Ultrasound Screening for Posterior Shoulder Dislocation in Infants with Persistent Brachial Plexus Birth Palsy

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Background: Early detection of posterior shoulder dislocation in infants with brachial plexus birth palsy (BPBP) is essential, but it may be difficult to accomplish with physical examination alone. The aim of this study was to determine the prevalence of shoulder dislocation in patients with BPBP using ultrasound and to identify which physical examination measurements correlated most with dislocation in these patients.

Methods: This study was a retrospective review of data obtained in an ultrasound screening program of infants with BPBP born from January 2011 to April 2014. Physical examination included the use of the Active Movement Scale (AMS) and measurement of passive external rotation of the shoulder. Ultrasound measurements included PHHD (percentage of the humeral head displaced posterior to the axis of the scapula) and the alpha angle (intersection of the posterior scapular margin with a line tangential to the humeral head through the glenoid). Shoulder dislocation was defined as both a PHHD of >0.5 and an alpha angle of >30°.

Results: Of sixty-six infants who had undergone a total of 118 ultrasound examinations (mean, 1.8; range, 1 to 5), 19 (29%) demonstrated shoulder dislocation with the shoulder positioned in internal rotation; the dislocation was first detected between 2.1 and 10.5 months of age. Infants with a dislocated shoulder demonstrated significantly less mean passive external rotation in adduction (mean, 45.8° versus 71.4°, $p < 0.001$) and a greater difference between internal rotation and external rotation AMS scores (mean, 5.5-point versus 3.3-point difference, $p < 0.001$) than those without shoulder dislocation. Passive external rotation in adduction was a better measure for discriminating between dislocation and no dislocation (area under receiver operating characteristic curve [AUC] = 0.89) than was the difference between internal and external rotation AMS scores (AUC = 0.73). A cutoff of 60° of passive external rotation in adduction (≤60° versus° >60) yielded a sensitivity of 94% and a specificity of 69%.

Conclusions: Shoulder dislocation is common in infants with BPBP; 29% of the infants presenting to our tertiary care center had a dislocation during their first year of life. Ultrasound shoulder screening is appropriate for infants with BPBP. If passive external rotation in adduction is used to determine which infants should undergo ultrasound, ≤60° should be utilized as the criterion to achieve appropriate sensitivity.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

Peer Review: This article was reviewed by the Editor-in-Chief and one Deputy Editor, and it underwent blinded review by two or more outside experts. The Deputy Editor reviewed each revision of the article, and it underwent a final review by the Editor-in-Chief prior to publication. Final corrections and clarifications occurred during one or more exchanges between the author(s) and copyeditors.

Glenohumeral joint dysplasia is a known sequela of brachial plexus birth palsy (BPBP). Although the exact mechanism is unknown, denervation of the upper limb during the neonatal period leads to impaired growth and eventual contracture of the affected muscles[1]. As the neonatal glenohumeral joint is entirely cartilaginous, these contractures can lead to altered joint mechanics and eventually to irreversible joint deformity. Although the glenohumeral joint deformity is

Disclosure: No funding was received for this study. On the **Disclosure of Potential Conflicts of Interest** forms, which are provided with the online version of the article, one or more of the authors checked “yes” to indicate that the author had a relevant financial relationship in the biomedical arena outside the submitted work (http://links.lww.com/JBJS/C818).

