Journal of Nursing Science
2019; 5(2): 6-16
http://www.aascit.org/journal/jns
ISSN: 2381-1056 (Print); ISSN: 2381-1064 (Online)

Hemiplegic Shoulder Pain in Stroke Patients: An Integrative Review

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Citation

Yufen Lin, Zheng Li, Dan Shi, Yiting Chen. Hemiplegic Shoulder Pain in Stroke Patients: An Integrative Review. *Journal of Nursing Science.* Vol. 5, No. 2, 2019, pp. 6-16.

Received: May 27, 2019; **Accepted**: August 7, 2019; **Published**: August 29, 2019

Abstract: Hemiplegic shoulder pain (HSP) is one of the most prevalent symptoms among stroke patients. The purposes of this integrative review were to explore multidimentional descriptions of this symptom, identify associated factors and describe the outcomes of hemiplegic shoulder pain. A literature search was conducted using MEDLINE, Embase and CINAHL from database inception through January 2019. The literature search initially yielded 2,791 articles, including 25 quantitative studies that met inclusion criteria. Three main categories were identified: the prevalence of HSP was dramatically varied from 19–72% and patients who experienced HSP varied in the dimensions of symptom experience (e.g. intensity, frequency); the factors with strongest support were impaired motor function, limited range of motion and sensation change; HSP decreased patients' quality of life and functional status, and increased duration of hospitalization. It suggests that accurate and comprehensive assessment and standardized diagnosis of HSP are necessary, symptom experience and its impact on quality of life need to be explored.

Keywords: Hemiplegic Should Pain, Symptom, Integrative Review

1. Introduction

Stroke is the leading cause of death or disability in the worldwide. Of more than 4.5 million stroke survivors alive in the United States today, 15% to 30% are permanently disabled [1]. In the recent past, an estimated 1.5–2 million new strokes occurred each year in mainland China [2]. Stroke has been proven to be the second largest cause of death for people in China [3]. Hemiplegic shoulder pain (HSP) has been described as one of the four most common medical complications after stroke, with others being depression, falls, and urinary tract infections [4]. Turner-Stokes and Jackson [5] reported the prevalence of HSP ranged from 5% to 84%. While this symptom has a wide range of estimated prevalence, investigators have found its high prevalence. A large population-based study of 1000 patients with stroke in Turkey demonstrated a prevalence of 54.8% for HSP [6]. The most common period of occurrence of HSP is at 8–10 weeks after stroke [7]. HSP can appear during the first week after stroke, or develop much later [8]. Although the etiology of HSP has not been clearly illustrated, three widely received mechanisms include impaired motor control (muscle tonus changes), soft-tissue lesions, and altered peripheral and central nervous activities [9]. The outcomes of HSP in stroke patients are serious and negative. With HSP undertreated, it can cause patients suffering from a reduced quality of life, poor functional recovery, depression, disturbed sleep, and prolonged hospitalization [9-10].

To date, limited original studies reported the definition, predictors and consequences of HSP. The lack of a universally accepted definition of HSP makes it impossible to create a standard diagnosis for HSP and difficult to synthesize related research results. Contributions to pain development are often multifactorial. The related factors of HSP proposed in previous studies included age, sex, hemisphere, glenohumeral subluxation, spasticity, range of motion limitation and prior shoulder pathology [11-12]. However, many of these factors have been suggested as contributing factors and the actual causes have not been well established. Complexities in factors create difficulties in the treatment of HSP, and literature reviews have revealed limited evidence to guide clinicians on effective treatment

options.

Hence, insight into the HPS symptom, exploiting valid measurements of HSP to support early identification of this symptom, and clarifying the influential factors of HSP would assist both clinicians and patients in finding appropriate management strategies. Therefore, the purposes of this review were 1) to describe HSP in the context of occurrence, definition, and symptom experience; 2) to analyze possible factors contributing to HSP after stroke; 3) to describe outcomes associated with HSP in stroke patients.

2. Methods

2.1. Literature Search

To obtain empirical studies on HSP after stroke, we conducted a comprehensive search of multiple databases from database inception through January 2019, including MEDLINE (OvidSP 1946-January 2019), Embase (OvidSP 1974-January, 2019), and CINAHL (1967-January, 2019) databases, using the search terms "shoulder pain", "hemiplegic", "stroke", and "cerebrovascular accident" to capture the meaning of HSP in post stroke patients. Because the search was intended to expose a large number of reports as well as the earliest literature on HSP among stroke patients, we applied no time limit on dates of the publication. Reference lists from relevant articles were cross-checked and pertinent journals were hand searched for articles.

The inclusion criteria were 1) studies that were limited to primary research studies, 2) written in English, 3) studies that involved HSP in the context of the post stroke period, described or defined the symptom of HSP, and addressed any aspect of features, factors or outcomes of HSP in stroke survivors. The exclusion criteria were 1) duplicates, review articles, unpublished manuscripts, electronic articles in non-journal formats, 2) studies that did not report HSP in their findings, 3) studies that discussed HSP in the non-stroke population only, 4) interventional studies.

2.2. Search Results and Data Evaluation

The initial search identified 2,791 publications. After eliminating duplicates and screening of titles, abstracts and key words, the full text of 80 articles was examined. Publications were screened by the first author according to the inclusion criteria and excluded 58 articles for the following reasons: 36 did not measure symptom, 16 were editorial or review articles and 6 were cases. In addition, 3 other articles were added through a manual search. The selection process is illustrated in Figure 1. The 25 articles were reviewed independently by two of the authors to assess for quality criteria [13]. If there was disagreement, it would be judged by the third reviewer. All of the 25 articles met the quality criteria and were included in the integrative review. The most important findings of the studies are presented in Table 1.

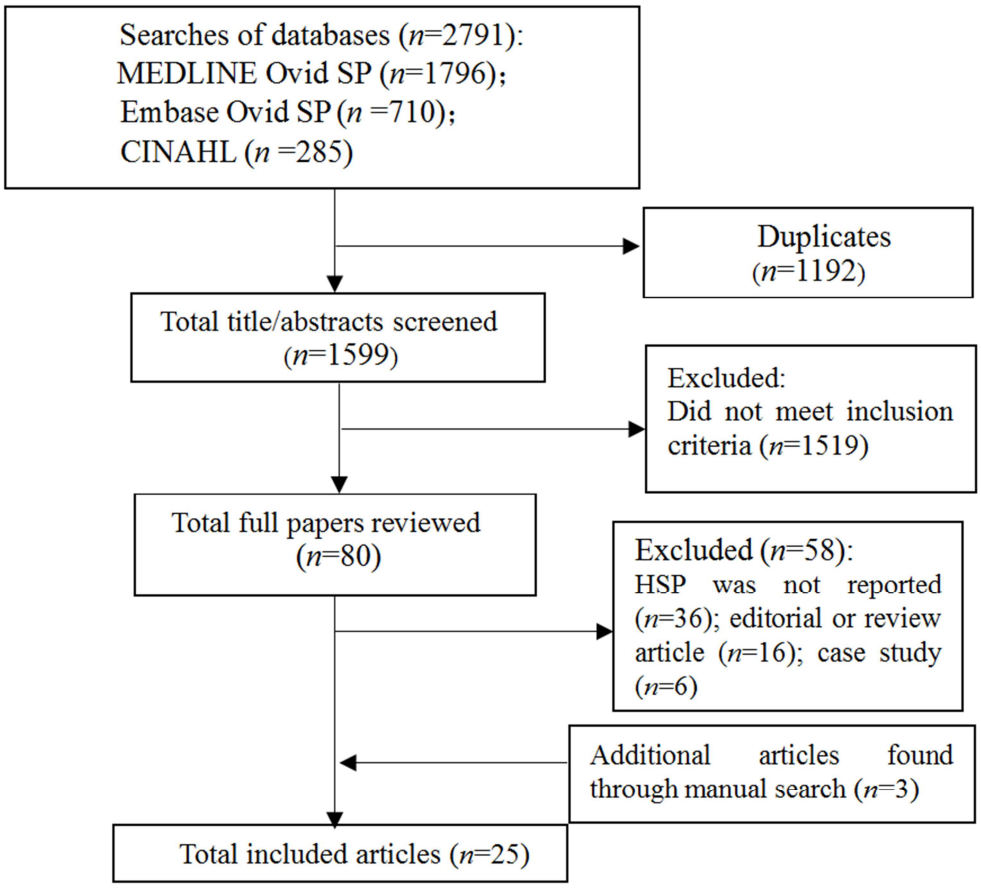

Figure 1. *Flowchart of the literature selection process.*

Table 1. *Summary of Literature Review Findings.*

Author, Year, Country	Design	Aim	HSP Evaluation	Sample	HSP Prevalence	Main findings
Adey-Wakeling et al. [15], 2015, Australia	Longitu-dinal	To provide the clinical profile, frequency, and determinants of poststroke HSP	intensity: VAS (0–100); time of onset; aggravating factors; ROM	Baseline, *n*=198 4mo, *n*=156 12mo, *n*=148 Total, *n*=226	19 (10%) 32 (21%) 31 (21%) 65 (29%)	At follow-up, pain was shown to be associated more with limited active and passive range of movement, and significantly fewer participants reported pain that was worse at rest or at night.
Adey-Wakeling et al. [35], 2016, Australia	Longitu-dinal	To determine the association of HSP with health-related quality of life at 12 months after first stroke	intensity: VAS (0–100); time of onset; aggravating factors	Baseline, *n*=300 4mo, *n*=226 12mo, *n*=195		HSP, depression, increased dependency, stroke severity, and absence of initial rehabilitation were each associated with reduction in quality of life.
Aras et al. [21], 2004, Turkey	Cross-sectional	To examine the occurrence of HSP in Turkish patients and clarify contributing factors	present/ absent; ROM	*n*=85	54 (64%)	Shoulder pain was significantly more frequent in subjects with reflex sympathetic dystrophy, lower motor functional level of shoulder and hand (p<0.001), subluxation, and limitation of external rotation and flexion of shoulder (p<0.05). Age was also a significant factor.
Barlak et al. [22], 2009, Turkey	Cross-sectional	To assess the possible causes of HSP, identify the correlation between HSP and clinical factors, and review the effects of HSP on functional outcomes	intensity: VAS (0–100); ROM	*n*=187	114 (61%)	No correlation was found between shoulder pain and clinical factors. The group without HSP showed significantly more improvement than the group with HSP in functional outcomes (P=0.01) and the hospitalization period was significantly shorter (p=0.03).
Blennerhassett et al. [31], 2010, Australia	Retro-spective	To determinewhat factors are associated with shoulder pain during stroke rehabilitation	Frequency: history documented; ROM	*n*=94	33 (35%)	Shoulder pain was reliably associated with two factors: reduced passive shoulder range (OR 14%, 95% CI 3 to 64), and Motor Assessment (OR 64%, 95% CI 43 to 96). The model accurately classified 85% of patients.
Chae et al. [18], 2007, USA	Cross-sectional	To assess the relationship between PSSP, upper-limb motor impairment, activity limitation, and pain-related QOL	intensity: NRS of the Brief Pain Inventory question 12 (BPI 12, rating 0–10); interference; ROM	*n*=61		Stepwise regression analyses indicated that poststroke shoulder pain is associated with the QOL (BPI 23), but not with the FMA, FIM, or AMAT scores.
Dromerick et al. [37], 2008, USA	Cross-sectional	To clarify the pathophysiology of HSP by abnormal shoulder physical diagnosis signs and the accuracy of self-report	intensity: VAS; location; radiation	*n*=46	17 (37%)	Self-reported pain was a poor predictor of abnormalities elicited on the examination maneuvers, even in those without neglect.
Gamble et al. [19], 2000, UK	Cross-sectional	To describe pain prevalence data for patients at various stages after stroke	intensity: VAS; time of onset; location; characteristics; bothersomeness	*n*=123	31 (25%)	There was a statistically significant association with ipsilateral sensory impairment (p<0.005), abnormal rheumatological examination (p<0.001) and depression score (p<0.005).
Gamble et al. [23], 2002, UK	Longitu-dinal	To describe the incidence of PSSP prospectively and identifies risk factors for developing pain	Intensity: VAS; time of onset; location; characteristics; bothersomeness	2wk, *n*=135 4mo, *n*=124 6mo, *n*=123	52 (40%)	There was a strong association between pain and abnormal shoulder joint examination, ipsilateral sensory abnormalities and arm weakness. Shoulder pain had resolved or improved at 6 months in 41 (80%).
Hadianfard et al. [17], 2008, Iran	Longitu-dinal	To find out the factors that can predict the HSP after stroke	intensity: VAS (0-10); time of onset; ROM	Six sessions (every two months) *n*=152	49 (32%)	The critical time for the occurrence of HSP was between 2 and 6 months. It showed that the best predictors for HSP were "Activity of daily living

