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Original Research

Prevalence of Posterior Shoulder Subluxation in Children With Neonatal Brachial Plexus Palsy After Early Full Passive Range of Motion Exercises

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Abstract

Background: Children with neonatal brachial plexus palsy (NBPP) are often prescribed shoulder range of motion (ROM) exercises; however, the extent and timing of exercise implementation remains controversial in the context of shoulder joint integrity. The association of ROM exercises to delayed posterior shoulder subluxation (PSS) is unknown.
Objective: To determine prevalence of PSS in children with NBPP who began full passive ROM exercises before 6 months of age, and characteristics associated with development or absence of PSS in children.
Design: Cross-sectional study.
Setting: Tertiary care NBPP referral center.
Participants: Forty-six children with NBPP, aged 24-57 months, who began full ROM exercises before 6 months of age.
Methods: One radiologist conducted bilateral shoulder ultrasound (US) on each child to evaluate for PSS. One occupational therapist evaluated each child clinically for PSS using defined parameters without knowledge of US results.
Main Outcome Measures: By US, 20% of children had PSS; 46% had PSS by clinical examination. Shoulder active ROM limitations and history of shoulder surgery were associated with presence of PSS. Extent of NBPP was not associated with PSS.
Results: Nine of 46 children (20%) met US criteria for PSS; α angle was $58^\circ \pm 21^\circ$ (mean $\pm$ standard deviation [SD]). Twenty-one children (46%) met clinical criteria. Mean age at examination was 35 ± 10 months. Shoulder active ROM ($P \leq .004$) was associated with PSS, whereas passive ROM was not ($P \geq .08$). History of secondary shoulder surgery and primary nerve graft repair were associated with PSS ($P = .04$). Extent of NBPP by Narakas classification was not associated with PSS ($P = .48$).
Conclusions: Early use of full-arc passive ROM home exercise program is not associated with increased prevalence of PSS in children with NBPP compared to prevalence of PSS in published literature. We suggest careful clinical examination, based on defined criteria, provides a reasonable screening examination for evaluating PSS that can be confirmed by noninvasive US.

Introduction

Posterior shoulder subluxation (PSS) associated with neonatal brachial plexus palsy (NBPP) can result from musculoskeletal deformities of the shoulder joint due to persistent muscle weakness or imbalance. In the 0.4-4.6 per 1000 live births affected by NBPP [1-5], PSS has a reported prevalence of 25% in children with a mean age of 47 months [6]; likewise, the reported prevalence of osseous deformity is 33% in children with a mean age of 43 months [7]. Persistent PSS leads to structural changes in the glenohumeral joint, resulting in decreased function and pain in the upper extremity and an increased need for surgical intervention [8-12]. Detection of PSS in the clinic comprises serial examinations of passive ROM [13,14], including external rotation of the shoulder; however, the sensitivity and specificity of this screening examination has not been determined [15,16]. Consequent confirmation of PSS has been reported using MRI [12-14,17-23], CT [23-27], arthroscopy [28], and more recently ultrasonography [15,16]. Prevention of PSS

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and subsequent osseous deformity may rely in part upon the regular performance of joint range of motion (ROM) exercises. However, both the arc of ROM (full versus limited) exercises at the shoulder and the timing of initiation of these exercises remain controversial with regard to resultant shoulder joint integrity. Neither consistent nor definitive published data exist to support or refute the use of early full ROM exercises at the shoulder with respect to the development of PSS in NBPP children. Based on anecdotal evidence, controversy exists with regard to an increase in the prevalence of PSS with early full ROM exercises. Therefore, our goal was to determine the prevalence of PSS in children who initiated full-arc ROM exercises before 6 months of age by radiologic and clinical criteria, and to compare the prevalence with historical cohorts who underwent undefined shoulder ROM exercises.

Methods

Using defined ultrasound (US) and clinical criteria, we performed a cross-sectional examination of shoulder integrity in children with NBPP aged 24-57 months, who initiated full-arc ROM exercises before 6 months of age. Approval of our study protocol was granted by our university institutional review board.

