Flinders UNIVERSITY

Incidence and Associations of Hemiplegic Shoulder Pain Post Stroke:

A prospective population based study

Abstract

Objective: To provide an epidemiological perspective of the clinical profile, frequency and determinants of post stroke hemiplegic shoulder pain.

Design: A prospective population-based study of an inception cohort of participants with 12 months follow up period.

Participants: Multiple ascertainment techniques were used to identify 318 confirmed stroke events in 301 individuals. Among 301 adults with stroke, data on shoulder pain were available for 198 (83% of survivors) at baseline, and 156 and 148 at 4 and 12 months, respectively.

Setting: Participants were recruited within a geographically defined metropolitan region with estimated population of 148,000 in Adelaide, Australia. Ascertainment and follow up included both general community and hospital settings.

Interventions: not applicable

Main Outcome Measures: Subjective reports of onset, severity and aggravating factors for pain, and three passive range of motion measures were collected at baseline, and follow-up at 4 and 12 months.

Results: 10% of participants reported shoulder pain at baseline, whilst 21% reported pain at each follow-up assessment. Overall, 29% of all assessed participants reported shoulder pain during 12 months follow up, with the median pain score (VAS = 40) highest at 4 months and more often associated with movement at later time points. Objective passive range of motion tests elicited higher frequencies of pain than self-report, and predicted later subjective shoulder pain (crude relative risk of 3.22 (95%CI 1.01-10.27).

Conclusions: The frequency of post-stroke shoulder pain is almost 30%. Peak onset and severity of hemiplegic shoulder pain in this study was at 4 months, outside of rehabilitation admission timeframes. Systematic use of objective assessment tools may aid in early identification and management of stroke survivors at risk of this common complication of stroke.

Key Words (3-7):

Stroke, epidemiology, hemiplegia, shoulder, pain

List of Abbreviations:

VAS Visual Analogue Scale

CI	Confidence Interval
LACS	Lacunar Syndrome
TACS	Total Anterior Circulation Syndrome
PACS	Partial Anterior Circulation Syndrome
POCS	Posterior Circulation Syndrome
NIHSS	National Institute of Health Stroke Scale
IQR	Inter-Quartile Range
OR	Odds Ratio

Hemiplegic shoulder pain has been described as one of the four most common medical complications following stroke[1], with others including depression, falls and urinary tract infections[1]. Earlier studies have reported the frequency of shoulder pain following stroke to be as high at 65-70%[2-4]. A more recent prospective Swedish study of 416 consecutive stroke patients reported that almost a third of stroke survivors developed shoulder pain, the majority of whom reported moderate to severe pain[5]. Contributions to pain development are often multifactorial; biomechanical factors are significant[6], and may occur in isolation or in addition to changes in tone[7] or neuropathic mechanisms[8]. Hemiplegic shoulder pain is associated with a reduction in functional use of the arm[9], interference with rehabilitation[9], increased length of stay[9] and higher rates of depression[10]. Complexities in aetiology and subsequent diagnosis mean that treatment of shoulder pain is difficult and reviews have found little evidence to guide clinicians on effective prophylactic and treatment options[11]. Understanding the pattern of presentation, and establishing tools to support early identification of those likely to develop pain would assist clinicians and patients.

The primary aim of this study was to determine the frequency, characteristics over time, and associations of hemiplegic shoulder pain in a defined metropolitan population of South Australia. The secondary aim was to evaluate the predictive use of three standardised passive objective measures of shoulder range as screening tools for development of shoulder pain. Objective assessment is necessary in conjunction with subjective questioning, as self-report alone has been shown to be a poor predictor of examination findings[6], and accurate clinical assessment and diagnosis is vital in establishing targeted management plans. A case control study suggested that a simple set of clinical assessments (three passive range of motion tests) conferred a 98% probability of predicting early hemiplegic shoulder pain at rest[12]. The generalizability of this finding is limited due to its small sample with multiple exclusion

criteria (thalamic infarcts, upper limb sensory deficit, previous shoulder injury, complex regional pain syndrome, dysphasia). We evaluated this same set of assessments on all participants in a stroke incidence study, based on the principles of complete ascertainment[13], to test their application as a predictor of development of hemiplegic shoulder pain.

