SCIENTIFIC ARTICLE

Postoperative Loss of Midline Function in Brachial Plexus Birth Palsy

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Purpose To identify the rate of and predictive variables for functionally limited shoulder internal rotation in postoperative patients with brachial plexus birth palsy.

Methods Records of patients with brachial plexus birth palsy who had surgery on the affected upper extremity during a 10-year period were retrospectively reviewed. Patient demographics, physical examinations, and all upper extremity procedures were recorded. Loss of midline function (LOM) was defined as a Modified Mallet Scale or Active Movement Scale (AMS) internal rotation score <3. Exclusion criteria were <1-year follow-up after the most recent procedure, insufficient documentation, or preexisting LOM. Multivariable logistic regression was performed on 3 different scenarios of candidate variables to identify those associated with LOM. All scenarios included each procedure as a candidate variable. Scenario A additionally analyzed preprocedural AMS scores. Scenario B additionally analyzed preprocedural Modified Mallet Scale scores. Scenario C isolated the surgical pathway without preprocedural examination scores.

Results Among 172 included patients, 34 (19.8%) developed LOM. Predictive variables associated with LOM included severity of initial palsy (C5-7, odds ratio 3.6; C5-T1, odds ratio 4.9), poor recovery of upper trunk motor function before the patient's first surgery (specifically Modified Mallet Scale abduction < 4, AMS elbow flexion < 3, and AMS wrist extension < 3), and patients who ultimately required surgical glenohumeral reduction (odds ratio 3.6). Age, number of procedures, closed shoulder reduction with casting, shoulder tendon transfers, and external rotation humeral osteotomies were not predictive of LOM.

Conclusions Approximately 1 in every 5 patients with brachial plexus birth palsy will develop LOM after entering a surgical algorithm designed to improve shoulder external rotation. Patients with a more severe initial palsy (C5-7 or global), poor spontaneous recovery of upper trunk motor function (elbow flexion or wrist extension) before their first procedure, and those who ultimately require surgical glenohumeral joint reduction should be counseled as having a higher odds of LOM development. *(J Hand Surg Am. 2017;■(■):■−■. Copyright © 2017 by the American Society for Surgery of the Hand. All rights reserved.)*

Type of study/level of evidence Therapeutic IV.

Key words Brachial plexus birth palsy, loss of midline function.