Participants

Sixty-one children with the diagnosis of NBPP, 24-57 months of age, who sequentially presented to our brachial plexus clinic from May 2011 to July 2013 were recruited for participation in this study. Forty-six children met the inclusion criteria; these children had begun full passive ROM (PROM) exercises before 6 months of age, with compliance reported via written survey. Exclusion criteria comprised home performance of non—full-arc ROM exercises, late (>6 months) initiation of full ROM exercises, other diagnoses conferring difficulties in arm movement or musculoskeletal abnormalities, and inability to attend scheduled clinic or radiographic appointments. One of 2 occupational therapists demonstrated the full PROM exercises to the children, and each patient received a DVD entitled "Home Exercise Therapy Program for Brachial Plexus Palsy" [29,30] to support the prescribed regimen of home exercises (at least 4-5 times per day). Standard clinical measurements included active ROM (AROM), PROM, manual muscle testing using Medical Research Council (MRC) scoring [31], limb length/circumference, self-reported presence or absence of pain, Narakas classification [32], modified version of Birch classification of shoulder deformity scoring [33], and acquisition of developmental milestones.

Determination of PSS by Ultrasound Examination

A single pediatric radiologist with >33 years of experience performed the shoulder US evaluation with an 8- to 12-MHz linear array transducer in an axial plane using a posterior approach [16,34], with the child seated on a stool. Static and dynamic motions and angles of the shoulder joint—during internal/external rotation with shoulder in adduction (with arm stabilized at side, elbow flexed at 90°, palm up), and shoulder internal/external rotation in abduction (arm supported in 90° of abduction, elbow flexed at 90°, forearm pronated), in forward flexion—were recorded by the radiologist, while the occupational therapist positioned the child within the child's available range of motion (Figure 1). To determine the α angle, a horizontal line was drawn along the posterior margin of the scapula, and another line drawn tangential to the humeral head, intersecting at the posterior glenoid labrum (just lateral to the spinoglenoid notch) (Figure 2) [16,35]. PSS was determined to be present if the α angle was $\geq$30° [16,36].

Determination of PSS by Clinical Examination

A single occupational therapist with >8 years of NBPP experience evaluated each child for PSS with a careful clinical examination using a goniometer, a standard flexible tape measure, the modified criteria from Moukoko et al [15] (criteria 1-4), and additional clinical criteria that we deemed relevant (criteria 5-8), as follows: 1) asymmetry of axillary and humeral skin folds; 2) discrepancy of >1 cm in length of humeral segment;

Figure 1. Radiologist-directed ultrasound using a posterior approach, and an occupational therapist assisting with performance of static and dynamic motion of shoulder forward flexion.

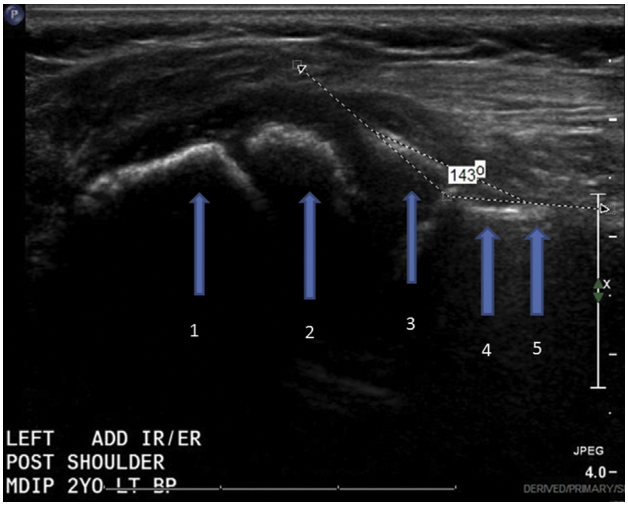

Figure 2. Ultrasound image of posterior shoulder subluxation. Arrow 1: ossified humeral diaphysis and metaphysis; arrow 2: partially ossified humeral head; arrow 3: cartilaginous glenoid labrum; arrow 4: spinoglenoid notch (contains suprascapular nerve); arrow 5: scapula. The α angle is 37° (180° − 143° = 37°).

3) asymmetric fullness of the posterior shoulder using "palpation [to] identify changes in the symmetry of the anterior and posterior shoulder regions"[15]; 4) PROM ≤50° of shoulder external rotation in adduction; 5) posterior glenohumeral joint angle ≤55° (the angle between the scapular crest and the humerus when the arm is positioned in forward flexion of 90° and full horizontal adduction) [25]; 6) AROM ≤90° of forward shoulder flexion; 7) AROM ≤50° of external rotation in adduction; and 8) AROM ≤50° of external rotation in abduction. PSS was determined to be present if any 5 of the 8 criteria were met. Criteria for the clinical determination of PSS were chosen based on the combined clinical experience of occupational therapists of >20 years in the context of relevant published literature [13,15,16,25].

Finally, child caretakers completed a brief survey reporting compliance with the prescribed full PROM home exercises.

Statistical Analysis

Participant demographics were summarized using descriptive statistics. Numerical variables (age, degree of AROM, PROM, and posterior glenohumeral joint angle) were compared for bilateral shoulders among subluxation and nonsubluxation groups by Student *t*-test and categorical variables by χ^2 or Fisher exact test. Median Birch classification scores were compared using 1-way analysis of variance. We explored the correlation between US and clinical impression of shoulder subluxation using Pearson's correlation. We used sensitivity and specificity to compare the relationship of the clinical impression of PSS with the US evaluation. Sensitivity is defined as the probability that a child had shoulder subluxation based on clinical impression and then confirmed by US study (true positive/true positive+false negative); specificity is the probability that the clinical impression indicates a child did not have shoulder subluxation and US confirmed that a child did not have shoulder subluxation (true negative/true negative+false positive). Analyses were performed using SPSS software, version 20 (SPSS Inc., Chicago, IL), with statistical significance established at $P < .05$.

Results

Nine study participants (20%) met the US criteria for PSS. The mean α angle for children with PSS was 58° ± 21° (mean ± SD) compared to <30° for those without US-diagnosed PSS. Mean age at the time of study participation was 34 ± 10 months. As expected, a demographic variable that demonstrated a significant difference between the subluxed and nonsubluxed groups was history of shoulder surgery via tendon transfer and/or muscle lengthening for external rotation ($P = .04$); by nature, children undergoing surgical intervention at the shoulder had more severe early glenohumeral dysfunction. No significant differences were found between children with and without PSS in regard to gender, involved extremity, race/ethnicity, age at time of US, Narakas classification, Birch classification, fracture history, primary nerve surgery, pain, or developmental achievement of crawling (Table 1).

Using clinical assessment alone, 46% (n = 21) of study participants met the criteria for PSS. We evaluated the relationship between radiographic and clinical determination of PSS using the Pearson correlation and found highly significant correlation ($R = 0.45$; $P = .002$). When compared to US examination, clinical examination determined PSS with a sensitivity of 100% and specificity of 68%. Regarding clinical evaluation alone for PSS, the AROM for shoulder forward flexion ($P = .001$), abduction ($P = .004$), external rotation in adduction ($P = .001$), and external rotation in abduction ($P < .0001$) were significantly greater in children without PSS when compared to those with PSS as determined by US; of note, no significant difference in PROM ($P > .1$) existed between the PSS and non-PSS groups for any shoulder movements or angles (Table 2).

Regarding the 8 clinical criteria for determining PSS, 2 of the 8 parameters were significantly different between the PSS and non-PSS groups (as determined by US criteria), namely, active shoulder forward flexion ($P = .0006$) and active external rotation in abduction ($P \leq .0001$). Passive external rotation in adduction ($P = .068$) and asymmetry of skin folds ($P = .071$) demonstrated a trend toward significance. The remaining 6 PSS screening parameters included active external rotation in abduction ($P = .090$), humeral length difference ($P = .132$), posterior shoulder fullness ($P = .102$), and posterior glenohumeral joint angle