Methods

Overview

The Adelaide stroke incidence study (ASCEND) was a prospective population-based stroke incidence study conducted in a defined region of the western suburbs of Adelaide, South Australia, with a census projected population of over 148,000. During the period from 15 July 2009 to 15 July 2010, multiple ascertainment methods were used to identify all occurrences of stroke. Ethics approval was obtained from every tertiary hospital in Adelaide and University of Adelaide and all participants provided consent prior to enrolment in the study. Detailed methodology has been previously described[14], including specific information regarding the study population and ascertainment techniques.

Following informed consent, participants were assessed at baseline, at 4 months and at 12 months. All data were collected as part of the larger ASCEND study and entered into a custom-designed online database. The data set specific to this study was extracted via an automated database query and checked against the raw database manually. Only data that were truly prospective were included for analyses, as retrospective report of subjective pain

measures was not deemed reliable and retrospective case note data would not include the objective tests.

Definitions

Stroke was defined as "rapidly developing clinical signs of focal (or global) disturbance of cerebral function lasting more than 24 hours (unless interrupted by surgery or death) with no apparent cause other than of vascular origin"[15]. Hemiplegic shoulder pain was defined as any subjective complaint of pain in the contralesional, or affected hemiplegic shoulder following stroke. Hemiplegic shoulder pain encompasses all aetiologies and we did not exclude patients on the basis of premorbid shoulder pathology. Pain was measured using a Visual Analogue Scale (VAS range 0-100) with severity classified into mild (10-30) and moderate-severe (40-100) in line with previous publications[16, 17]. Upper limb motor function was determined using question 5 from the NIHSS – motor arm score of 3 or above was classified as 'no motor function' (score 3 = no effort against gravity; score 4 = no movement), and reduced motor function was score 1-2 (score 1 = drift; score 2 = limited effort against gravity).

Demographic Data, Subjective and Objective Assessments

The subset of data of interest in the study included record of demographic data, and baseline and follow up subjective and objective measures pertaining specifically to shoulder pain.

Demographic and clinical characteristics were recorded to characterise the subsets within the study population and to explore any associations with risk of development of shoulder pain.

Data included age, gender, significant medical history, stroke subtype and aetiology, affected hemisphere, and motor arm component of the National Institute of Health Stroke Scale (NIHSS).

Subjective information included history of shoulder pain prior to stroke and presence of shoulder pain on affected side. If pain was reported, further questions regarding time of onset, severity of pain, and aggravating factors were asked. Patients were asked if pain was worse at rest, on movement (active or passive), or at night. Pain severity was scored using a vertical VAS. Each consented participant was assessed by a trained study nurse.

A rehabilitation physician taught all data collectors a standardised approach to objective tests, and a video support package was made and provided for ongoing reference.

Objective measures of the participants' affected upper limb included:

- the modified Neer test (forced passive forward flexion) tested in a seated position
- passive Hand-Behind-Neck test (passive abduction, external rotation) tested in a seated position, and
- passive external rotation as compared to unaffected limb. Passive external rotation was measured with the patient in a seated position. Range was measured using a goniometer.

Any pain on modified Neer or passive hand-behind-neck was scored as a positive result. Affected limb passive external rotation range of more than 10^{o} less than the unaffected limb was scored as positive limitation of range of movement.