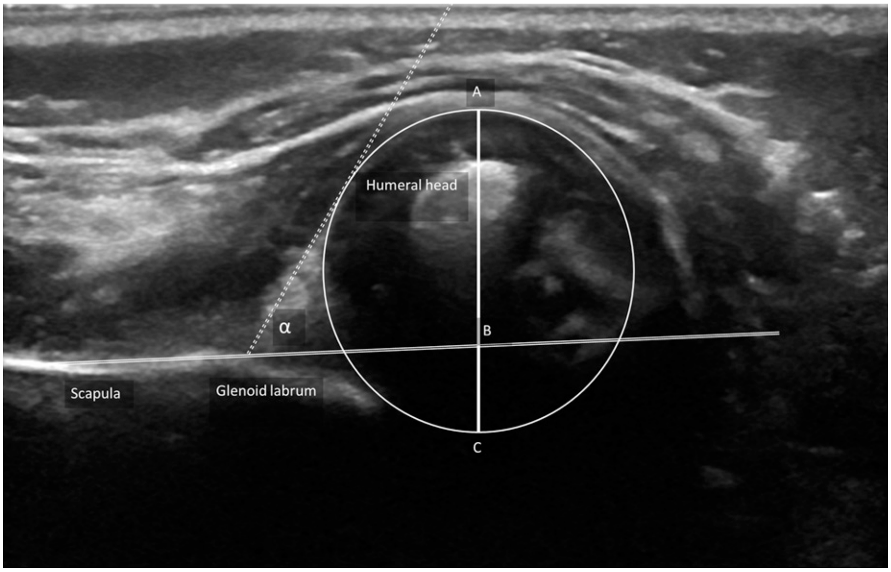

Fig. 1
Ultrasound image of a dislocated shoulder in an infant. A line is first drawn along the scapular border (double solid line). The α angle is the angle created by the intersection of this line at the echogenic glenoid labrum with a tangent drawn to the humeral head (double dotted line). Next, a circle is drawn to approximate the contour of the humeral head. The circle is bisected at its widest point (single line). Point B is where this line intersects with the scapular border line (double line). The PHHD is the ratio of line AB to line AC.

etiologically secondary to the nerve injury, changes to the joint can happen quite early in life, such that glenohumeral joint dislocation can occur during the infantile period. This is not a true dislocation, with complete loss of contact at the articular surfaces, but rather a developmental dislocation in which the humeral head and glenoid develop together into a posteriorly-oriented alignment. Glenohumeral joint dysplasia in association with BPBP has been recognized since early in the twentieth century[2]. The characteristic posterior shoulder dysplasia associated with BPBP was classified with magnetic resonance imaging (MRI) by Waters et al. in 1998[3], with inclusion of a category (type VI) for infantile dislocation. The concept of infantile dislocation was refined in 2004 by Moukoko and colleagues[4], who found an 8% prevalence of dislocation in a cohort of infants presenting to a tertiary care center for BPBP. The authors noted physical examination findings characteristic of dislocation, most notably the sudden loss of passive external rotation, and recommended ultrasound evaluation of infants with such findings. Other physical examination signs that should raise concern that the shoulder is dislocated are fullness in the posterior aspect of the shoulder, apparent shortening of the humerus, and asymmetry in the soft-tissue folds of the upper arm[4].

Early diagnosis of infantile glenohumeral dysplasia associated with BPBP is essential in order to treat it appropriately. Earlier diagnosis enables earlier treatment to reduce the glenohumeral joint and potentially provide more normal shoulder development. It is likely that, on the basis of the type and extent of the nerve injury, some infants with BPBP are more at risk for infantile dislocation than others. One characteristic physical examination finding is a lack of passive external rotation of the shoulder, but it is likely that physical examination alone will miss some dislocations. There is a strong precedent for targeted ultrasound screening of infants at risk for developmental hip dysplasia, making ultrasound a natural choice for screening for glenohumeral joint dysplasia in infants with BPBP[5,6]. The technique of shoulder ultrasound in BPBP has been well-described, but which infants with BPBP should undergo ultrasound screening is not yet understood[7].

The aim of this study was to evaluate the use of ultrasound for identifying infantile shoulder dislocation. Specifically, we sought to identify the prevalence of infantile shoulder dislocation as detected on ultrasound in infants with BPBP seen at a tertiary referral center as well as to identify which physical examination measures correlated with dislocation.

Materials and Methods

Subjects

All patients seen at our BPBP clinic undergo a routine shoulder ultrasound to evaluate them for the development of glenohumeral dysplasia. As a result of the wide range of ages at the time of presentation to the clinic, there is no standard age at which these ultrasounds are obtained. All patients also undergo a standardized physical examination at each clinic visit with use of the Toronto Score[8] and Active Movement Scale (AMS)[9] as well as measurement of the passive range of motion throughout the upper extremity. The frequency of visits during the first year depends on the injury severity and ranges from

TABLE I Demographics of Study Subjects

Variable	No. *(%)*
Sex	
Male	29 (44%)
Female	37 (56%)
Side	
Left	32 (48%)
Right	34 (52%)
Narakas grade of injury	
1 (upper trunk palsy)	41 (62%)
2 (extended upper trunk palsy)	14 (21%)
3 (global palsy)	3 (5%)
4 (global palsy with Horner syndrome)	8 (12%)

TABLE II Number of Ultrasound Evaluations for Each Patient

	Internal Rotation Ultrasound	External Rotation Ultrasound
No. (%) of patients with:		
1 ultrasound	30 (45%)	25 (47%)
2 ultrasounds	25 (38%)	18 (34%)
3 ultrasounds	7 (11%)	6 (11%)
4 ultrasounds	3 (5%)	3 (6%)
5 ultrasounds	1 (2%)	1 (2%)
Total no. of patients	66 (100%)	53 (100%)
No. of ultrasounds		
Total	118	96
Average	1.79	1.81

monthly to every 3 months. Whether additional ultrasounds are performed after the initial ultrasound is at the discretion of the treating provider.