Author, Year, Country	Design	Aim	HSP Evaluation	Sample	HSP Prevalence	Main findings
						ability", "Increased light touch threshold" and "Increased vibration threshold".
Karaahmet et al. [24], 2014, Turkey	Longitu-dinal	To analyze the incidence of and the factors associated with shoulder pain in people with hemiplegia	present/absent; time of onset	*n*=55, on admission, at discharge, and 1 month after discharge	34 (62%)	The major risk factors were disease duration and poor initial motor function. In both groups, the FMA, FAT, and FIM scores showed significant changes, but it did not differ between the two groups. It demonstrated that the incidence of depression was 36.4% and was not significantly associated with HSP.
Kashi et al. [34], 2018, Israel	Cross-sectional	To find out whether HSP is associated with deficient pain modulation.	intensity: NRS (0–10)	*HSP, n=16; no HSP, n=14; control, n=20*		HSP is related to the patients' deficient of pain adaptation ability. The magnitude of pain adaptation in the HSP group was significantly smaller than in either the non-HSP group (P<0.05) and the control group (P<0.01), and the insufficient pain adaptation can be an explanation for the transformation of acute HSP patients into chronic HSP patients.
Lindgren, Jönsson et al. [20], 2007, Sweden	Longitu-dinal	To describe the prevalence, predictors, and outcome of HSP in unselected stroke populations	intensity: VAS (0–100); time of onset; frequency; inducements	Baseline, *n*=4164mo, *n*=327 16mo, *n*=305 Total, n=327	71 (22%) 74 (24%) 99 (30%)	Lost or impaired arm motor function and high National Institutes of Health Stroke Scale score were predictors of shoulder pain.
Lindgren, Lexell et al. [32], 2012, Sweden	Longitu-dinal	To determine the proportion of persons with PSSP and to assess the risk factors	intensity: VAS (0–100); frequency; ROM	HSP/non-HSP at 4 mo, (58/247); HSP/non-HSP at 16mo, (42/16)	42 (72%)	Left-sided hemiparesis, pain reported frequently, and decreased passive shoulder range of abduction at 4 months are predictors of long-lasting poststroke shoulder pain and require increased attention in the rehabilitation setting.
Lindgren, Brogardh et al. [33], 2014, Sweden	Cross-sectional	To assess the differences between individuals with and without PSSP, and to determine how PSSP is associated with these variables	intensity: VAS (0–100); duration; inducements; ROM	*n*=49	24 (49%)	The PSSP group had significantly decreased passive shoulder abduction (*p*=001) and upper extremity motor function (*p*=03) in comparison with the non-PSSP group, but there were no significant differences between the groups in daily hand activities, perceived participation, or life satisfaction.
Pong et al. [25], 2012, Taiwan	Longitu-dinal	To investigate the correlation between HSP factors during the acute and chronic stages of stroke recovery	intensity: VAS (0–10); ROM	*n*=76, on admission, in acute stage, and in chronic stage	acute, 45 (59%); chronic, 62 (82%)	Significant correlations were found between HSP and the lower motor function levels and shoulder ROM limitations in both acute and chronic stage patients. Shoulder spasticity and abnormal sonographic findings were highly correlated with HSP in chronic stage patients only.
Ratnasabapathy et al. [28], 2003 New Zealand	Longitu-dinal	To measure the occurrence of HSP and to identify the factors that predict risk of shoulder pain after stroke	presene/ absent; time of onset	1wk, *n*=1474 1mo, *n*=1336 6mo, *n*=1201 Total, *n*=1349	256 (17%) 261 (20%) 284 (23%) 529 (39%)	Shoulder pain was positively associated with motor deficit, side of deficit and severity of deficit. The risk of shoulder pain was higher in those with severe upper limb motor deficit (OR 4.94; 95% CI 3.06-7.98) and in diabetics (OR 1.57, 95% CI 1.15-2.14).
Roosink et al. [14], 2011 Netherlands	Longitu-dinal	To identify factors associated with pPSSP in the first 6 months after stroke	intensity: NRS (0–10); time of onset; frequency; distribution; pattern; ROM	Baseline, *n*=37 3mo, *n*=34 6mo, *n*=34	8 (22%) 11 (32%) 9 (26%)	pPSSP was significantly associated with impaired voluntary motor control, abnormal sensation, spasticity, ROM for both shoulder abduction and external rotation, trophic changes and type 2 diabetes

Author, Year, Country	Design	Aim	HSP Evaluation	Sample	HSP Prevalence	Main findings
						mellitus.
Roy et al. [26], 1995, UK	Longitu-dinal	To decide whether shoulder pain in stroke is a marker of severity, or an independent predictor of poor outcome	intensity: VAS (0–10); frequency	*n*=76	55 (72%)	Shoulder pain on movement was the most important predictor of poor recovery of arm power and function, and an important contributor to length of stay in hospital.
Suethanapornkul et al. [30], 2008, Thailand	Longitu-dinal	To study the occurrence of HSP and identify associated factors during rehabilitation period	present/absent	*n*=327, at the begin-ning and the end of study	62 (19%)	Shoulder pain was significantly more frequent in subjects with shoulder subluxation (OR 2.48, 95%CI 1.38-4.46) and at 2-6 months after stroke onset (OR 4.0, 95%CI 2.06-7.79). No significant functional and quality of life impact was found.
Wanklyn et al. [29], 1996, UK	Longitu-dinal	To analyze the incidence of and the factors associated with HSP after stroke	intensity; time of onset; frequency; pattern; aggravating and relieving factors; ROM	*n*=108 at discharge, at 2 mo and at 6 mo after discharge	69 (64%)	Reduced shoulder shrug was associated with HSP at all times and reduced pinch grip was also associated with HSP at discharge. Patients who required help with transfers were more likely to suffer with HSP.
Zeilig et al. [1], 2013, Israel	Cross-sectional	To explore the neuropathic components in HSP, and whether they are specific to the shoulder or characteristic of the entire affected side	intensity: VAS (0-10); time of onset; location; duration; quality; dynamics; ameliorating and exacerbating factors	HSP, *n*=16; no HSP, *n*=14; control, *n*=15		Both poststroke groups exhibited higher sensory thresholds than healthy controls. Those with HSP had higher heat-pain thresholds in the affected shoulder (p<0.001), and more often reported chronic pain throughout the affected side (p<0.001) than those without HSP.
Zhu et al. [46], 2013, China	Retro-spective	To analyze the incidence of and the factors associated with HSP after stroke	intensity: VAS (0–10)	*n*=223	106 (46%)	The number of patients with HSP post stroke increased yearly, attacking mainly males 50-69 years of age. Shoulder pain, upper limb motor function, and function independence were significantly improved after comprehensive rehabilitation.
Zorowitz et al. [27], 1996, USA	Cross-sectional	To explore whether shoulder pain after stroke is related to age, subluxation, ROM, and upper extremity motor impairment	intensity: VAS (0–10); ROM	*n*=20		Shoulder pain after stroke was not correlated with age, vertical, horizontal, or total asymmetry, shoulder flexion or abduction; or Fugl Meyer scores. Shoulder pain was strongly correlated with degree of shoulder external rotation.

Note: HSP, hemiplegic shoulder pain; PSSP, poststroke shoulder pain; pPSSP, persistent poststroke shoulder pain; mo, month; wk, week; VAS, Visual Analogy Scale; NRS, Number Rating Scale.

3. Results

The selected articles were observational studies published between 1995 and 2018. Of the 25 articles included in the analysis, 11 used cross-sectional designs and 14 used longitudinal designs; 23were prospective studies and 2 were retrospective studies. The sample size ranged from 20 to 1,474. Studies originated from 12 countries or districts, United Kingdom (*n*=5), United States of America (*n*=3), Australia (*n*=3), Turkey (*n*=3), Sweden (*n*=3), Israel (n=2), Iran, South Korea, New Zealand, Thailand, Taiwan, and mainland China (*n*=1, respectively). The samples of stroke patients varied in terms of age (average age ranged from 57 to 74 years), gender (female 21% to 66%), types of severity of stroke, stage after the stroke (acute or chronic), and comorbidity.

3.1. Symptom Profile of HSP

3.1.1. Conceptual and Operational Definitions of HSP

Conceptual definition. The nomenclature of HSP used interchangeable terms among articles including HSP syndrome, post stroke shoulder pain (PSSP) and persistent post stoke shoulder pain (pPPSP). 3 articles gave explicit conceptual definitions of HSP. Roosink et al. [14] confined shoulder pain to the shoulder and/or C5 dermatome of the contralesional side with an onset after stroke and present during rest or during active or passive motion. Another latest definition of HSP was clarified as any subjective complaint of pain in the contralesional or affected hemiplegic shoulder

after stroke [15]. Kim's [16] study considered HSP to be a general term for shoulder pain on the hemiplegic side after stroke. Thus, there was an agreement that HSP referred to patient reported shoulder pain on the hemiplegic side after stroke.

Operational definition. Operationalization of HSP in stroke patients varied across studies, with different assessing contents and criteria. 20 of 25 studies evaluated the intensity of HSP. Two subjective methods for quantifying the intensity of HSP were Visual Analog Scale (VAS) and Number Rating Scale (NRS). In the 20 articles, 15 studies used VAS, 4 studies used NRS, and no details were reported in the remaining one (Table 1). In the review, VAS was measured in two ways (i.e. score on a 100mm line or score on an 11-point scale), and it could both be transformed equally to NRS. Thus, we considered them as similar methods in the present study. Even when using the same instrument, the criteria for defining HSP were diverse in these studies. For example, most studies identified a score>0 as determining a patient having HSP. However, one study defined HSP by the NRS point scale of 4 to10 [16]. Another study considered HSP was present for more than 5 cm of 10cm in VAS [17]. Besides, timing of assessment for HSP intensity also varied in these studies. Chae et al. [18] asked the participants to rate their worst shoulder pain in the last week. Gamble et al. [19] defined significant pain as lasting 24h within the last month or since stroke. Lindgren et al. [20] evaluated the worst shoulder pain perceived by the patients during the last 48 hours. In order to achieve accurate assessment and diagnosis, objective measures such as range of motion (ROM), consisting of flexion, abduction, rotation testing, was usually used as pain-related explanatory measurement (Table 1). Radiographs and/or ultrasonographs were used as diagnostic techniques to provide imaging information of shoulders [16, 18, 19, 21-27].

3.1.2. Prevalence of HSP

The prevalence of HSP found in the studies varied from 19 to72% (Table 1). 16 of 25 studies that reported the occurrence of HSP documented the prevalence at more than 30%. Nine of 25 studies reported the prevalence at almost or over 50%. Among these studies, a large population-based longitudinal study consisting of 1349 patients with stroke in New Zealand demonstrated that 529 (39%) patients developed HSP after stroke during a period of six months [28].

3.1.3. Symptom Experience of HSP Patients

Symptom experience of HSP is multidimensional and persistent. Most of these studies reported the intensity of HSP was moderate to severe level of pain. In Lindgren's study [20], one third of the 327 patients developed shoulder pain, the majority of whom had moderate to severe pain (VAS=40 to 100). Adey-wakeling et al. [15] found the median pain score was 40 (measured by VAS). In addition to concerning the intensity of HSP, 18 of 25 studies described the other aspects of the symptom such as frequency, location, exacerbating and alleviating factors, etc. (Table 1). Roosink's group [14] discussed HSP frequency (sometimes, at least 2days per week, daily, continuous) and location, indicating two-thirds of all patients reported daily pain and others reported pain at least twice per week or sometimes. The location of shoulder pain was appeared in anterior positions and radiated to the arm and chest. Another study reported the locations of shoulder pain were: the entire shoulder region (69%), the posterior aspect of the shoulder (37.5%), the entire shoulder region and the elbow region (19%), and the entire upper limb (12.5%). The exacerbating factors of HSP were: active or passive movement (87%), pressure (37%), lack of support to the shoulder (12.5%), and end of day pain (12.5%). The alleviating factors were: rest (56%), medications (31%), supporting or restraining the movement of the shoulder (31%) and physical therapy and massage (25%) [1]. It is consistent with the results of Wanklyn et al. [29], showing the most common exacerbating factor was movement and the most frequent relief was resting the arm.

As for prognosis of HSP, more than 70% of 31 people with stroke still reported pain one year later, which indicated that shoulder pain could be a long-lasting problem after stroke [14]. The other two studies demonstrated inconsistent results but confirmed that HSP is a long-lasting symptom for some stroke patients. Suethanapornkul et al. [30] reported 25/62 (40.3%) pain was resolved, 23/62 (37.1%) improved after treatment and only 13/62 (21%) patients still had shoulder pain upon discharge. Also, Gamble et al. [23] found 41/52 (79%) patients improved or had resolution of their pain during the six months study period.

3.2. Contributing Factors with HSP

The related factors of HSP comprise conditions that might cause post stroke shoulder pain and conditions that impede the recovery of HSP patients while producing negative outcomes in stroke survivors. Although many factors seem distinguishable from one another, the authors consider them to be interrelated and interacting with each other.

3.2.1. Factors with Strongest Empirical Support

Arm Motor deficits. 10 studies examined arm motor function, and all these studies found it was strongly associated with the risk of shoulder pain [14-16, 20, 21, 23-25, 28, 31]. One study using the Frenchay Arm Test (p=0.023, p<0.05) [24] and 2 studies using National Institutes of Health Stroke Scale item 5 (p=0.026 and p=0.03, p<0.05) [16, 20] showed significant correlation between weakness of upper limb and HSP. Crude and multivariate analysis found an absence of upper limb motor function was strongly associated with the risk of shoulder pain (odds ratio [OR]=3.19; 95% confidence interval [CI] 1.77–6.9; p=0.0003) [15]. Similarly, Blennerhassett and colleagues [31] found poor arm motor function was correlated with HSP indicated by Motor Assessment Scale Upper Arm item score (r=−0.45, p<0.05). In a longitudinal study, Pong et al. [25] found poor shoulder motor function levels were associated with HSP, in both acute (r=−0.30; p=0.01, p<0.05) and chronic stage

patients (r=−0.28; p=0.02, p<0.05).

Limited Range of motion (ROM). The findings of 8 studies supported an association between limited ROM and HSP [14, 15, 21, 25, 27, 31-33]. In acute and chronic stages, HSP was both significantly associated with severity of ROM restriction in all five motion planes (r=−0.25 to −0.57, p<0.01; r=−0.38 to −0.61, p<0.01, respectively) [25]. In Wanklyn's group [29], presence of HSP was strongly associated with reduced shoulder shrug ability at each time. Additionally, the proportion of patients who had HSP reported limited passive shoulder ROM increased from 26/39 (67%) at discharge, 45/59 (76%) at 8 weeks to 29/36 (81%) at 26 weeks. In a longitudinal study, passive range of abduction at 4 months after stoke was one of predictors for long-lasting shoulder pain (1 year follow- up) (p=0.02) [32].

Abnormal Sensation. A significant association was observed between a reduction in sensitivity to light touch (p<0.005), a reduction in sensitivity to temperature sensation (p<0.005) and the development of shoulder pain. Pain was associated with sensory loss (thermal and touch) [19]. In Zeilig's study [1], both post stroke groups exhibited higher sensory thresholds than healthy controls (p<0.05). Sensory testing revealed that individuals with HSP did have a significantly higher heat-pain threshold in the affected shoulder compared to those without HSP, which positively associated with the intensity of HSP (p<0.05) [1]. Also, significant statistical difference was found between the pain and non-pain groups in relation to reduced cold temperature sensibility (p<0.001), light touch sensation (p<0.001) [23]. A latest study showed that the magnitude of pain adaptation in the HSP group was significantly smaller than in either the non-HSP group (p<0.05) and the control group (p<0.01), and the insufficient pain adaptation can be an explanation for the transformation of acute HSP patients into chronic HSP patients [34]. A logistic regression showed that the best predictors for HSP were increased light touch threshold, increased vibration threshold and activity of daily living ability, and the overall percentage correctly predicted by the model was 97.4% [17]. However, one study in this review found no association between PSSP and somatosensation in a univariate analysis [16].

3.2.2. Factors with Equivocal Empirical Support

Subluxation. Two studies confirmed that subluxation was correlated to underlying HSP [20, 21]. A striking finding in Aras's study [21] was that almost all patients with Reflex Sympathetic Dystrophy (RSD) had shoulder subluxation, which suggested that glenohumeral joint subluxation may be a causative factor for RSD and, as a result, pain. However, four studies found no significant correlation between HSP and glenohumeral subluxation, especially during the chronic stage [1, 22, 25, 27]. Wanklyn et al. [29] reported that subluxation was detected clinically in 31/108 (29%) patients at discharge, 24/106 (23%) at 8 weeks and 27/106 (26%) at 26 weeks. No relationship was found between pain and the presence of subluxation except at the 26-week assessment. However, Kim et al. [16] found that subluxation was significantly associated with HSP at 1 and 3 months after a stroke, but not associated with HSP during the first 6 months after a stroke.

Spasticity. Spasticity is common in individuals with HSP. Higher spasticity scores were found in the HSP group when compared with the non-HSP group, which suggests an association between HSP and spasticity [1]. Lindgren et al. [20] reported similar results. However, a study conducted by Karaahmet et al. [24] showed the difference was not statistically significant, although the majority of patients with spasticity experienced HSP. In addition, Pong et al. [25] confirmed shoulder spasticity was correlated with HSP during the chronic stage, but no significant correlations were observed between HSP and shoulder spasticity during the acute stage. Another study contradicted this finding by showing a borderline significant association existed between spasticity and HSP at the chronic stage [16]. Thus, it is difficult to determine whether spasticity is an underlying factor of HSP.

Depression and anxiety. Seven studies analyzed not only physical factors, but also psychological factors related to shoulder pain. Four of them found no relationship between HSP and anxiety and depression scores, which might indicate mood disorders were not a risk factor for HSP [23, 24, 29, 30]. Similar findings were reported on the association between depression and HSP by Roosink et al. [14]. However, two studies pointed out that there was a significant association between depression and the development of shoulder pain (p<0.005) [17, 19]. In addition, one of them revealed a significant association between anxiety and HSP (p<0.005) [17].

3.2.3. Factors with Weak or No Support

Several factors examined in the 21 investigations included in this review had little or no association with HSP. For example, 10 studies found that there was no association between shoulder pain and basic demographic characteristics (age, sex, body weight, etc.), the affected hemisphere, stroke type, stroke severity, Brunnstrom stage, and living condition [1, 15-17, 21-23, 31-33]. Although Hadianfard and Hadianfard [17] revealed that age was a significant factor in developing HSP and Kim et al. [16] suggested younger age was an independent risk factor for HSP during the first 6 months after stroke, most studies did not consider age to be a risk factor for HSP. In addition, 2 studies showed that shoulder pain was positively associated with type 2 diabetes mellitus (OR 1.57, 95% CI 1.15–2.14; 44% vs 9%, p=0.024, p<0.05) [14, 28].

3.3. Outcomes of HSP

The outcomes associated with HSP in stroke patients were serious, but the findings were controversial in some studies. The types of these outcomes can be divided into three main categories: quality of life, functional status, and length of hospitalization. One of the most important outcomes is health-related quality of life (HRQOL). Mean HRQOL was

lower in participants who reported HSP during the follow-up period (participants with/without HSP, 0.572 ± 0.357/0.725 ± 0.369, p=0.017, p<0.05). It showed that HSP was an independent determinant of subsequent HRQOL [35]. Chae et al. [18] measured HSP using question 12 of the Brief Pain Inventory that was directly related to pain-related quality of life (QOL), explaining 20% of the variance in the model. However, one study found no significant difference in QOL outcome between patients with or without shoulder pain. This could be caused by a good outcome after treatment [30].

The functional outcomes mostly consisted of issues related to daily hand activities, self-care activities or activities of daily life [18, 22, 26, 30]. In Barlak's study [22], the group without HSP showed significantly more improvement than the group with HSP in functional outcomes measured by a Functional Independence Index (FIM) (p=0.01, p<0.05). Another study, however, failed to demonstrate a statistical relationship between HSP and activity limitation assessed by FIM [18]. No significant differences in daily hand activities (p=0.15), perceived participation (p=0.11, p>0.05), and life satisfaction (p=0.15, p>0.05) were found between an HSP group and non-HSP group by Lindgren and Brogårdh [33]. The third outcome associated with HSP highlighted in these studies was duration of hospitalization. Roy et al. [26] used logistic regression analysis and showed that shoulder pain was an important contributor to length of stay in hospital (p=0.026, p<0.05). Furthermore, Barlak et al. [22] showed that the hospitalization period was significantly shorter in the group without HSP (p=0.03, p<0.05).

4. Discussion

HSP is highly prevalent in stroke patients that it should be taken into serious consideration. Different prevalence reported in the studies possibly derived from either difference in measurement or disparity in recruitment criteria (e.g. duration of hemiplegia and severity of paralysis). Another possible reason is the inconsistency in effect of care and treatment of post stroke patients. Although only three studies defined the conceptual definition of HSP in this review, all studies provided the operational measurement of HSP among stroke patients. The studies used a basically consistent meaning of shoulder pain after stroke, but the terminology employed was diverse among those studies. Certainly, a standardization of the definitions and terminology of HSP in the future, will benefit clinicians and help them to recognize, diagnose and treat this symptom as best as possible.

Our review is related to stroke patients and to numerous factors that may associate with HSP in this population. The evidence in this area varies in its support for different variables. Support is strongest for impaired motor function, ROM and sensation, with equivocal results for subluxation, spasticity and depression. It provided little evidence that many of the risk factors were empirically linked to HSP (e.g., demographic characteristics, hemisphere and stroke type). Other studies provided divergent findings. Kalichman et al. [9] reviewed the most frequent factors associated with HSP including age and sex, preexisting shoulder pain and degeneration, subluxation, time since stroke, hemiplegic side and co-morbidities, the analysis of some of which (i.e. age, subluxation) still provided ambiguous results. In the present study, we classified these factors into three categories based on the strength of supporting data that could help to guide healthcare professionals in developing interventions and choose treatments that are targeted on factors related to HSP.

The goal of HSP symptom management is to avert or delay negative outcomes (e.g. decreasing QOL) through effective and professional treatment strategies. Management begins with the assessment of the symptom experience (e.g., severity, frequency) of stroke patients; then, this is followed by the identification of the factors related to developing HSP, and the evaluation of outcomes and the management process. Note that symptom management is a dynamic process, often requiring changes in strategies over time or in response to a patient's acceptance (or lack of acceptance) of a specific strategy [36]. Clinicians are also supposed to add patient preferences to symptom intervention. Additionally, determining the interrelationship with symptom experience, factors, and outcomes for persons with HSP would help health providers understand how to integrate different strategies of symptom management. Clinicians might develop clinical pathways that could be used to examine the efficacy and feasibility of strategies in symptom management of HSP.

5. Implications to Practice and Research

There are two methods, one subjective and one objective, for assessing and measuring shoulder pain symptom. Subject measurements mainly consisted of VAS and NRS. However, objective methods, such as the modified Neer test (forced passive forward flexion) and passive external rotation, are thought to be more accurate than subjective tests [15]. Dromerick et al. [37] found that patient self-reporting of pain significantly underestimated the extent of pain found during physical examination. Nonetheless, researchers often favor subjective over objective measures when studying HSP in stroke patients, likely because of the lower cost and time saved when using subjective measurement [38, 39]. As a result, it suggests to combine subjective instruments with some rapidly administered objective methods for more thorough symptom assessment of stroke patients. This may result in potentially uncovering the presence of HSP when it might otherwise have been overlooked or found with difficulty.

Moreover, we found that VAS and NRS were the most frequently used subjective measures of HSP in people with stroke. Although these two methods are simple and rapid, they are often used for general pain not specific pain such as shoulder pain or these methods may only allow the evaluation of the severity of pain not reflect other dimensions of pain (e.g., frequency, distress). While HSP has been

identified as one of the most prevalent symptoms, specific symptom assessment instruments need to be developed based on high-quality research evidence to provide a useful framework of symptom theories or models. For example, some articles have indicated that patients with more severe HSP are more likely to avoid rehabilitation or even detest being touched by others [40, 41]. Thus, sensitivity to touch or willingness to seek rehabilitation could be additional indications of painful severity that could be used as measuring items. Apart from the severity, a new instrument could evaluate the frequency, distress, quality of HSP based on the Theory of Unpleasant Symptoms [42]. Furthermore, patients with cognitive or communication difficulties are often excluded in many studies; instruments that involve patients who have difficulty communicating should be considered. For instance, the Revised Faces Pain Scale and the Iowa Pain Thermometer had been used for the analysis of samples from cognitively impaired patients [43]. Therefore, researchers should develop instruments for assessing the symptom of HSP that consider particular symptomatic characteristics, more assessment dimensions and special populations.

To date, a large number of treatments have been proven to be efficacious to some extent but cannot radically cure HSP for stroke survivors because of the complicated etiology and mechanisms related to HSP [44, 45]. Patients who have persistent complications are more concerned with the experience of distressing symptom. Though we have identified factors which were associated with the symptom between groups with or without the existence of HSP, very few studies have analyzed the factors that influenced the severity and distress of shoulder pain after stroke. Nursing practice is often supposed to care more about people's feelings and experiences of symptoms. Recognizing the factors that can affect symptom experience of HSP will be a new direction for future research.

It was shown that symptom outcomes of HSP were involved in three aspects, QOL, functional status and hospitalization period. Mean QOL score was lower in participants who reported HSP at any time in the first year after the onset of stroke [35]. Further research may explore the relationship between different dimensional variables of pain and QOL to find out how HSP affect quality of life in stroke patients.

6. Conclusion

We reviewed relevant studies on prevalence, definition, experience, associated factors and outcomes of HSP in people with stroke. The definition or diagnostic criteria of HSP need to be unified. Accurate and multidimentional assessment of HSP is necessary in monitoring of the symptom. Healthcare providers should be directed to strengthen awareness of assessing HSP comprehensively. There is divergence of opinions related to contributing factors and outcomes of HSP in stroke survivors, which emphasizes the need for further research to provide evidence. Clinicians and researchers should develop innovative interventions to support symptoms management, including targeting the experience, factors, and outcomes related to the HSP symptom. Further exploring the characteristics of HSP will provide a foundation for developing future interventions for efficient, effective symptom management in patients with stroke.

Conflict of Interest

The authors have no conflict of interest to disclose.

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