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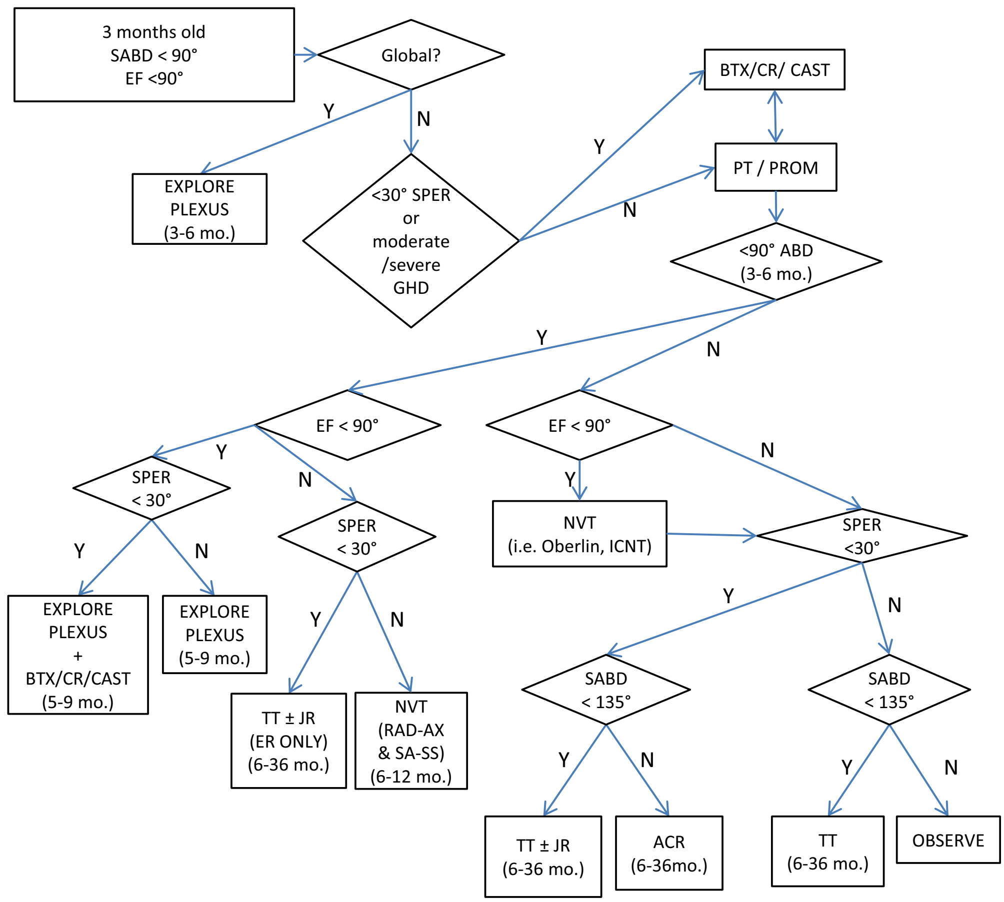
Despite advances in perinatal care, brachial plexus birth palsy (BPBP) continues to affect 1.5 per 1,000 live births in the United States.[1] Although most infants have spontaneous recovery of upper extremity function, as many as 35% exhibit persistent neurologic deficits, resulting in abnormally balanced muscular forces followed by progressive joint deformity and limited motion about the shoulder.[2] Even if antigravity elbow flexion is seen before 6 months of age, patients with persistent upper trunk palsy may never achieve full shoulder abduction and external rotation.[3,4] These patients can develop anterior shoulder capsular and muscular contractures that are likely due to a complex combination of intrinsic fibrosis secondary to denervation as well as dynamic muscular shoulder imbalance.[5] Shoulder internal rotators that are at least partially C7-innervated (eg, pectoralis major, teres major, latissimus dorsi, subscapularis) overpower the weakened C5-6 innervated external rotators (eg, infraspinatus, teres minor), resulting in decreased active, then passive, external rotation, a persistent internal rotation posture, glenohumeral dysplasia, posterior humeral head subluxation, and subsequently, lifelong shoulder dysfunction.[6]

Physical therapy initially aims to restore passive external rotation and abduction, but several surgical procedures, that can be performed subsequently, are available to balance internal and external rotators or restore glenohumeral alignment with hopes of optimizing joint congruity. These include closed shoulder reduction and casting (with or without onabotulinumtoxin A injection), arthroscopic or open joint reduction techniques, and tendon transfers.[7–11] When glenohumeral dysplasia is deemed irreversible, humeral rotational osteotomies may be considered to improve the position of the upper extremity. Although these procedures are designed to improve overall upper extremity function, they may lead to a loss of shoulder internal rotation and/or a functional loss of the ability to reach the midline.[9] Loss of midline function (LOM) impairs a child's ability to perform activities of daily living such as dressing, bimanual tabletop work, and perineal hygiene. Still, LOM is an infrequently discussed and poorly quantified clinical problem. The goals of this study were to quantify the prevalence of LOM and to identify a risk profile that may predispose certain patients with BPBP to LOM function.

METHODS

After internal review board approval, medical records of patients with BPBP treated between January 2002 and June 2013 were reviewed. The patients who were included underwent procedures aimed at improving external rotation and abduction (closed reduction and casting with or without onabotulinumtoxin A injections, arthroscopic or open glenohumeral joint reduction, latissimus dorsi and/or teres major tendon transfers about the shoulder, and/or humeral external rotation osteotomy). The current treatment algorithm used by our institution is provided in Figure 1. In brief, brachial plexus exploration, nerve grafting, and/or nerve transfer is performed for patients with global palsy as young as 3 months old and in patients with upper trunk involvement who have not recovered elbow flexion by 6 months old. We used a history of any sural nerve grafting and/or nerve transfer as a surrogate for surgical exploration to represent early intervention for more severe injury. Indications for shoulder closed reduction and casting included no passive external rotation past neutral, or progressive loss of external rotation in patients younger than 2 years. Tendon transfers were performed for children who lacked external rotation after 18 months old or as early as 6 months old in cases of severe dysplasia that failed casting. Older children (age 3 years and more) were offered humeral osteotomy if they had persistent internal or external rotation deficiency or moderate-to-severe glenohumeral dysplasia.

Charts were reviewed for demographic information, levels of palsy, laterality, history of sural nerve grafting or nerve transfer, and other surgical interventions. The newly Modified Mallet Scale (MMS)[9,12] that includes internal rotation as a sixth category and the Active Movement Scale (AMS)[13] were recorded both preoperatively (less than 1 month before the procedure) and during the most recent follow-up examination (after a minimum of 1 year after surgery for the patient's last procedure). LOM was defined as either an MMS or AMS internal rotation score less than 3. This definition was selected based on clinical relevance, as scores less than 3 in either category imply that the child is unable to position his or her arm near his or her umbilicus and thus cannot actively participate in midline activities with the affected limb. Whenever possible, both AMS and MMS were obtained. However, the MMS was not always attainable in younger patients. There were no instances whereby both MMS and AMS scores were available and had conflicting values with respect to LOM. It is important to note that we did not use hand to spine to represent shoulder internal rotation because this also incorporates movements of shoulder abduction and extension.[14] Patients with less than 1 year of follow-up, insufficient documentation, or preoperative LOM were excluded.



All movements are active anti-gravity unless specified as passive.
Plexus exploration involves intraoperative decision to proceed with nerve transfers and/or sural nerve grafting.
All TT/ACR indications can be extended past 36 months if minimal GHD.
Shoulder ultrasound/MRI performed throughout process as appropriate.
Salvage options (i.e. humeral osteotomy) considered when >3 years old if shoulder rotational deficiencies persist.

SABD: shoulder abduction
EF: elbow flexion
SPER: shoulder passive external rotation
GHD: glenohumeral deformity/dysplasia
BTX/CR/CAST: botox, closed reduction, and casting in abduction/external rotation
NVT: nerve transfers
RAD-AX: radial to axillary nerve transfer
SS-SA: suprascapular to subscapularis nerve transfer
TT: tendon transfers to augment shoulder external rotation (latissimus teres + teres major, teres major only, or lower trapezius)
JR: joint reduction (open or arthroscopic, +/- soft-tissue releases)
ACR: arthroscopic capsular release

FIGURE 1: Brachial plexus birth palsy treatment algorithm.

Examinations were performed by licensed occupational therapists with at least 5 years' experience working in a busy BPBP clinic.

The study sample was divided into 2 groups based on postoperative maintenance, versus LOM. Continuous variables were assessed using the *t* test. Categorical variable frequencies were compared using the chi-square or Fisher exact test. MMS and AMS subscores were grouped into clinically relevant aggregates (scores <3 and scores ≥ 3) because certain MMS or AMS scores are rarely achieved by our patients with brachial plexopathies (ie, excellent external rotation at the time of presentation in this cohort is rare).

Multivariable logistic regression for 3 different scenarios of candidate variables was performed with the binary outcome of interest being LOM (yes or no). All scenarios (A, B, and C) included levels of palsy, number of total procedures, and all procedures performed (ie, brachial plexus exploration and neurolysis, nerve grafting, nerve transfer, closed glenohumeral reduction and casting with or without onabotulinumtoxin A, external rotation tendon transfers, surgical glenohumeral reduction, and external rotation humeral osteotomy) as candidate variables. Scenarios A and B additionally analyzed preprocedural AMS or MMS values, respectively. Preprocedural AMS and MMS examination scores were not collectively analyzed in 1 scenario because any patient who did not have both a preprocedural AMS and MMS available would be excluded. To predict LOM based on a patient's treatment course (regardless of preprocedural examination scores), candidate variables in scenario C did not include preprocedural examination scores. Given the preponderance of arthroscopic reductions included in our study (likely due in part to a separate study responsible for meticulous data collection in

