Glenohumeral Deformity in Children with Brachial Plexus Birth Injuries
Early and Late Management Strategies

Abstract
Shoulder deformity remains the most common musculoskeletal sequela following a brachial plexus birth injury. The natural history of untreated glenohumeral deformity is one of progression in this unique patient population. In infants and young children with persistent neurological deficits, shoulder dysfunction becomes a major source of morbidity, as these children have extreme difficulty placing the hand in space. The functional limitations due to muscle denervation and the resultant periarticular soft tissue contractures and progressive osseous deformities have been well-characterized. Increasing attention is being given to the glenohumeral dysplasia (GHD) and the associated prevalence of early posterior dislocation of the shoulder in infants with brachial plexus birth injuries. GHD represents a spectrum of findings, including glenoid and humeral head articular incongruities and dysplasia, subluxation, and frank dislocation. This article presents our comprehensive, temporally-based management strategies for the glenohumeral joint deformities in these children utilizing soft tissue and bony reconstructive procedures.

In infants with brachial plexus injuries, shoulder dysfunction represents a major source of disability. Glenohumeral deformity occurs early and is progressive in children who do not demonstrate adequate spontaneous recovery. Initial deformity is secondary to the muscle imbalances that develop about the shoulder, or primary bony trauma to the shoulder. In children with persistent neurological deficits, weakness of abduction, forward elevation, and external rotation is due to incomplete recovery of the axillary and suprascapular nerves. In C5/C6 or upper trunk lesions, the medial rotators of the shoulder are relatively “spared,” and often there are associated internal rotation myostatic contractures of the subscapularis and pectoralis major. Additionally, significant periarticular capsuloligamentous tightness may develop, necessitating releases. Ultimately, if left untreated, these soft-tissue imbalances about the glenohumeral articulation result in secondary subluxation or dislocation and even a fixed articular deformity of both the glenoid and humeral head and overgrowth of the coracoid. Alternatively, osseous deformity may be due to a primary and direct trauma to the proximal humerus physis at the time that the neurologic injury is sustained. Goddard has divided direct shoulder trauma sustained at birth into simple (distortion of the humeral head), complicated (proximal humerus epiphyseal separation, such as epiphysseolysis), and clavicular trauma. Further, Dunkerton reported on the occurrence of proximal humeral metaphyseal fractures and posterior dislocation of the shoulder at birth in association with brachial plexus lesions. These soft tissue and osseous changes in the normal anatomy of the glenohumeral joint also result in abnormal...
scapulothoracic mechanics.\textsuperscript{6}

In a longitudinal study of 25 patients, Pearl and Edgerton\textsuperscript{1} observed that approximately 70% of children who present with an internal rotation contracture will have concomitant glenohumeral deformity. Therefore, in the setting of persistent neurologic deficits and muscular imbalances about the shoulder, it is clear that glenohumeral dysplasia and possible joint instability appears very early, progresses with age, and usually correlates with the magnitude of internal rotation contracture seen in these children.

**Evaluation and Classification of Glenohumeral Dysplasia**

While much research has focused on developmental dysplasia of the hip in the pediatric orthopaedic literature, increasing attention is now being given to the glenohumeral dysplasia (GHD) and the associated prevalence of early posterior dislocation of the shoulder in infants with brachial plexus birth injuries. Glenohumeral dysplasia, too, represents a spectrum of findings, including glenoid and humeral head articular incongruities and dysplasia, subluxation, and frank dislocation. Several modalities have been used to evaluate the magnitude of glenoid hypoplasia, humeral head deformity, coracoid overgrowth (“hooking”), and articular incongruity in these children.

Ultrasound has been used in select centers to assess the location of the ossific nucleus of the proximal humerus in these infants. Further, it allows for the dynamic assessment of the glenohumeral joint, and thus the reducibility of the subluxation-dislocation may be studied. In a study of 134 consecutive infants with brachial plexus birth injuries treated at Texas Scottish Rite Hospital, 8% had ultrasonographic evidence of early posterior shoulder dislocation.\textsuperscript{9} Children with early subluxation or dislocation of the humeral head demonstrated asymmetric axillary or proximal arm skin folds, apparent shortening of the humeral segment, palpable fullness in the posterior shoulder, and palpable “clicks” during passive glenohumeral manipulation; and these represent similar findings to those seen in the infant with hip dysplasia. The critical role of serial, consistent examinations of the glenohumeral and scapulothoracic joints in the neonatal period cannot be over-emphasized. The rapid loss of passive external rotation between monthly examinations is indicative of progressive capsular and muscular contracture, and thus the onset of subluxation or dislocation.