This study was a retrospective record review of all infants who underwent at least 1 ultrasound evaluation for glenohumeral dysplasia prior to the age of 1 year. Subjects were excluded after they had reached 1 year of age or had had any procedure to treat the glenohumeral joint, such as Botox injection, application of spica casts, or surgical intervention. The age limit of 1 year was chosen because progressive ossification of the humeral head makes interpretation of ultrasound increasingly difficult after that age.

Physical Examination Measurements

We retrospectively recorded details of the standardized physical examination, including the AMS score and passive range of motion of the shoulder, as they were reported during each clinic visit. The passive shoulder external rotation was recorded with the arm in adduction at the side (passive external rotation in adduction) as well as with the arm in 90° of abduction (passive external rotation in abduction). Several subscores of the AMS were calculated, included the total shoulder score (sum of the AMS scores for shoulder flexion, abduction, adduction, external rotation, and internal rotation), shoulder rotation score (sum of the AMS scores for shoulder external rotation and internal rotation), and the difference between the AMS scores for internal rotation and external rotation.

Ultrasound Technique

All ultrasounds were performed by an experienced ultrasound technician, with use of a previously described technique[10], and supervised by a pediatric musculoskeletal radiologist. Still images saved at the time of each examination, with the shoulder position labeled as *internal rotation* or *external rotation,* were measured directly by us to calculate the PHHD (percentage of the humeral head displaced posterior to the axis of the scapula) and the alpha angle (intersection of the posterior scapular margin with a line tangential to the humeral head through the glenoid) for the purposes of this study (Fig. 1). These previously reported ultrasound measurements have been shown to have high interrater and intrarater reliability[7]. We defined glenohumeral dislocation as both a PHHD of >0.50 and an alpha angle of >30° because this is the point at which procedural intervention for the shoulder is considered in our practice. One of us performed all of the ultrasound measurements without knowledge of the corresponding physical examinations, while another two of us recorded all physical examination data from the medical record.

Statistical Methods

Physical examination results were compared between infants demonstrating dislocation at any point during the first year of life and infants with no evidence of dislocation. A mixed model with a random subject effect was used to account for clustering in these analyses, as most subjects had >1 set of physical examination results. We used receiver operating characteristic (ROC) curve analysis to illustrate the association between physical examination and concurrent ultrasound results, with the area under the ROC curve (AUC) as a measure of how well the physical examination result discriminates between dislocated and nondislocated shoulders. The exact method was used to calculate 95% confidence intervals (CIs) for sensitivity and specificity. All p values are 2-sided, and p values of <0.05 were considered significant.

This work was approved by the institutional review board at our institution. Subject consent was not required as this was a retrospective study.

Results

Prevalence of Shoulder Dislocation

Sixty-six subjects, born between January 2011 and April 2014, had at least 1 ultrasound study with the shoulder positioned in internal rotation during the first year of life (Table I). Of these, 53 also had at least 1 ultrasound with the shoulder positioned in attempted external rotation. Because this positioning was done by the ultrasound technician, we cannot be certain how externally rotated the shoulder was at the time of the imaging. Most subjects had >1 ultrasound during their first year, with a total of 118 internal rotation ultrasounds and 96 external rotation ultrasounds available for review (Table II) after 14 internal rotation ultrasounds of 12 patients and 13 external rotation ultrasounds of 11 patients had been excluded because they were obtained after the patient had undergone an intervention. Of the 66 subjects, 19 (29%) met the criteria for dislocation on at least 1 internal rotation ultrasound; 19 (36%) of the 53 patients who underwent ultrasound in external rotation met criteria for dislocation on at least 1 external rotation ultrasound. There was substantial overlap between the subjects identified as having dislocation in internal rotation and those identified as having it in external