TABLE 1. Patient Demographics (n = 172)

	Total	% LOM		*P* Value	
Study population	172	19.8			
Race					
Caucasian	104	20.2		.658	
African American	45	22.2			
Other	23	13			
Sex					
M	102	19.6		.949	
F	70	20			
Laterality					
R	72	23.6		.283	
L	100	17			
Number of procedures					
1	61	13.1		.084	
2	60	18.3			
3	31	22.6			
4	10	30			
5	7	42.9			
6	3	66.7			
Level			*Odds ratio (CI)*	.002	
C5-6	100	11			
C5-7	55	30.9	3.6 (1.6–8.5)		
C5-T1	16	37.5	4.9 (1.4–16.0)		
					Average age (y)
Brachial plexus exploration	33	33.3	2.5 (1.1–5.9)	.029	6.7 mo
Sural nerve grafting	28	35.7	2.8 (1.1–6.8)	.021	7 mo
Nerve transfers	22	27.3		.390	8.7 mo
Closed GH reduction + casting*	60	23.3		.390	14.2 mo
ER tendon transfers	107	17.8		.396	3.6 y
Surgical GH reduction (arthroscopic or open)	106	26.4	3.6 (1.4–9.2)	.006	2.6 y
Arthroscopic	89	25.8	2.3 (1.03–5.0)	.038	2.6 y
Open	19	26.3		.540	2.7 y
ER HO	4	0		.586	8.4 y
IR HO	12	n/a		n/a	6 y

CI, confidence interval; ER, external rotation; GH, glenohumeral joint; HO, human osteotomy; IR, internal rotation (performed after LOM); LOM, loss of midline function.

*With or without onabotulinumtoxin A.