Table 1
Patient demographics at time of ultrasound examination

Parameter	Total patients (N = 46)	Subluxed shoulder (n = 9)	Nonsubluxed shoulder (n = 37)	*P*
Mean age ± SD (mo)	35 ± 10	34 ± 11	36 ± 10	.66*
Gender				.06†
Male	19 (41%)	1 (11%)	18 (49%)	
Female	27 (59%)	8 (89%)	19 (51%)	
Side				.26†
Left	25 (54%)	3 (33%)	22 (59%)	
Right	21 (46%)	6 (67%)	15 (41%)	
Child race				.96†
White	27 (59%)	6 (67%)	21 (57%)	
Black	12 (26%)	2 (22%)	10 (27%)	
Hispanic	1 (2%)	—	1 (3%)	
Mixed	6 (13%)	1 (11%)	5 (14%)	
Narakas classification§				.48†
I-II	21 (46%)	3 (33%)	18 (49%)	
III-IV	25 (54%)	6 (67%)	19 (51%)	
Clavicle or humerus fracture	6 (13%)	2 (22%)	4 (11%)	.58†
Shoulder pain	9 (20%)	3 (33%)	6 (16%)	.35†
Crawling	44 (96%)	8 (89%)	36 (97%)	.36†
Median Birch classification score (range)	0 (0-1)	1 (0-1)	0 (0-1)	.07‡
Primary nerve surgery	17 (37%)	5 (56%)	12 (32%)	.26†
Shoulder tendon transfer/muscle lengthening for internal rotation contracture or to increase external rotation	5 (11%)	3 (33%)	2 (5%)	.04†

SD = standard deviation.
* Student *t*-test applied for group comparison.
† χ^2 Test or Fisher exact test applied for group comparison.
‡ One-way analysis of variance applied for group comparison.
§ Narakas classification I-II is less involved (Erb's palsy group) and III-IV is more involved (panplexopathy group).

(*P* = .999), for which there was no significant difference between children with PSS and those without PSS (Table 3).

Discussion

We found the prevalence of PSS using US criteria to be 20% in NBPP children aged 37 ± 10 months who began full-arc ROM exercises before 6 months of age. This result is not inconsistent with the prevalence reported by Kambhampati et al [6], a 25% prevalence of shoulder subluxation in children at an average age of 47 months (range, 3-204 months). Regarding frank osseous deformity, 33%-80% of NBPP children aged 12 months to 14 years demonstrate glenohumeral joint deformity [5,7,37], but not all osseous deformities are associated with PSS. For example, history of humeral and/or clavicle fractures in the perinatal period was not associated with PSS [38,39]. Furthermore, the reported prevalence of PSS in later childhood may decrease with increasing age (64% prevalence at 6.1 years of age [40] and 16% prevalence at 13.5 years of age [41]). These reported values for prevalence of PSS occurred in children performing variable shoulder exercises with regard to arc and timing of initiation. Consequently, we suggest that

Table 2
Comparison of degree of passive vs active range of motion as determined by ultrasound examination in patients with or without shoulder subluxation (mean ± SD)*

Parameter	Total patients (N = 46)	Subluxed shoulder (n =9)	Nonsubluxed shoulder (n = 37)	*P*
Passive range of motion,°				
Shoulder flexion	171 ± 15	162 ± 28	174 ± 9	.23*
Shoulder abduction	171 ± 17	158 ± 30	174 ± 12	.17
External rotation adduction	74 ± 26	57 ± 31	78 ± 23	.08
External rotation abduction	84 ± 19	72 ± 27	87 ± 15	.14
Gleno-humeral posterior angle	54 ± 9	51 ± 9	55 ± 8	.33
Active range of motion,°				
Shoulder flexion	131 ± 46	88 ± 33	141 ± 43	.001
Shoulder abduction	127 ± 49	81 ± 44	138 ± 43	.004
External rotation adduction	22 ± 54	−31 ± 38	34 ± 50	.001
External rotation abduction	49 ± 44	−11 ± 30	64 ± 32	<.001

* Independent *t*-test applied for group comparison.

Table 3
Eight parameters of clinical examination used to screen for posterior shoulder subluxation that can be confirmed via ultrasound

Clinical assessment	Subluxed shoulder (n = 9)	Nonsubluxed shoulder (n = 37)	*P* *
Exorotation adduction, ≤50° (passive)	56%	19%	.068
Humeral length difference, ≥1 cm	67%	32%	.132
Asymmetry skin folds	100%	62%	.071
Posterior shoulder fullness	78%	41%	.102
Posterior glenohumeral angle, ≤55°	67%	59%	.999
Shoulder flexion, ≤90° (active)	89%	22%	.0006
External rotation adduction, ≤50° (active)	89%	51%	.090
External rotation abduction, ≤0° (active)	100%	11%	<.0001

* χ^2 Test was applied for group comparison.

performance of early full-arc ROM exercises is not associated with an increased prevalence of PSS.

Of note, the extent of nerve root involvement in NBPP (Narakas classification: group I = C5-6, group II = C5-7, group III-IV = C5-T1) is not significantly associated with the presence of PSS and is consistent with reports in the literature denying a significant relationship between the Narakas classification groups and shoulder deformities [38-41]. However, some investigators do report a higher prevalence of shoulder deformity in Narakas group I-II children, suggesting that global muscle weakness in Narakas group III-IV produces a lesser degree of muscle imbalance [20,24,42]. Because all Narakas groupings include injury of the C5 and C6 nerve roots (primary nerve root innervation to the shoulder muscles), we would not expect an association between higher Narakas groupings and the presence of PSS [39,43]. Despite the favorable prognostic outlook of 69%-90% functional recovery in Narakas groups I and II [44], we suggest that all practitioners be aware that PSS occurs with similar prevalence, regardless of the extent of nerve root injury in NBPP.

As we expected, history of musculoskeletal shoulder surgery was associated with PSS. Although this phenomenon may reflect a selection bias for clinically worse children, the need for musculoskeletal shoulder surgery is indicative of the severity (with which each nerve root is injured) rather than the extent (number of nerve roots injured) of the NBPP. Children who underwent both primary nerve reconstruction and secondary musculoskeletal operations had worse shoulder function than those children who underwent only primary nerve reconstruction [45]. All of our children who underwent musculoskeletal shoulder surgery also had a prior nerve reconstruction. Therefore, we suggest that healthcare providers be particularly attentive when screening for PSS in children who have undergone both nerve reconstruction and musculoskeletal shoulder surgery, as these children may be more likely to develop PSS because of the initial severity of the nerve injury.

With regard to developmental milestones, some authors have suggested that weight-bearing in a crawling position contributes to the development of PSS [46], but others contradict this suggestion [15]. In our study, 97% of children in the nonsubluxed group achieved crawling, which is consistent with the latter observation that weight-bearing in the crawling position is not likely associated with PSS. As symmetrical weight-bearing will foster appropriate postural control for later development [47], and as prone positioning may be the optimal position for prevention of internal rotation contractures in infants [2], we encourage supervised "belly to play" activities as part of the NBPP treatment paradigm.

Prevention of PSS begins with prevention of contracture formation and retention of optimal PROM, despite the muscle imbalances inherent to NBPP [4,48]. PSS can progress with age in diagnosed children, if the condition remains untreated [48,49]. Although the literature supports therapeutic interventions and home exercise programs inclusive of PROM exercises beginning as early as 7-10 days of life [4,13], the arc of motion, timing, and frequency of these exercises is barely described in the literature. Resultant shoulder function (PROM and AROM) is closely associated with the degree of deformation of the glenoid as well as with the extent of posterior humeral head dislocation [40], but our study demonstrates that AROM may be the more important factor associated with PSS. Because AROM is limited functionally by PROM, we support the use of early full-arc PROM exercises, consistent with that proposed by other investigators [4,43], with attention to scapular stabilization during external rotation movement [13,50]. Survey responses indicated that all families performed full-arc PROM exercises and that 78% of children (36 of 46) performed the exercises at a frequency of at least once daily. In addition, 48% of the children (22 of 46) participated in formal therapy programs, and 68% (15 of 22) reported a therapy frequency of at least once a week. Multimedia assistance has been associated with increased compliance and performance accuracy of at-home ROM exercises [29,30].

Various methods used to determine the presence or absence of PSS exist in the published literature [26,28,51]. We reviewed the available literature for other studies using US techniques and/or clinical assessments [15,16]. US is the preferred radiographic method with regard to cost, lack of invasiveness, lack of ionizing radiation exposure, and ability to perform dynamic measurements (reduction of subluxation with movement) and has been used successfully in children <1 year of age [15,16,34,36,52-54]. The intraobserver reproducibility of 0.91 and interobserver reliability of 0.875 for shoulder US has been reported by Vathana et al [36]. The differences reported in the prevalence of PSS among studies by Moukoko et al [15] and Poyhia et al [16] are likely attributable to the differences in child age and inclusion/exclusion of children with prior surgical history (Table 4). Mean α angle was not reported by Moukoko et al [15], but their results were similar to those of Poyhia et al [16]. Our clinical assessment for PSS using 8 criteria (of which the presence of 5 would suggest PSS) has 100% sensitivity and 68% specificity, compared to the 84% sensitivity for PSS reported by Poyhia et al when using only external rotation ROM [16]. Moukoko et al [15] did not indicate sensitivity or specificity, as the US was completed on only 11 of 134 children, in contrast to our study, which included all children. When compared to the criterion standard of a US examination, our findings indicate that the clinical examination is a justifiably low-cost but highly sensitive means of screening for PSS. Clinical examination might be effective for detection of PSS and could prevent unnecessary recommendations for radiological imaging.

Given the potential implications and progression of undiagnosed (untreated) PSS, a careful clinical screening examination with 100% sensitivity is desirable. We developed the 8-criteria clinical examination protocol based on literature reports and clinical experience. We used 4 of the 5 criteria reported by Moukoko et al [15]; however, we excluded screening for the presence or absence of the "click" with shoulder movement, as we could not reliably detect the "click." We added 4 other criteria to assist us with our clinical screening for the presence of PSS, which includes the posterior glenohumeral joint angle, AROM of forward shoulder flexion (≤75°), AROM of external rotation in adduction (≤50°), and AROM of external rotation of abduction (≤50°). We focused on criteria reflecting the AROM, supported by the lack of statistically significant difference in PROM, but a significant difference in AROM in our study and others [24]. In contrast, traditional examination of children to determine PSS primarily comprised parameters reflecting the loss of PROM (particularly external rotation in adduction [16], as demonstrated with the Birch classification [33] and somewhat in abduction. However, we suggest that the strength of the 8-point clinical method for determining PSS relies on AROM, particularly active forward shoulder flexion, active external rotation in abduction [51], and less with active external rotation in adduction [40], asymmetry of the skin folds [15], and passive external rotation in adduction [40]). Overall, using a simple clinical screening examination for detection of PSS can direct those with clinically suspicious PSS toward the appropriate treatment to improve overall outcomes after NBPP.

Study Limitations

The limitations of this study include the single-institution data (tertiary care NBPP referral center), sample size, and the use of a single cohort study. Our study did not incorporate a typically developing cohort, nor did other studies have a typically developing cohort when using US as a method to screen for PSS. A randomized study that would deny the use of full PROM was not instituted, as we did not want to withhold treatment from these children. Maturation of the skeletal system, in particular the humeral head, can result in ossification that can obscure imagery of the US. Although this is a potential limitation, we supplemented the posterior approach with techniques used for the examination of the adult rotator cuff and

Table 4
Comparison of study results for ultrasound procedures used to determine posterior shoulder subluxation

Ultrasound parameter	Current study	Moukoko et al [15]	Poyhia et al [14]
Total no. of patients in ultrasound study	46	11	96
Mean age at time of PSS diagnosis (range)	34 (24-57)	6 (3-10)	6 (3-12)
Median Narakas classification (range)	3 (1-4)	1 (1-4)	2 (1-4)
Patients with PSS, by age group, mo			
<12 mo	—	11 (8%)*	19 (20%)
>36 mo	9 (20%)	—	—
Clinical examination correlates with ultrasound			
Sensitivity	100%	NA*	84%
Specificity	68%	NA*	NA
Mean posterior subluxation,° (range)	58° (34°-83°)	NA	49° (36°-78°)
Passive range of motion exercises	Yes; full	Yes; unsure arc of motion	Yes; unsure arc of motion
Included patients with primary nerve graft	Yes	No	Yes
Included patients with shoulder surgery	Yes	No	Yes

PSS = posterior shoulder subluxation.
* In the Moukoko et al study, ultrasound was conducted based on the clinical impression of shoulder subluxation; therefore 11 of 134 patients underwent ultrasound evaluation.

identification of the suprascapular notch and spinoglenoid notch [55-57].

Conclusions

PSS is a significant but undesirable consequence of NBPP. Prevention of PSS, encouraging appropriate formation of the glenohumeral joint, and halting progression of PSS relies on maintaining muscle balance and ROM at the shoulder joint. In this regard, we demonstrate that the use of an early, full-arc PROM home exercise program is not associated with an increased prevalence of PSS in children with NBPP. In addition, we suggest that health care providers use both history (prior nerve reconstruction or musculoskeletal surgery, regardless of the extent of NBPP) and a simple clinical assessment (8-criteria tool) when diagnosing PSS in the outpatient setting, which can be confirmed with a facile, noninvasive US study.

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