Characterization of glenohumeral deformities secondary to brachial plexus birth palsy with plain radiography is not possible because the glenohumeral joint does not completely ossify until puberty. Most institutions utilize MRI scans to assess the cartilaginous surfaces of the glenohumeral joint in younger children and reserve CT imaging for older children. Classification of the magnitude of glenohumeral deformity is essential in the treatment algorithm.\textsuperscript{2} In a prospective study of 94 patients with brachial plexus birth injuries, the association between persistent palsy, age-related musculo-skeletal deformity, and functional limitations were studied. Seven radiographic glenohumeral subtypes, as measured by axial CT or MRI images, were delineated based upon the magnitude of glenoid retroversion and the percentage of humeral head subluxation relative to a bisecting line drawn through the scapular spine. As the severity of the dysplasia increases, a “pseudoglenoid” may be seen along with the progressive subluxation (i.e., type IV deformity). Type VI deformity represents infantile posterior dislocation, and type VII is characterized by proximal humeral physeal growth arrest.

**A Longitudinal Treatment Strategy**

In cases of soft tissue or bony glenohumeral deformity with loss of passive or active shoulder motion, treatment options available depend upon time of recognition and age at diagnosis (“early” versus “late”), the magnitude of GHD, and the reducibility of the subluxation or dislocation if present. The goals of treatment always include restoration of full passive range of motion, maintenance of a concentric, stable glenohumeral articulation, and muscle rebalancing in order to facilitate glenoid remodeling.

**Age 0-24 Months, Early Intervention**

The variable patterns of nerve injury, and thus the potential for neurological improvement in the majority of infants born with a brachial plexus birth injury, are the impetus behind the initial period of observation in the immediate postnatal period.\textsuperscript{10} It is essential during this period of observation and potential early neurological recovery that the glenohumeral and scapulothoracic joint be specifically evaluated as part of the upper extremity examination during serial (i.e., monthly) examinations. Goals during this time period include obtaining full passive glenohumeral motion, prevention of myostatic and periarticular contractures, and maintaining a concentrically reduced humeral head within the glenoid cavity. A multidisciplinary team that includes specialized therapists is crucial in optimizing shoulder outcome in this early phase.\textsuperscript{11} The glenohumeral joint must be isolated by stabilizing the scapulothoracic articulation and working to increase external rotation with the arm adducted at the side and the elbow flexed. Because of the complexity of the biomechanics of the shoulder, maintaining full external rotation in abduction only will not prevent contracture of the subscapularis muscle.\textsuperscript{12}

A small percentage of infants will present with a traumatic true congenital glenohumeral dislocation sustained in conjunction with the initial nerve injury, and this clinical entity manifests as an early, fixed medial rotation contracture of the shoulder.\textsuperscript{13} However, the majority of infants will develop gradual glenohumeral deformity and subluxation or dislocation as indicated by the progressive loss of passive external rotation on serial examinations.\textsuperscript{9,14}

In addition to physical therapy, abduction and external rotation splints and spica casts may be used to improve and
maintain the passive arc of motion. Botulinum toxin type A injections are also being increasingly used in our practice (JAIG) at this stage preceding microneurosurgical intervention to prevent contracture and maintain full passive range of motion by balancing the shoulder. Functional electrical stimulation may also be added. At present, no Level I evidence exists to support formal guidelines for indications of these interventions nor data on expected outcomes.

**Established Periarticular Contracture and Deformity**

Early, persistent, or progressive medial rotation contracture and glenohumeral deformity is always addressed at the time of primary plexus microneurosurgical reconstruction and suprascapular nerve reinnervation procedures. Primary focus is placed on suprascapular nerve reinnervation to restore shoulder elevation and external rotation. Direct distal spinal accessory nerve (SAN) transfer into the suprascapular nerve outside the zone of injury represents our present reconstructive strategy of choice for reinnervation of the supraspinatus and infraspinatus in order to promote shoulder stability and restore active external rotation. The SAN is an extraplexal pure motor nerve that lies in close proximity to the upper trunk and suprascapular nerve allowing for potential direct coaptation without the need for an interposition nerve graft. While a direct end-to-end repair between the spinal accessory and suprascapular nerve is preferred, the use of a short, interposition graft yields results comparable to outcomes reported following partial accessory nerve transfer into the suprascapular nerve using a proximal end-to-side technique with cervical plexus or reversed sural nerve grafts.

