head disturbs the muscular balance of the rotator cuff, providing increased tension in the subscapularis and a slackening of the infraspinatus. This probably contributes strongly to the contracture in internal rotation that is so typical in BPBP. When we examined a subgroup of 8 patients in whom relocation had failed, they had less benefit from the operation.[12] In our experience, it is impossible to rebalance the joint unless the head can be relocated and lined up correctly on the long axis of the scapula.

Conclusion

The findings of our study confirm that although the glenohumeral joint can develop severe incongruence and deformity very quickly in young children with BPBP, the joint also has an impressive and consistent potential for remodeling after relocation, certainly up to the age of 5 years. Our findings suggest that there is potential for successful relocation with useful remodeling even in adolescents, challenging the notion that derotational osteotomies are the treatment of choice in older children.[3,27,29]

Disclaimer

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