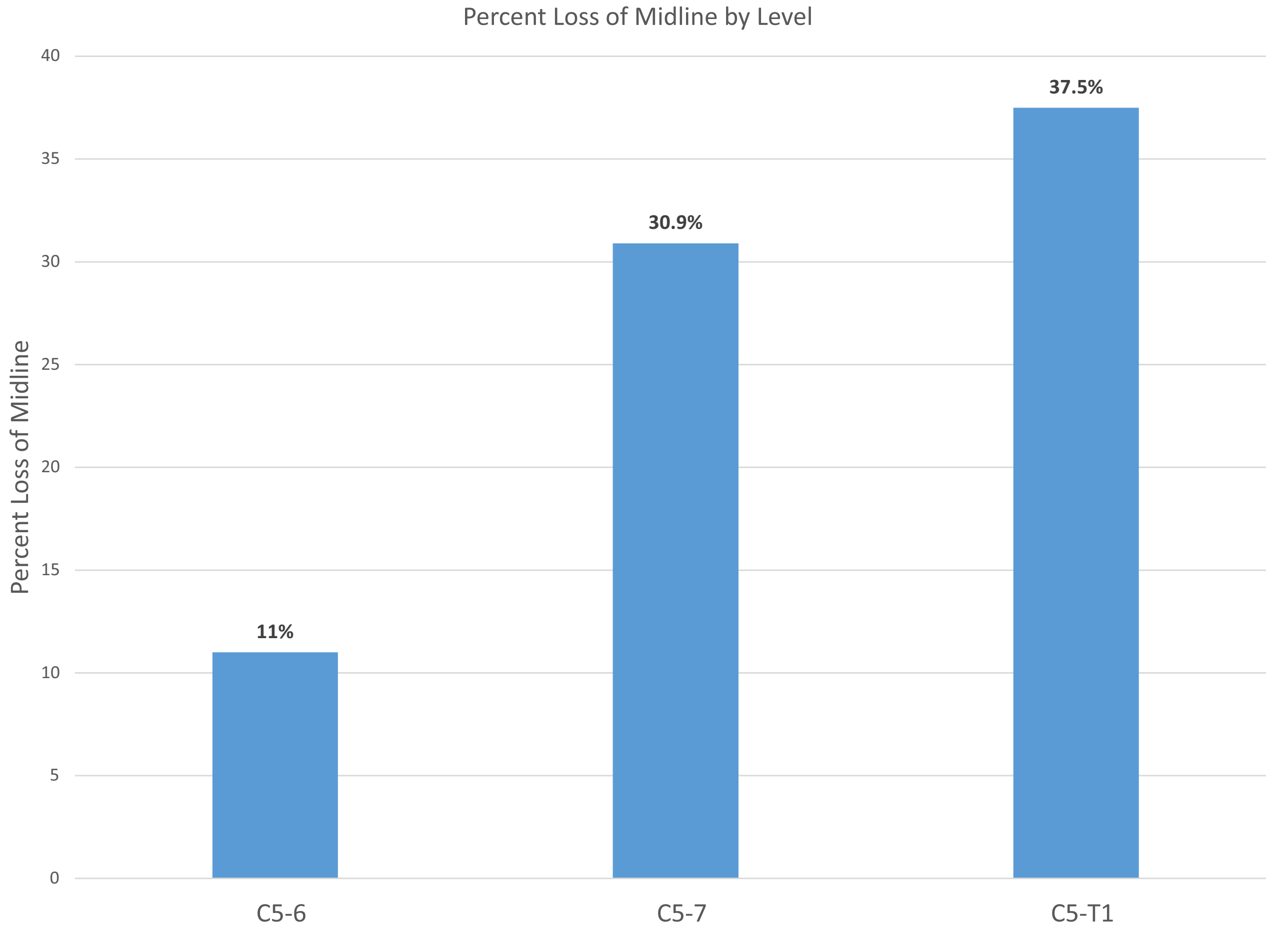


FIGURE 2: Percent LOM by level of palsy.

those patients), arthroscopic and open glenohumeral reductions were combined into 1 single potential predictive variable (termed glenohumeral joint reduction). All logistic regression models identified were then compared based on the c-statistic to identify any differences in predictive power.

All P values are 2-sided when applicable. Statistical significance was defined as $P < .05$.

RESULTS

A total of 172 patients with BPBP satisfied the criteria for inclusion in the study. The median follow-up duration after surgery was 49 months (range, 12−159 mo). LOM occurred in 34 of the 172 included patients (19.8%). Among 270 identified patients who underwent a procedure of interest, 20 were excluded because of baseline internal rotation deficits before any surgical intervention and 78 patients had insufficient documentation or follow-up. Among the included patients, there were 111 preprocedural MMS, 92 preprocedural AMS, 166 final follow-up MMS, and 35 final follow-up AMS scores completely documented. A summary of patients with their associated procedures is in Table 1.

Most patients who developed LOM (68.4%) had either an initial C5-7 injury (n = 17, 30.9%) or a global palsy (n = 6, 37.5%) (Fig. 2). Compared with a C5-6 injury, there were significantly higher odds of LOM associated with patients with a C5-7 palsy (odds ratio = 3.6, 95% confidence interval 1.6−8.5, $P < .05$) and a global palsy (odds ratio = 4.9, 95% confidence interval 1.4−16.0, $P < .05$).

Average preprocedural AMS scores (n = 92) for internal rotation were 6.8 ± 0.6, 7.0 ± 0.01, and 5.7 ± 2.4 among C5-6, C5-7, and global palsies, respectively. Although global palsies tended to have lower preprocedural scores, none of these values demonstrated statistically significant differences. In 8 of the 15 AMS categories, a lower preprocedural AMS score was associated with LOM (Table 2). The strongest odds of LOM tended to be associated with decreased recovery of elbow flexion (C5-6) or wrist extension (C6-7) before the patient's first procedure and poor initial lower trunk function (C8-T1).

TABLE 2. Analysis of Preprocedural Active Movement Scale (AMS) Scores (n = 92)

AMS Score	No.	% LOM	OR	Confidence Interval	*P* value	Test
Wrist extension						
<3	28	53.6%	5.0	(1.9, 13.2)	<.001	chi-square
≥3	64	18.8%				
Elbow flexion						
<3	38	44.7%	3.6	(1.4, 9.1)	.007	chi-square
≥3	54	18.5%				
Shoulder flexion						
<3	45	42.2%	3.6	(1.4, 9.3)	.008	chi-square
≥3	47	17.0%				
Forearm supination						
<3	74	35.1%	9.2	(1.2, 50)	.013	chi-square
≥3	18	5.6%				
Thumb extension						
<3	25	48.0%	3.2	(1.2, 8.5)	.016	chi-square
≥3	67	22.4%				
Finger extension						
<3	18	50.0%	3.1	(1.1, 9.0)	.032	chi-square
≥3	74	24.3%				
Shoulder abduction						
<3	46	39.1%	2.6	(1.03, 6.8)	.039	chi-square
≥3	46	19.6%				
Finger flexion						
<3	8	62.5%	4.7	(1.04, 21.3)	.045	Fisher's exact
≥3	84	26.2%				
Thumb flexion						
<3	10	60.0%	4.4	(1.1, 17.0)	.059	Fisher's exact
≥3	82	25.6%				
Elbow extension						
<3	13	46.2%	2.4	(0.71, 7.9)	.191	Fisher's exact
≥3	79	26.6%				
Wrist flexion						
<3	13	46.2%	2.4	(0.71, 7.9)	.191	Fisher's exact
≥3	79	26.6%				
Shoulder external rotation						
<3	87	31.0%	n/a		.317	Fisher's exact
≥3	5	0.0%				
Shoulder adduction						
<3	6	50.0%	2.6	(0.49, 13.7)	.354	chi-square
≥3	86	27.9%				
Forearm pronation						
<3	13	23.1%	0.7	(0.17, 2.7)	.749	Fisher's exact
≥3	79	30.4%				

(Continued)

TABLE 2. Analysis of Preprocedural Active Movement Scale (AMS) Scores (n = 92) (Continued)

AMS Score	No.	% LOM	OR	Confidence Interval	*P* value	Test
Shoulder internal rotation						
<3	2	0.0%	n/a		1	Fisher's exact
≥3	90	30.0%				

LOM, loss of midline function; OR, odds ratio.

TABLE 3. Analysis of Preprocedural Modified Mallet Scale (MMS) Scores (n = 111)

Preprocedural MMS	No.	% LOM	OR	Confidence Interval	*P* Value	Test
Abduction						
<4	55	21.8%	7.5	(1.6, 35.5)	.004	chi-square
≥4	56	3.6%				
External rotation						
<3	83	15.7%	5.0	(0.6, 40.2)	.113	Fisher's exact
≥3	28	3.6%				
Internal rotation						
<4	13	15.4%	1.4	(0.3, 7.3)	.650	Fisher's exact
≥4	97	11.3%				
Hand to neck						
<3	86	11.6%	0.88	(0.2, 3.5)	1	Fisher's exact
≥3	23	13.0%				
Hand to spine						
<3	89	13.5%	2.8	(0.3, 23.0)	.457	Fisher's exact
≥3	19	5.3%				
Hand to mouth						
<3	70	12.9%	1.3	(0.4, 4.5)	.768	Fisher's exact
≥3	39	10.3%				

LOM, loss of midline function; OR, odds ratio.

Average preprocedural MMS scores (n = 111) for internal rotation were 3.8 ± 0.4, 3.9 ± 0.4, and 3.8 ± 0.5 among C5-6, C5-7, and global palsies, respectively. None of these values demonstrated statistically significant differences. Patients with limited preprocedural abduction (MMS abduction <4) had greater odds of developing LOM than those with adequate abduction (21.8% vs 3.6%, respectively; $P < .05$). None of the other preprocedural MMS variables were predictive of LOM, including internal rotation and hand to spine (Table 3).