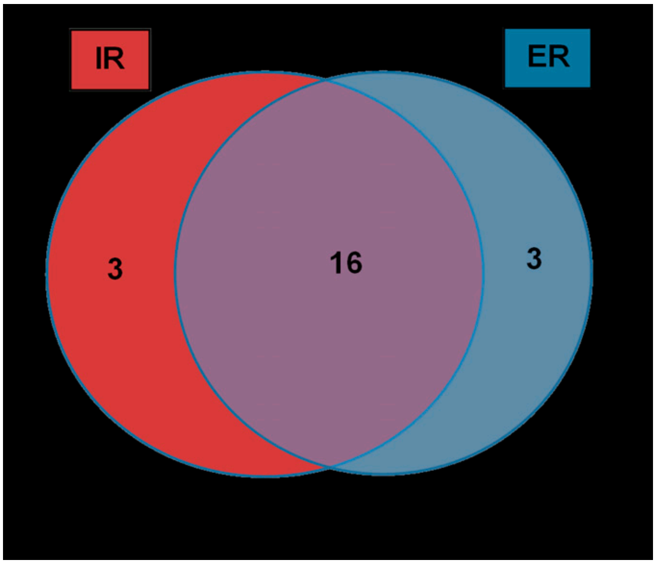

Fig. 2

Twenty-two subjects demonstrated glenohumeral dislocation on ultrasound with the shoulder positioned in internal rotation (IR) and/or external rotation (ER), with substantial overlap between groups.

TABLE III Prevalence of Dislocation Seen on Ultrasound According to Age

	No. of Ultrasounds in Age Group/No. Showing Dislocation (% Showing Dislocation)			
	0 to 91 Days Old	92 to 182 Days Old	183 to 273 Days Old	274 to 365 Days Old
Dislocation in internal rotation	1/17 (6%)	14/38 (37%)	4/35 (11%)	5/28 (18%)
Dislocation in external rotation	1/15 (7%)	13/32 (41%)	6/28 (21%)	4/21 (19%)

rotation, but they were not the exact same 19 subjects (Fig. 2). A total of 22 infants demonstrated dislocation on either internal rotation and/or external rotation imaging. As these groups were not statistically different and imaging is easier to obtain with the shoulder in its resting position of internal rotation, the data presented here refer to the 19 infants who demonstrated a dislocation in internal rotation. The median age at the ultrasound diagnosis of dislocation was 161 days (range, 63 to 318 days) for the internal rotation group and 180 days (range, 64 to 318 days) for the external rotation group. Fourteen subjects (74%) in each group demonstrated the dislocation on their first ultrasound, whereas the remainder were diagnosed on the basis of either their second (4 subjects; 21%) or third (1 subject; 5%) ultrasound. All available ultrasounds were reviewed to determine the prevalence of dislocation detected on ultrasound according to age (Table III).

Physical Examination Findings Associated with Ultrasound Evidence of Dislocation

Physical examination measurements at each time point were compared between infants who had demonstrated dislocation during their first year of life and those who had not (Table IV). Subjects who underwent a physical examination and ultrasound on the same date were included in this portion of the analysis. The physical examination measurements that showed the strongest associations with detection of dislocation on ultrasound were the difference between the AMS subscores for shoulder internal rotation and shoulder external rotation and passive shoulder external rotation in adduction. These associations were similar regardless of whether internal rotation or external rotation ultrasounds were used to identify dislocation. ROC curves were created for these 2 physical examination measurements to illustrate how well they could discriminate

TABLE IV Association of Physical Examination Findings with Dislocation Seen on Ultrasound

	Internal Rotation Ultrasound			External Rotation Ultrasound		
	No.*	Mean (Stand. Dev.)†	P Value	No.*	Mean (Stand. Dev.)†	P Value
Total AMS score			0.26			0.50
Dislocation	33	5.0 (1.0)		31	5.1 (1.0)	
No dislocation	76	5.5 (1.3)		63	5.4 (1.3)	
Total shoulder AMS score			0.008			0.05
Dislocation	34	3.9 (1.1)		32	3.9 (0.9)	
No dislocation	76	4.8 (1.5)		63	4.5 (1.5)	
Shoulder rotation AMS score			0.002			0.02
Dislocation	35	3.7 (1.4)		33	3.8 (1.1)	
No dislocation	78	4.9 (1.7)		64	4.6 (1.8)	
Difference between internal and external rotation AMS scores			<0.001			<0.001
Dislocation	35	5.5 (2.3)		33	5.6 (1.9)	
No dislocation	78	3.3 (2.7)		64	3.5 (2.8)	
Passive external rotation in adduction			<0.001			<0.001
Dislocation	25	45.8 (33.2)		23	43.9 (34.8)	
No dislocation	53	71.4 (19.9)		46	70.5 (18.4)	
Passive external rotation in abduction			0.08			0.03
Dislocation	22	76.1 (23.8)		22	75.7 (23.8)	
No dislocation	47	86.4 (10.7)		41	87.3 (8.4)	

*The sample sizes are based on the number of subject visits with a complete data set available, which varies according to the variable being analyzed. †The values are given in points for the AMS variables and in degrees for the passive external rotation variables.

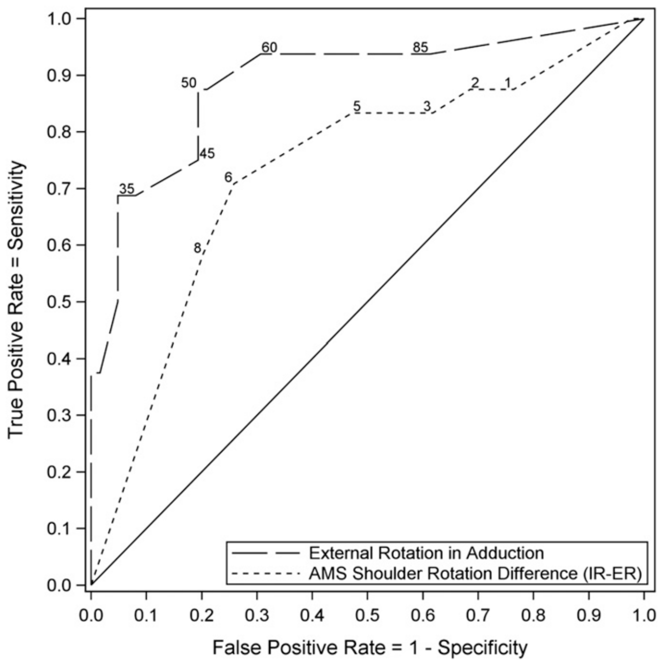

Fig. 3

ROC curves for discriminating between dislocated and nondislocated shoulders on ultrasounds performed with the shoulder in internal rotation. The curves show true-positive and false-positive rates at each cut-point of the physical examination measurement, with selected cut-points labeled. For example, cut-point 60 for external rotation in adduction represents ≤60° versus >60°; cut-point 2 for the difference between the AMS scores for internal rotation (IR) and external rotation (ER) represents a difference of ≥2 points versus a difference of <2 points.

between the dislocated and nondislocated shoulders. Figure 3 shows the ROC curves for internal rotation ultrasounds; results were similar for external rotation ultrasounds. The AUC for the difference between internal rotation and external rotation AMS subscores was 0.73 (95% CI = 0.61 to 0.85) for ultrasounds done in internal rotation and 0.74 (95% CI = 0.63 to 0.85) for ultrasounds done in external rotation. The AUC for passive external rotation in adduction was 0.89 (95% CI = 0.78 to 0.99) for internal rotation ultrasounds and 0.85 (95% CI = 0.72 to 0.98) for external rotation ultrasounds. Thus, passive external rotation in adduction was better for discriminating between dislocated and nondislocated shoulders.

The estimated sensitivity of using passive external rotation in adduction of ≤50° to identify glenohumeral dislocation on internal rotation ultrasound was 14 of 16, or 87.5% (95% CI = 61.7% to 98.4%). Specificity was 50 of 62, or 80.6% (95% CI = 68.6% to 89.6%). Increasing the cut-point from 50° to 60° yielded a sensitivity of 15 of 16, or 93.8% (95% CI = 69.8% to 99.8%), and a specificity of 43 of 62, or 69.4% (95% CI = 56.3% to 80.4%).

Discussion

The natural history of infantile dislocation is progressive shoulder dysfunction with limitations in abduction, external rotation, and overhead activity[3,11]. The 5 subjects with infantile dislocation in the 1998 study by Waters et al. had severe limitations in shoulder function at an average age of 1.9 years[3]. The literature regarding when and how to best treat infantile glenohumeral dysplasia associated with BPBP is limited, but earlier treatment is likely to be more effective. Closed reduction of the shoulder under anesthesia along with onabotulinum toxin-A injection into the internal rotators and use of shoulder spica casts have been proposed as a treatment that may be more successful in younger infants[12]. Similarly, it has been shown that extra-articular tendon transfers are more likely to lead to remodeling of the glenohumeral joint if they are done before 2 years of age[13]. After using a combination of arthroscopic shoulder reduction and extra-articular tendon transfers, Pearl and colleagues found that glenohumeral joint remodeling was better in children <4 years of age than in older children[14]. A reliable means for identifying infantile glenohumeral dislocation in the BPBP population would enable earlier treatment and potentially improved outcomes.