Following the microneurosurgical reconstruction, attention is turned to the shoulder. Priority is placed on achieving full passive lateral rotation followed by concentric reduction of the glenohumeral joint. In 1918, Sever first described the release of the pectoralis major and subscapularis to address the medial rotation contracture of the shoulder. Simultaneous release of a shoulder medial rotation contracture is indicated at the time of primary microsurgical intervention or tendon transfers when full passive external rotation of the shoulder cannot be obtained with the arm adducted.

We have adopted an “a la carte” approach to the management of the medial rotation contracture in all children with brachial plexus birth injuries. Following examination under anesthesia, the magnitude of the contracture is established. If near-full passive external rotation is appreciated, botulinum toxin A is injected into the pectoralis major and latissimus dorsi-teres major complex. Severe medial rotation contracture necessitates intramuscular lengthening of the pectoralis major, release of the subscapularis, or both. When longstanding subluxation or dislocation is present, partial coracoid resection, with or without release of the coracohumeral ligament, and coracobrachialis fractional lengthening may be necessary to obtain concentric glenohumeral reduction.

Subscapularis lengthening for infants who fail to regain full passive external rotation of the shoulder may be performed by a variety of methods (arthroscopically, such as by release of the insertion; anterior Z-lengthening; musculotendinous lengthening; or subperiosteal release of the origin off the anterior face of the scapula [subscapularis slide]). Advocates of arthroscopic capsular and subscapularis releases in these children with medial rotation contractures cite the direct visualization of the articular deformity as a major advantage of this technique. Several disadvantages and complications with this approach have been noted: 1. excessive loss of internal rotation strength with subscapularis insertional release may necessitate internal rotation humeral osteotomy; 2. the proximity of the axillary nerve puts it at risk for injury; and 3. anterior instability may be created with release of the associated glenohumeral ligaments.

Our group’s preference is for an open “subscapularis slide” in these children (Fig. 1), and this technique represents a refinement of that initially presented by Carlioz and Brahimi and popularized by Gilbert. The incision is based over the lateral border of the scapula and may be extended posteriorly if concomitant latissimus dorsi-teres major transfer is contemplated. The raphe between the subscapularis and teres major is identified and the neurovascular pedicles are protected. Subperiosteal release of the subscapularis from its origin on the anterior surface of the scapula is performed. A fasciotomy along the deep investing fascia of the subscapularis may add external rotation if lateral rotation is not full following the subperiosteal slide. With this technique, the anterior stabilizing function of the subscapularis and glenohumeral ligaments is preserved.

Botulinum toxin type A inhibits the release of acetylcholine vesicles into the neuromuscular junction by cleaving the SNARE proteins that facilitate the binding of the vesicle membranes to the nerve terminal membrane. There is data to support consideration of botulinum toxin type A injections into the medial rotation muscle groups about the shoulder (i.e., subscapularis major, pectoralis major, and the latissimus dorsi-teres major complex) at the time of the primary plexus and shoulder reconstructions. Use of botulinum toxin has been associated with early and sustained improvements in shoulder outcomes at a mean of 3 years postoperatively following open subscapularis slide with concomitant external rotation tendon transfers. Botulinum toxin A appears to prevent the tendency to develop an exaggerated medial rotation posture early after upper plexus neurolysis and reconstruction. It is hypothesized that by temporarily weakening the medial rotators through its action on α and γ motor endings, botulinum toxin type A reduces their afferent input to the motor cortex and thereby allows cortical recruitment of injured nerves to increase.

Success with addressing the shoulder at the time of primary nerve surgery has been previously reported. Grossman and colleagues reported an improvement of two grades or more on the modified Gilbert shoulder scale in 22 infants.
who underwent “late” combined plexus and shoulder reconstructions at a mean age of 16 months. Specifically, all children in this study underwent microsurgical neurolysis of the upper plexus and bypass nerve grafting from C5 or C6 selectively into the upper-trunk divisions and the suprascapular nerve, and four underwent transfer of the spinal accessory nerve into the suprascapular nerve with a short interpositional graft. All underwent concomitant release of the associated shoulder contracture by subscapularis slide, and 11 with persistent posterior instability after subscapularis slide underwent simultaneous open reduction and posterior capsulorrhaphy. Additionally, all received botulinum toxin type A into the pectoralis major and latissimus-teres major complex. Further, in a recent cohort of 34 patients undergoing primary plexus reconstruction including distal direct spinal accessory nerve to suprascapular nerve, 14 of 34 (41%) of infants underwent simultaneous reconstruction of a shoulder medial rotation contracture. At a minimum of 2-year follow-up, overall mean active external rotation measured approximately 70°, mean Gilbert score was 4.1, and the mean Miami score was 7.1, corresponding to an overall good functional shoulder outcomes on each scale (Tables 1 and 2).

Therefore, once passive lateral rotation is obtained, glenohumeral reduction usually ensues. When the subluxation-dislocation has existed for an extended interval, it may be necessary to address the secondary changes preventing concentric reduction. Open posterior capsulorrhaphy is performed in cases of unstable reductions. Simultaneous anterior procedures and posterior capsulorraphy are used to confer stability to the reduced humeral head.

The priority placed upon obtaining and maintaining concentric glenohumeral reduction in these children is derived from the demonstrated potential for glenoid remodeling as demonstrated by Hui and Torode. A mean 30° of improvement in the magnitude of glenoid retroversion in the first year postoperatively was observed following open reduction and release of the associated internal rotation contracture. Further, glenoid version improved at an overall rate of 9% per year. Pedowitz and coworkers have reported MRI evidence of significant improvements in the magnitude of humeral head subluxation and mean glenoid version following arthroscopic anterior capsular release, subscapularis tenotomy, and glenohumeral reduction in 22 children. It is important to note that unlike the acetabulum, there is no published data that speaks to the upper age limit at which spontaneous improvement in the degree of glenoid retroversion may be seen.

**Age 2 Years to 5 Years**

This age groups represents a heterogeneous group and may include children with or without prior primary plexus reconstructions at a mean age of 16 months. Specifically, all children in this study underwent microsurgical neurolysis of the upper plexus and bypass nerve grafting from C5 or C6 selectively into the upper-trunk divisions and the suprascapular nerve, and four underwent transfer of the spinal accessory nerve into the suprascapular nerve with a short interpositional graft. All underwent concomitant release of the associated shoulder contracture by subscapularis slide, and 11 with persistent posterior instability after subscapularis slide underwent simultaneous open reduction and posterior capsulorrhaphy. Additionally, all received botulinum toxin type A into the pectoralis major and latissimus-teres major complex. Further, in a recent cohort of 34 patients undergoing primary plexus reconstruction including distal direct spinal accessory nerve to suprascapular nerve, 14 of 34 (41%) of infants underwent simultaneous reconstruction of a shoulder medial rotation contracture. At a minimum of 2-year follow-up, overall mean active external rotation measured approximately 70°, mean Gilbert score was 4.1, and the mean Miami score was 7.1, corresponding to an overall good functional shoulder outcomes on each scale (Tables 1 and 2).

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surgery, those with recurrent or progressive medial rotation contractures, new or recurrent glenohumeral subluxation-dislocation, or those with significant glenoid dysplasia. At this age (i.e., after 24 to 30 months), these children are not candidates for primary microneurosurgical reinnervation. Treatment in these children is based on rotator cuff and periscapular muscle strength, the magnitude of medial rotation contracture, and status of the glenohumeral joint. The medial rotation contracture is addressed using the step-wise progressive “a la carte” approach described above, and glenohumeral reduction is achieved by closed or open means. Additionally, glenoid osteotomy and extra-articular tendon transfer to restore active external rotation are considerations in these children.

If we extrapolate our knowledge of hip dysplasia and the associated efficacy of pelvis and periacetabular osteotomy to this problem of GHD in these patients, the question becomes should we be considering the plausibility of glenoid osteotomy? The use of glenoid osteotomy has not been explicitly reported in the literature. Based on the senior author’s (AEP) experience with two children, corrective posterior glenoid osteotomy performed with a bone block has only been rarely required (Fig. 2). Specifically, in two cases, the indications have been recurrent posterior instability secondary to severe glenoid retroversion and the presence of biconcave “pseudoglenoid.” Based on the limited experience with this procedure, the optimal timing at which to perform this procedure is not known. Enthusiasm for glenoid osteotomy may grow in select cases in which the magnitude of glenoid remodeling potential is limited with soft tissue procedures alone.

Loss of active external rotation is manifested by functional limitations as the child is unable to effectively place the hand in space. External rotation of the proximal humerus is necessary in activities occurring above the horizontal as this motion allows the greater tuberosity to clear the subacromial arch, and the biceps assumes a position to act as a forward elevator of the upper limb. Further, scapulothoracic winging cannot compensate for weak external rotation.

In 1934, L’Episcopo first described latissimus dorsi transfer performed in conjunction with medial rotation contracture release in order to restore active external rotation. This approach has yielded satisfactory results at centers with large experiences in children with minimal glenohumeral dysplasia/deformity, a concentrically reduced shoulder, and residual active external rotation weakness. Hoffer and Phipps refined this technique by transferring the latissimus dorsi tendon as lateral and anterior on the infraspinatus insertion in order to gain the biomechanical advantage of elevation as well as external rotation with a single transfer. We also advocate concomitant latissimus dorsi-teres major transfer to achieve muscle re-balancing to help to minimize recurrent subluxation or dislocation and to facilitate glenoid remodeling (Fig. 3). It useful to characterize the degree of axillary nerve-teres minor recovery in the decision-making process of whether to transfer the latissimus dorsi tendon as lateral and anterior on the infraspinatus insertion in order to gain the biomechanical advantage of elevation as well as external rotation with a single transfer.

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between the long head of the triceps and posterior deltoid. The tendon(s) is then anchored into the posterior aspect of the greater tuberosity as high up on the insertion of the infraspinatus as possible, similar to the approach described by Hoffer and associates.\(^{42}\) Extra-articular tendon transfers to restore external rotation strength yield suboptimal results.

**Figure 2.**

A. Preoperative MRI demonstrates glenohumeral dysplasia with posterior subluxation of the humeral head. B. Axial CT image demonstrates incorporation of the bone-block graft following glenoid osteotomy for severe glenoid dysplasia and retroversion that led to posterior instability of the shoulder.

**Figure 3.**

A. Following incision and dissection over the lateral border of the scapula, regional axillary adhesions are released. B. In this case, the latissimus and teres major were isolated and transferred as a single conjoined tendon. C. The tendons are re-routed together posterior to the long head of the triceps and under the posterior deltoid. D. They are anchored into the posterior aspect of the greater tuberosity at the insertion of the infraspinatus, similar to the approach of Hoffer and colleagues.\(^{42}\)
in the child with advanced glenohumeral deformity, and the underlying incongruities continue to restrict abduction and external rotation.

**The Older Child and Salvage Options**

Humeral external derotational osteotomy represents a salvage option in the older child with advanced GHD and long-standing medial rotation contracture of the shoulder. When indicated, derotational humeral osteotomy improves the position and function of the upper limb. In a recent study of 27 patients who underwent external rotation humeral osteotomy for medial rotation contracture and external rotation weakness in the setting of advanced glenohumeral deformity, mean modified Mallet scores improved from 13 preoperatively to 18 at a mean follow-up of 3.7 years, and external rotation averaged 64°. These improvements in global shoulder function were seen without any significant changes in glenoid morphology. When mild glenohumeral deformity is present with a profound loss of shoulder strength, we have added a latissimus dorsi to infraspinatus tendon transfer to gain elevation and external rotation at the time of derotational osteotomy.

It is the status of the glenohumeral joint rather than the age of the patient that guides the selection of the procedure performed. As discussed earlier, unlike the acetabulum, there is no published data that speaks to the upper age limit at which spontaneous improvement in the degree of glenoid retroversion may be seen. Derotational osteotomy of the humerus may also be used in the younger child (i.e., greater than age 4 years) with mild-to-moderate GHD and persistent external rotation weakness following previous soft-tissue procedures.

A deltopectoral approach is utilized, followed by subperiosteal exposure of the proximal humerus. A transverse osteotomy is performed between the deltoid and pectoralis major insertions. The proximal holes are then drilled. Intraoperatively, the humeral cortex can be scored proximal and distal to the plate or parallel k-wires can be used as guides to assess the magnitude of external rotation achieved once the osteotomy is completed. The osteotomy is then fixed with size-appropriate internal fixation in a standard fashion. In most cases, 45° to 60° of external rotation is desirable, with size-appropriate internal fixation in a standard fashion. To confirm that over-rotation prohibiting the child from reaching the midline and perineum has not occurred. This osteotomy does not alter overall glenohumeral motion but rather serves to improve upper limb function by reorienting the arc of shoulder motion into a more functional range.

**Summary**

A multidisciplinary approach is crucial to optimizing the general overall outcome of these children, and in particular shoulder outcome. Our model consists of a team comprised of specialists in child neurology, neurophysiology, and occupational therapy who serially evaluate these infants. The collective combined expertise of these specialists is used in collaboration with the treating orthopaedic and peripheral nerve and hand surgeons for decision making in the immediate perioperative period and during long-term follow-up. It must be stressed that despite evidence-based guidelines for the treatment of the shoulder pathology in these children, there is no “cookie-cutter” approach for the management of the musculoskeletal sequelae that these children face. Long-term, prospective data is still needed to ascertain whether “early” shoulder microneurosurgical innervation yields superior results to “delayed” glenohumeral reconstructions.

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