Multivariable logistic regression also suggested that initial injury severity is associated with increased odds of developing postoperative LOM. Quantitative results of the 3 multivariable logistic regression scenarios are summarized in Table 4. Notably, the c-statistics suggest that all reported models have acceptable discrimination. Also, 2 bivariate logistic models were discovered in scenario A. However, a separate chi-square analysis concluded that there was a statistically significant association between those 2 alternative variables (preprocedural AMS elbow flexion and wrist extension, $P < .05$), thus making both 2-variable models under scenario A acceptable. Scenario A demonstrates that poor spontaneous recovery of the upper trunk before surgery and persistent glenohumeral malalignment is associated with higher odds of LOM. Scenario C demonstrates a very similar conclusion while isolating the surgical pathway (preprocedural examination scores were not candidate predictive variables) by reiterating that a more severe initial plexopathy and persistent glenohumeral abnormality is associated with increased odds of LOM. Lastly, there was no statistically

TABLE 4. Multivariable Logistic Regression: Predictive Variables for Loss of Midline Function

	Predictive Variables	OR	CI	*P* Value
Scenario A: Candidate Variables = All Procedures + Pretreatment AMS Scores (n = 92)				
Regression model 1	Glenohumeral joint reduction	9.2	(2.1, 39.0)	.003
	AMS elbow flexion <3	6.6	(2.2, 19.4)	<.001
			c-statistic = 0.754	
	-OR-			
Regression model 2	Glenohumeral joint reduction	4.2	(1.1, 16.6)	.037
	AMS wrist extension <3	4.9	(1.8, 13.4)	.002
			c-statistic = 0.727	
Scenario B: Candidate Variables = All Procedures + Pretreatment MMS Scores (n = 111)				
	MMS abduction	7.5	(1.6, 35.5)	.011
			c-statistic = 0.707	
Scenario C: Candidate Variables = Only Procedural History (n = 172)				
	C5-T1 (vs C5-6)	4.9	(1.4, 16.8)	.002
	C5-7 (vs C5-6)	4.1	(1.7, 9.9)	.002
	Glenohumeral joint reduction	4	(1.4, 10.6)	.005
			c-statistic = 0.731	

AMS, active movement scale; CI, confidence interval; MMS, modified mallet scale; OR, odds ratio.

significant difference between the predictive power of each scenario.

DISCUSSION

Historically, early surgical treatment of shoulder dysfunction in BPBP appropriately focused on the restoration of external rotation and abduction with a concurrent goal of eliminating internal rotation contractures.[3,7,8,10,15–17] The concern for decreased midline function throughout these patients' treatment course is not well documented despite the important role of internal rotation during several activities of daily living. Within 3 popular physical examination systems specifically validated to monitor the upper extremity in BPBP (Mallet classification, AMS, and Toronto Test score), only 1 maneuver out of 25 specifically isolates internal rotation (AMS internal rotation).[18] During an extensive review of his own clinical research regarding surgical options for children with BPBP, Kozin[19] described the absence of outcomes data available to assess a patient's ability to reach the midline in front of the body as a limitation. Pearl et al[20] echoed similar concerns after reviewing a series of arthroscopic releases with tendon transfers whereby, in certain cases, his patients demonstrated functional problems associated with postoperative shoulder internal rotation deficits. Abzug et al[9] subsequently recommended a dedicated internal rotation maneuver to their MMS scale to assess outcomes after external rotation humeral osteotomies. They concluded that the additional isolated internal rotation component is better than hand to spine at assessing midline function. Greenhill et al[21] compared 171 MMS scores (both with and without the internal rotation component) and concluded that patients with good external rotation but limited internal rotation can score deceptively high on the original composite Mallet scale, which is normally used to represent overall upper extremity function. Russo et al[14] used motion analysis to further verify that the MMS hand-to-belly component better represents true internal rotation when compared with hand to spine.

There are several unanswered questions regarding this concept of LOM, and this study answers only a few of them. The true incidence among all patients with BPBP is unknown, and controversy exists regarding the clinical relevance of LOM. For example, some authors claim that internal rotation lost after tendon transfers may be regained with time.[22,23] For this reason we included only patients with at least 1-year follow-up, although there is no current literature to indicate if this time frame is adequate for maximal improvement. Terzis and Kokkalis[24] observed loss of internal rotation within a functional range. Thus, they considered it negligible when compared with the functional gains observed in external rotation after shoulder tendon transfers. Unfortunately, we do not have patient selfassessments available for further analysis, but this should be a subject of further study. Moreover, wrist and finger flexion may also play a role in one's ability to reach his or her umbilicus. Finally, there was a 0% incidence of LOM in patients who underwent external rotation humeral osteotomy. We try to avoid LOM after external rotation humeral osteotomies by limiting the gains in external rotation proportional to the available surplus in internal rotation and on occasion by using triplanar osteotomy cuts.[25] However, it may be that maintenance of shoulder extension after humeral osteotomy, rather than precise rotational adjustments, may guard against LOM by allowing the hand to move closer to the belly. Regardless, knowing the rate at which postoperative patients lose midline function as well as which patients are initially at high risk may help guide further studies and patient follow-up when limited patient data are available.

This study suggests that the odds of LOM after reconstructive shoulder procedures in children with BPBP correlates with the severity of initial neurologic injury. Patients with poor recovery of upper trunk function before surgical intervention and those who ultimately required surgical glenohumeral reduction have an increased risk of LOM development compared with patients with isolated upper trunk injuries that recovered elbow flexion spontaneously. We believe that the weak internal rotation and denervation-related abduction and external rotation contractures associated with more severe injury (ie, global injury, poor spontaneous recovery, persistent glenohumeral dysplasia) can make the maintenance of shoulder dynamics after procedures designed to improve external rotation less predictable. Thus, the association of inadequate preprocedural abduction with LOM is also not surprising. Good abduction implies a less severe upper trunk injury, less imbalance, and a potentially better glenohumeral joint than more severely affected patients.[26]

Limited abduction and associated contractures in children with BPBP may be more prevalent than originally acknowledged, and its role in the treatment of children with BPBP is currently being elucidated.[27−29] The findings in this study suggest that a careful preoperative evaluation should weigh the potential gains in external rotation with the concomitant functional loss of internal rotation as measured by the MMS. It is now our practice to both counsel parents regarding the incidence of decreased midline function and compare the potential benefits of external rotation gains against the potential loss of internal rotation during surgical decision making and operative planning. We also caution parents about the possibility of an elective internal rotation humeral osteotomy if LOM occurs, as was the case in 12 of our 172 patients.

The current study has several limitations. It is a retrospective review whereby strict documentation and adequate follow-up narrowed the sample size in an already uncommon patient population. Thus, certain surgical subgroups were underpowered to conclude that they are not associated with LOM (ie, only 4 patients underwent external rotation humeral osteotomy). Another limitation of the study is the absence of a control group of patients in whom no surgery was attempted, thus making it impossible to conclude that all cases of LOM were iatrogenic as opposed to simply a consequence of the natural history. For example, the fact that 20 patients were initially excluded because of baseline internal rotation deficits before any surgical intervention implies that surgery alone is not the only risk factor for poor midline function. Nonsurgical risk factors also deserve further investigation. In addition, there may be a selection bias within this study population that protects shoulder external rotation tendon transfers and external rotation humeral osteotomies from losing midline function. Although there are no absolute contraindications to surgically enhance external rotation at our institution, we consider an MMS internal rotation score <4 a relative contraindication. Among included patients, only 13 of 111 preoperative MMS and 5 of 92 preoperative AMS internal rotation subscores were <4. Therefore, we were unable to evaluate if limited preoperative internal rotation is a potential variable associated with postoperative LOM. Finally, 21.5% of patients were evaluated pre- and postoperatively with different scoring systems because the age and cooperation of the child determines which system is used.

In conclusion, approximately 1 in every 5 patients with BPBP will develop LOM after entering a surgical algorithm designed to improve shoulder external rotation. Patients with a more severe initial palsy (C5-7 or global), poor spontaneous recovery of upper trunk motor function (elbow flexion or wrist extension) before their first procedure, and those who ultimately require surgical glenohumeral joint reduction have a higher odds of LOM development and those families should be counseled appropriately.

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