Statistical Analysis

Comparisons were made of baseline demographics for participants with and without shoulder pain using Wilcoxon tests for continuous variables or chi-squared tests for categorical variables. Non-parametric tests (i.e. Wilcoxon or Kruskal-Wallis tests) were selected in the context of analysis of continuous variables because some variables (such as VAS and NIHSS) had skewed distribution. The primary outcome was onset of shoulder pain within the first year of stroke onset. Measures of shoulder function (subjective report of pain, pain severity, aggravating factors, and objective assessments) at each visit were compared using Kruskal-Wallis or chi-squared tests. Associations between baseline demographic subsets and development of shoulder pain were assessed using logistic regression models and statistically significant predictors were included into multivariable logistic regression models. Data are reported with the standard level of significance ($P < 0.05$) and with 95% confidence intervals (CI). All analyses were performed using SAS software version 9.2 (Cary, NC, USA).

Results

As some participants had more than one stroke event, a total of 318 strokes were confirmed in 301 people in the study population. Excluded were 103 people without a shoulder assessment due to death (60%), retrospective ascertainment (12%), or non-consent to participation (28%) (see Figure 1). For baseline assessments, 73% of all recruited patients were assessed within one week of symptoms onset[14] (average 8.7 days post onset). At baseline, a shoulder assessment was completed on 198 (83%) of 239 survivors, 156 (75%) at 4 months, and 148 (77%) at 12 months. A total of 226 shoulder assessments were performed at any assessment point within the follow-up period, with complete data from all 3 time

points available for 105 participants surviving to 12 month follow-up. Among survivors, baseline characteristics were comparable between participants with and without pain, except severity of upper limb deficits and history of premorbid shoulder pain which were significantly greater in those participants reporting subjective pain (Table 1). The demographic and clinical variables of participants receiving shoulder assessment as compared to those not receiving any assessment are summarised in Supplementary data Table I. In the group who did not receive a shoulder assessment, there were significantly more haemorrhagic strokes (25% versus 9%) and Total Anterior Circulation Syndrome (TACS) strokes (67% versus 18%), reflecting higher mortality from more severe strokes. Data from patients who did not receive shoulder assessment were excluded from further analysis.

Table 2 summarises the incidence of shoulder pain over 12 months. Comparison of participants receiving any assessment (n=226) to participants receiving assessments at all time points (n=105) demonstrated similar frequencies at each follow up, with a clear pattern of increasing frequency of pain over 12 months. Of stroke survivors receiving any assessment, 10% reported pain at baseline and 21% at each follow up period. Overall, approximately one third (65/226=29%) of individual participants reported onset of shoulder pain within the 12 months following their stroke. In the cohort of participants receiving shoulder assessment at all three time points (n=105), Figure 2 shows that shoulder pain increased in frequency over time: 8% at baseline, 18% at 4 months, and 21% at 12 months A relatively low rate of pain resolution at each time point is demonstrated (6% at 4 months and 14% at 12 months respectively).

Subjective reports of severity and factors aggravating hemiplegic shoulder pain amongst participants receiving any assessment are summarised in Table 3. The median pain score (VAS = 40) was highest at 4 months. Pain characteristics in the early weeks demonstrated milder pain (median VAS = 15) which was more prominent at rest (including night). At follow up, pain was shown to be more associated with limited active and passive range of movement and significantly fewer participants reported pain which was worse at rest or at night (Figure 3).

Crude and multivariable analysis found a strong association between premorbid shoulder pain and post-stroke hemiplegic shoulder pain (Table 4). Additionally, an absence of upper limb motor function was strongly associated with risk of shoulder pain (OR 3.19 (1.77-6.9) p=0.0003). The odds ratio (CI 95%) for pain associated with reduced arm function was 1.24 (0.7-2.17) p=0.458. A large proportion (86%) of participants with TACS strokes died before the baseline assessment. There was no association of shoulder pain and basic demographics, stroke syndrome, affected hemisphere, or stroke severity.

In stroke survivors who reported pain at baseline, baseline passive range of motion tests were not consistently positive (not all patients reporting pain had positive objective tests). Follow-up assessments demonstrated increasing frequency of positive objective tests in those with reported pain, and objective passive range of motion tests were associated with higher frequencies of pain than were elicited by self-report alone. Further evaluation revealed that positive baseline objective assessments, despite the absence of subjectively reported pain, conferred a statistically significant crude relative risk of 3.22 (95% CI 1.01 to 10.27) for future development of hemiplegic shoulder pain within a 12 month period. Multivariate

analysis, adjusting for high NIHSS score (>5 above median) and significant motor upper limb deficit, demonstrated an odds ratio of 2.13 (CI 0.54 to 8.35) although this was not significant (Table 5).

Discussion

In a field in need of greater research focus, this study contributes data on early incidence of pain and pain characteristics in the first year post stroke. Additionally, the study supports the predictive value of easily reproducible objective screening tests.

This study found that approximately one third of stroke survivors experienced shoulder pain at some stage in the 12 months post stroke, with peak incidence of pain at 4 months. Congruous data in studies of comparable methodology[2, 5, 18] lend weight to this finding regarding rate of shoulder pain (previous papers reported rates as high as 70%)[2, 19, 20]. A pertinent issue to consider, in the context of persistently significant rates of hemiplegic shoulder pain, is the possibility that this may reflect a lack of improved prevention measures regarding education and shoulder care over more recent years. Thus, despite previous studies highlighting the amplitude of this issue, it is postulated that minimal gains in evidence-based treatment and prevention options, or translation of the same into practice are indicated.

A novel finding of our study is the comparatively low frequency of very early (average 8.7 days) hemiplegic shoulder pain (10%). Lindgren et al[5] followed up 416 people from a Stroke Register, with specific study pain questions and assessment at 4 and 16 months; at follow up I (4 months), almost 40% of participants reported that their pain begun between 0-2 weeks post stroke. In the current study, prospective data regarding baseline pain were collected.

Interestingly, patients who reported pain within the first few days following stroke were not necessarily those who went on to have persistent pain complaints. There was a much higher rate of new onset pain at 4 month follow-up compared to pain persisting from baseline assessment, highlighting the need for ongoing monitoring after hospital discharge. A relatively low rate of pain resolution at each time point was demonstrated (6% at 4 months and 14% at 12 months respectively), further indicating the need to establish an increased pool of effective evidence-based treatment options. The increasing association of pain with range of movement (active and passive) over time may represent cumulative musculoskeletal contributors and adaptive mechanisms, with pain on movement recognised as one of the cardinal features of musculoskeletal pain[21]. Mechanisms of pain may differ and additional research exploring evidence-based treatment options that address early versus later onset hemiplegic shoulder pain are needed.

The predominant associations between clinical profile and risk of shoulder pain were in participants with premorbid pain and those with more marked upper limb motor deficit. Whilst previous population-based studies[5] have found motor deficit to be predictive, they have not demonstrated premorbid shoulder pain as a risk factor for developing pain. In this study, history of shoulder pain was reported in 27% of participants with hemiplegic shoulder pain, compared to only 4% of those who did not report pain. This differs from Lindgren et al[5], who found similar rates of premorbid shoulder pain reported by those who subsequently developed pain and those who did not (23% versus 22% respectively). Pain history is a simple question easily added to clinical screening assessment battery and further helps identify an at-risk cohort.

The association of compromised range of motion with persistent pain is supported by recent studies. Research supports that persistent pain is more likely in patients with left sided weakness[22], and in those who demonstrate reduced passive abduction range[22, 23], as well as patients with reduced external rotation range, impaired voluntary motor control and spasticity[22, 23]. We did not find an association between affected hemisphere and pain development, but our data does support the previous findings that pain is associated with reduced passive abduction and external rotation (passive hand-behind-neck and external rotation tests respectively), and impaired motor function. Testing of passive range is often impacted by increasing tone, though formal spasticity assessment was not included in this study.

With the three passive range of motion tests used, it was possible to identify those likely to develop pain. Those patients who demonstrated a positive response on an objective passive range of movement test at baseline trended to be at increased risk of later pain, suggesting that these tests may serve a useful screen among at-risk patients, namely those with more severe upper limb paresis. