Outcomes of Surgical Treatment for Secondary Shoulder Contractures and Deformities in Brachial Plexus Birth Palsy: A Systematic Review

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Brachial plexus birth palsy can be a severely disabling condition for an otherwise healthy infant. Activities of daily living, such as feeding and grooming, are restricted by the internal rotation and adduction contractures that develop from muscle imbalance about the shoulder. A spectrum of orthopaedic procedures, from contracture releases to humeral derotational osteotomies, are performed to improve range of motion and enhance the functional ability of the extremity. The Mallet classification has been used widely in the literature to evaluate improvements in shoulder motion in patients with brachial plexus birth palsy. This study is a systematic review of the literature regarding surgical treatment of shoulder contractures and deformities with soft tissue procedures and humeral osteotomies. Both techniques have shown consistent improvements in motion according to the Mallet classification. However, these gains still do not provide a normally functioning shoulder and may diminish slightly over time.

Introduction

Brachial plexus birth palsy (BPBP) is a traction or compression injury of the brachial plexus caused by complicated delivery of the infant during birth. It results in a varying degree of paralysis of the upper extremity, depending on the extent of the injury. The incidence of BPBP, which ranges from 0.04% to 0.5% of live births, has been increasing and may be related to the rise in infant birth size[1-4]. While most children recover fully without intervention, those who experience long-term sequelae of BPBP can have enormous enhancements in quality of life with orthopaedic surgery.

The most common and mildest form of BPBP involves the C5 and C6 nerve roots, the classic Erb's palsy. In this form, the child's shoulder is adducted and internally rotated, the elbow extended, the forearm pronated, and wrist flexed ("waiter's tip" position). C7 is variably affected in this group and portends a worse prognosis when involved. Pure lower root palsies are rare and primarily demonstrate deficits with hand function. Global palsies (C5-T1) have the worst prognosis, especially if associated with a Horner's syndrome or phrenic nerve palsy[1,3-4].

Infants with BPBP should be monitored very closely over the first several months of life as the severity of the injury declares itself. As many as 90% of cases resolve spontaneously,[1-2] although some experts argue that many of these cases likely have residual deficits that simply do not have a notable effect on overall function[2]. The presence of anti-gravity biceps function by 3 months of age is used as the standard for determining which patients can expect a full or near-full recovery with physical therapy alone, and who may benefit from surgical intervention[1-2,4-5]. Children with no biceps function by 3-6 months of age are considered candidates for microsurgery to repair or reconstruct the damaged nerves. However, while microsurgical techniques aim to enhance function and alter the natural history of BPBP, the expectation of a normally-functioning limb is not realistic. Most patients who do not recover spontaneously will develop secondary contractures and progressive deformity of the upper extremity despite a "successful" microsurgical intervention.[1,4].

Contractures and deformities, most commonly involving the shoulder, develop from the muscle imbalance inherent in the palsy. Weak shoulder external rotators and abductors are overpowered by the unaffected internal rotators and adductors. The imbalance also can affect the forces present at the glenohumeral joint. The humeral head is forced into the posterior glenoid, which causes dysplasia of the developing glenohumeral joint and subsequent instability.[1-2] Contractures can be seen in patients as young as 5 months of age and tend to be progressive, with significant glenohumeral deformity by 2 years of age.[1-2,4,6]. Internal rotation/adduction contractures limit the ability to bring the hand to the face or over the head. Children develop awkward compensatory maneuvers, relying on neck flexion, shoulder abduction, and/or scapulothoracic motion to bring the hand to the mouth[4,7].

A variety of orthopaedic procedures are performed to address these contractures and deformities. Most patients will benefit from a combination of soft tissue releases and tendon transfers. Older patients with more advanced deformity and those who continue to have poor function despite soft tissue procedures may be treated with humeral derotational osteotomy to reposition the upper extremity[4,8-9]. Currently,

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TABLE I. Mallet Classification[10]

	1	2	3	4	5
Global External Rotation	No active motion	<0°	0-20°	>20°	Normal active motion
Global Abduction	No active motion	<30°	30-90°	>90°	Normal active motion
Hand-to-Mouth	No active motion	Marked trumpet sign*	Partial trumpet sign	<40° of abduction	Normal active motion
Hand-to-Neck	No active motion	Not possible	Difficult	Easy	Normal active motion
Hand-to-Spine	No active motion	Not possible	S1	T12	Normal active motion

* The trumpet sign refers to shoulder abduction to bring the hand to the mouth, as if playing a trumpet, to compensate for loss of external rotation.

there are no distinct guidelines on how aggressive surgeons should be in their management. For instance, the literature is inconsistent in terms of which tendons and other anatomic structures should be released, lengthened, or transferred. Treatment may be as simple as a subscapularis release or as complex as lengthening of the pectoralis major, subscapularis and conjoint tendon with transfer of the latissimus dorsi and teres major. Surgery can also be performed open or arthroscopically.

The purpose of this study is to systematically evaluate the current literature on brachial plexus birth palsy to characterize patient outcomes following the different surgical treatments for secondary shoulder contractures and deformities. The Mallet classification[10] (Table I) is a method of grading motor function of the shoulder that has been used in the literature as a reliable tool for evaluating patients with BPBP. The Mallet score assesses hand-to-mouth, hand-to-neck, and hand-to-spine movements as well as global abduction and global external rotation. Each motion is given a score of 1 (no function) to 5 (normal function), and the sum of these numbers is the overall Mallet score. Because it focuses on coordinated movements that are important for activities of daily living in addition to single-plane range of motion, we chose to use the Mallet classification as the primary outcome of interest.

Methods

We performed a search of the PubMed computerized literature database for articles associated with the keywords "brachial plexus birth palsy" with limits of "English language" and "human." The list of returned references was then scrutinized for pertinent articles first based on title, then abstract, then full text. Inclusion criteria were as follows: (1) level I, II, III, or IV study design by Journal of Bone and Joint Surgery criteria; (2) surgical treatment of the brachial plexus birth palsy; (3) treatment for shoulder pathology (rather than elbow, wrist, or hand pathology); (4) treatment involving soft tissue release, soft tissue lengthening, tendon transfer, and/or humeral osteotomy; and (5) results of treatment classified by Mallet score. Exclusion criteria were (1) isolated microsurgical treatment of the palsy (e.g. nerve repair, nerve grafting, or neurolysis); and (2) inability to separate out patients by tables or text in studies that had multiple different treatments.

The literature search returned 361 unique references. We excluded 264 articles based on title alone because they were not pertinent to our subject or did not meet the study design criterion (eg. reviews, case reports, letters to the editor, or erratum). An additional 43 articles were excluded based on the abstract for failure to meet the inclusion criteria. The remaining 54 articles were reviewed in their entirety. Thirty-

TABLE II. Articles Excluded Following Full Text Review

Reason for Exclusion	Number of Articles Excluded
Mallet scores not reported	18
Multiple treatments not distinguished in article	5
Isolated microsurgical treatment	4
Osteotomy not performed on humerus	4
Study design criterion not met	3
No description of surgical procedure	1
Non-obstetric brachial plexus palsy included	1
No post-operative outcomes listed	1

TABLE III. Study Characteristics for Soft Tissue Procedures

Study	Year	Level of Evidence	N	Type of Palsy	Surgery
Cohen et al.[11]	2010	IV	32	26 C5-6, 2 C5-7, 2 C5-C8, 2 C5-T1	Subscapularis release, +/− latissimus and teres major transfer, +/− capsulorraphy
Kozin et al.[12]	2010	IV	44	36 C5-6, 8 C5-7	Arthroscopic latissimus and teres major transfer, partial subscapularis release, capsular release, joint reduction
Kozin et al.[6]	2010	IV	24	19 C5-6, 5 C5-7	Latissimus and teres major transfer, +/− subscapularis release, +/− pectoralis major lengthening
Terzis and Kostopoulos[13]	2010	IV	46	16 C5-6, 30 C5-T1	Latissimus and teres major rerouting, pectoralis major transfer, subscapularis and conjoint tendon lengthening
Waters and Bae[8]	2008	IV	23	Not specified	Latissimus and teres major transfer, pectoralis major lengthening, +/− subscapularis lengthening, +/− posterior capsule reefing, joint reduction
Van Kooten et al.[14]	2008	IV	9	Not specified	Latissimus transfer
Ahmed and Hashmi[15]	2006	IV	10	10 C5-6	Latissimus and teres major transfer, pectoralis major, subscapularis and conjoint tendon lengthening
Kozin et al.[16]	2006	IV	23	18 C5-6, 5 C5-7	Latissimus and teres major transfer, +/− subscapularis release, +/− pectoralis major lengthening
Waters and Bae[17]	2005	IV	25	Not specified	Latissimus and teres major transfer, +/− pectoralis major, subscapularis and conjoint tendon lengthening
Waters and Peljovich[18]	1999	III	32	10 C5-6, 14 C5-7, 8 C5-T1	Latissimus and teres major transfer, pectoralis major release
Waters[19]	1999	III	9	2 C5-6, 5 C5-7, 2 C8-T1	Latissimus and teres major transfer, pectoralis major release
Chuang et al.[20]	1998	IV	29	Not specified	Teres major and pectoralis major transfer (clavicular part), pectoralis major lengthening (sternal part), +/− latissimus lengthening
TOTAL			306	154 C5-C6, 44 C5-C7, 2 C5-C8, 2 C8-T1, 40 C5-T1	

seven of these articles were excluded according to the reasons listed in table II.

Seventeen studies[6,8-9,11-24] remained that met all inclusion and exclusion criteria and are the focus of this review. We gathered data on type of palsy, type of surgery, length of follow up, and Mallet scores. In studies that included patients who underwent different treatments or did not meet study criteria, we extracted pertinent data directly from the tables that listed outcomes individually. The studies were divided into soft tissue procedures and humeral osteotomy procedures for analysis.

Results

The majority of studies were retrospective case series (Level IV) reporting on a single surgical technique (Tables III and IV). Soft tissue procedures had some variation in the extent of tissue releases or lengthenings, usually based on the intraoperative gains in range of motion. Two studies[18-19] were comparison studies generated from a prospectively-collected database (Level III) that listed data for patients who underwent soft tissue procedures and patients who underwent humeral osteotomy procedures separately. Therefore, these studies were included in both groups for analysis.

TABLE IV. Study Characteristics for Humeral Osteotomy Procedures

Study	Year	Level of Evidence	N	Type of Palsy	Location of Osteotomy
Abzug et al.[9]	2010	IV	23	15 C5-6, 3 C5-7, 5 C5-T1	Diaphyseal
Waters and Bae[21]	2006	IV	27	Not specified	Proximal to deltoid insertion
Al-Qattan[22]	2002	IV	15	15 C5-6	Distal to deltoid insertion
Waters and Peljovich[18]	1999	III	16	1 C5-6, 11 C5-7, 2 C5-T1	Proximal to deltoid insertion
Waters[19]	1999	III	7	2 C5-6, 3 C5-7, 2 C8-T1	Proximal humerus
Kirkos and Papadopoulos[23]	1998	IV	22	18 C5-6, 4 C5-T1	Between pectoralis major and subscapularis insertions
Glez Cuesta et al.[24]	1982	IV	6	8 C5-6	Between deltoid and pectoralis major insertions
TOTAL			116	59 C5-6, 17 C5-7, 2 C8-T1, 11 C5-T1	

The seventeen studies represented a total of 422 patients. Three-hundred six were treated with soft tissue procedures, and 116 were treated with humeral derotational osteotomy procedures. The distribution in type of palsy followed the overall incidence reported in the literature. Most patients in both groups had a C5-C6 (Erb's) palsy. The next most common palsy was C5-C7, then C5-T1. A pure lower plexus palsy (C8-T1) was rare. One of the soft tissue procedure studies[13] had an unusually high number of patients with complete plexus involvement. Mean follow up was 3.2 years for the soft tissue procedure studies and 5.9 years for the humeral osteotomy procedure studies.

In the soft tissue procedure group, tendon transfers of the latissimus dorsi and teres major served as the basis of most surgeries. Additional surgical components included release or lengthening of pectoralis major, subscapularis, and/or the conjoint tendon, joint reduction, and capsule release or capsulorrhaphy. One study[12] performed the surgery arthroscopically.

TABLE V. Mallet Scores for Soft Tissue Procedures

Study	Follow up (years)	Mean Mallet Score (ER)	Mean Δ Mallet (ER)	Mean Mallet Score (Abd)	Mean Δ Mallet (Abd)	Mean Mallet Score (Total)	Mean Δ Mallet (Total)
Cohen et al.[11]	9.5	N/A	N/A	N/A	N/A	15.8	N/A
Kozin et al.[1]2	1	3.7	1.6	3.9	0	17.1	4.4
Kozin et al.[6]	2.9	3.4	0.8	3.9	0.3	16	1.3
Terzis and Kostopoulos[13]	6.6	4	2	4	2	N/A	N/A
Waters and Bae[8]	2.6	4	2	N/A	N/A	18	8
Van Kooten et al.[14]	N/A	2	0.1	N/A	0	N/A	N/A
Ahmed and Hashmi[15]	2.1	4.3	1.5	4.7	1.4	21.4	6
Kozin et al.[16]	1.1	3.2	0.6	4.1	0.5	16.4	1.59
Waters and Bae[17]	3.6	3.5	1	4	0.92	18	5
Waters and Peljovich[18]	1.6	3.7	1.7	4	1.1	15.6	6.1
Waters[19]	2.4	4	2	3.9	1	N/A	N/A
Chuang et al.[20]	2	N/A	N/A	86% 4-5, 14% 3	N/A	N/A	N/A
MEAN	3.2	3.6	1.3	4.1	0.8	17.3	4.6

ER = external rotation, Abd = abduction, Δ = change in, N/A = not available

TABLE VI. Mallet Scores for Humeral Osteotomy Procedures

Study	Follow up (years)	Mean Mallet Score (ER)	Mean Δ Mallet (ER)	Mean Mallet Score (Abd)	Mean Δ Mallet (Abd)	Mean Mallet Score (Total)	Mean Δ Mallet (Total)
Abzug et al.[9]	2.2	3.3	0.9	3.7	0.2	16.1	2.3
Waters and Bae[21]	3.7	4	2	N/A	N/A	18	5
Al-Qattan[22]	3	4*	1.8*	N/A	N/A	N/A	N/A
Waters and Peljovich[18]	3.1	3.8	1.7	3.8	0.8	15.1	5.6
Waters[19]	9.1	3.9	1.9	3.9	1	N/A	N/A
Kirkos et al.[7]	14	3*	1.8*	N/A	N/A	N/A	N/A
Glez Cuesta et al.[24]	N/A	2.8*	1*	N/A	N/A	N/A	N/A
MEAN	5.9	3.5	1.6	3.8	0.7	16.4	4.3

ER = external rotation, Abd = abduction, Δ = change in, N/A = not available

*Mallet score not distinguished for a specific category in the study

There was some variation in reporting of Mallet scores. Typically, the 5 categories evaluated by the Mallet classification are each given a score from 1 to 5 and the total Mallet score represents the sum of these 5 scores. Whenever available, the total Mallet score as well as the Mallet scores for external rotation and abduction are listed. A few studies only reported a single Mallet score in the range of 1 to 5 without distinguishing to which category it referred or if it was the mean of all 5 categories. These studies are marked with an asterisk in tables V and VI, and the results are listed under the Mallet score for external rotation as that was the most universally reported category.

All Mallet scores improved regardless of the surgery performed. Mean Mallet scores and mean gains in Mallet score were similar for the soft tissue procedure group and the humeral osteotomy group. Tables V and VI list Mallet scores for each study.

Discussion

Brachial plexus birth palsy can be a devastating diagnosis because of its potential to cause lifelong shoulder problems for the affected infant. It demonstrates the basic principle of pediatric musculoskeletal development that muscle imbalance in a growing child results in bone and joint abnormalities[4]. For the majority of cases that resolve spontaneously in the first few months of life, physical therapy to maintain passive range of motion and prevent contractures can preserve shoulder motion while awaiting the return of motor function. Patients who do not have anti-gravity biceps function by 3 months of age should be expeditiously referred to a specialist to begin more aggressive management as contractures and joint deformity can appear as early as 5 months of age[1-2].

The limitations in external rotation and abduction that develop from BPBP restrict the ability to raise the hand above the level of the shoulder, interfering with basic tasks such as feeding and grooming. Children may also fatigue more quickly when using the hand away from the body because of weak shoulder muscles[4]. The goals of surgical intervention are to restore functional range of motion and provide a more balanced joint to minimize the progression of glenohumeral joint deformity. Glenohumeral dysplasia can result in infantile subluxation or dislocation. Recent literature has stressed the importance of adding intra-articular manipulation (eg. joint reduction, capsular stabilization) to the standard tendon transfers and releases;[1,6] the pediatric shoulder has the ability to remodel in the setting of a reduced and rebalanced glenohumeral joint[2,6].

The studies included in this review demonstrate that surgical intervention for BPBP generally results in improved Mallet scores, regardless of the type of surgery performed. The improvements are clinically relevant, as 1 to 2 points on the Mallet score can mean the difference in a previously impossible task, such as bringing the hand to the mouth, becoming possible or even "easy". However, it is important to note that the total Mallet scores are still far from the normal score of 25. Parents should not be given unrealistic expectations when discussing surgical options. Furthermore, gains may diminish over time. Kozin et al. found slightly lower Mallet scores in their 3-year follow up study[6] as compared to their 1-year follow up study[16]. As such, exercises to prevent return of contractures are an important part of long-term therapy in the setting of persistent neurologic deficits.

The specific components of the soft tissue procedures varied greatly between studies and even within studies, making it difficult to endorse a common technique. Some authors indicated that the decision to release or lengthen additional tissue was based on the residual restrictions in motion following their standard procedure. Yet even the baseline procedures varied, including the option of performing the surgery arthroscopically. The difference may be partly surgeon preference, but often such variation in technique makes it impossible to determine which technique is best. This knowledge deficit may guide future investigations.

The decision to perform a humeral derotational osteotomy rather than a soft tissue procedure is typically based on the severity of the glenohumeral joint deformity. Experts agree that humeral osteotomy is indicated for cases of advanced deformity with humeral head flattening, glenoid dysplasia, and subluxation or dislocation[1-2,4,8,21]. Such cases are past the point of remodeling capability, so functional improvements will be hindered by joint asymmetry even with better muscle balance. The goal of humeral osteotomy is not to fix the joint itself, but to rotate the extremity to a more functional arc of motion.[1-2,9,21] The studies in this review did not consistently report severity of joint deformity, so we cannot say that the groups are equivalent or that one has better outcomes. However, presuming that patients treated with osteotomy generally had more severe deformity than those treated with soft tissue procedures, Mallet scores were surprisingly good for the osteotomy group.

As with all systematic reviews, this review is limited by the inherent biases and weaknesses of the included studies. The large majority of studies were retrospective case series. Randomized control trials may be inappropriate for this patient population, but our understanding of how to best treat patients with BPBP could be greatly enhanced by more well-designed comparative studies. We chose to use Mallet scores as a primary outcome measure because it provided a standardized and clinically relevant tool to analyze studies. Unfortunately, this eliminated 18 additional studies that might have similarly important results to contribute. Another limitation is that 1 author had 3 studies and 1 author had 5 studies included in the review. With a small total number of studies, the biases of these 2 authors may overshadow the results of other authors. This drawback is difficult to avoid in rare injuries because the majority of affected patients are typically treated at regional centers.

In summary, brachial plexus birth palsy can be a severely disabling condition for an otherwise healthy infant, but there has been an enormous effort by the orthopaedic community to develop treatments that address the various stages of disability. Early and aggressive treatment of contractures and joint deformity with tendon transfers, contracture releases, and joint reduction is warranted to halt or reverse these changes. When cases are too advanced to salvage the glenohumeral joint, humeral derotational osteotomy is indicated to restore function. Both techniques have shown consistent improvements in motion according to the Mallet classification. Future comparative studies may help orthopaedic surgeons choose the most appropriate surgery for individual patients.

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