This study is not the first to investigate the use of ultrasound for identifying infantile shoulder dislocation. Moukoko and colleagues found an 8% prevalence of dislocation in a group of infants with BPBP who had been selected for ultrasound on the basis of physical examination findings[4]. Our study demonstrated a much higher rate of dislocation, which was found in approximately 1 in 3 infants presenting to our BPBP clinic. These numbers may differ for several reasons. First, the severity of the BPBP injury may differ between the studied populations. Second, our definition of dislocation likely included some cases that would have been considered dysplasia but not dislocation in the previous study, in which dislocation was defined as the ossification center of the humeral head being displaced behind the scapular line. We chose our definition of dislocation on ultrasound (a PHHD of >0.50 and an alpha angle of >30°) to identify those shoulders most at risk for progressive dysfunction and thus likely to be selected by us for early intervention. Finally, most infants in our study underwent >1 ultrasound examination during their first year of life, which improved our ability to identify infants who developed a dislocation later in their first year. Because we defined dislocation according to ultrasound findings, we do not know if any subjects had a shoulder dislocation not evident on ultrasound. Further study is necessary to compare ultrasound findings with those of other modalities, such as MRI, or intraoperative findings to understand if ultrasound could fail to identify a dislocated shoulder.

The important question of when to best identify infantile shoulder dislocations was addressed by Pöyhiä and colleagues[15], who found posterior glenohumeral joint subluxation (defined as an alpha angle of >30°) in one-third of infants with permanent BPBP. In their study, 24 infants had shoulder ultrasound performed at 1, 3, 6, and 12 months of age; 5 of the 9 cases of glenohumeral joint subluxation were identified at 3 months of age and 8 of the 9, by 6 months of age. The authors thus concluded that ultrasound screening was best utilized in the 3 to 6-month age group. Although imaging in the current study was not performed at as regular intervals as those in the study by Pöyhiä et al., our results support their findings. Our reported prevalence of dislocation of 29% is very similar to theirs (one-third of infants with permanent BPBP), and most of our cases were identified between 3 and 6 months of age as well.

This study has several limitations. First, it is a retrospective review of clinical practice. Although repeated ultrasounds at regular intervals were recommended, most patients had only 1 or 2 ultrasounds; thus, our data on the exact timing of infantile dislocation are limited. Similarly, the timing and frequency of the ultrasound examinations, which were decided by the treating physician and the parents, and the exclusion of data from time points subsequent to intervention may have created selection biases that make age-specific prevalence estimates difficult to interpret. It also must be remembered that our patients were seen at a tertiary care referral center for BPBP, so our findings cannot be extrapolated to infants with transient neurapraxias after birth. Finally, the relatively small sample size limits the statistical precision of our estimates. In particular, the CI widths for the sensitivity estimates are wide as a result of the small number of positive ultrasounds with concurrent physical examination measurements. We attempted to minimize bias by having two members of our research team review patient records and a different member measure the ultrasounds. However, as this was a retrospective study, clinicians treating the patients and performing the physical examination may have had access to the ultrasound images at the time of their physical examinations.

The results presented here can advance our understanding of the use of ultrasound for infants with BPBP in 2 main ways. First, our finding of only minor differences between the results of the internal rotation and external rotation ultrasounds suggests that static ultrasound can be done with the shoulder in the position that is most comfortable for the infant. Second, our quantification of the relationship between shoulder dislocation and various physical examination measurements showed that passive shoulder external rotation in adduction was the best physical examination parameter for detecting dislocation, with an AUC of 0.89. Using a cutoff of 60° of passive external rotation in adduction identified infants with dislocation with 94% sensitivity and 69% specificity. We contend that routine shoulder ultrasound is appropriate for infants with permanent BPBP because of the high prevalence of shoulder dislocation, and we agree with prior investigators[15] that 3 to 6 months is likely the best age for this imaging. We suggest that, if physical examination is used to identify which infants should undergo shoulder ultrasound, passive external rotation in adduction of ≤60° should be the criterion for performing the ultrasound. ■

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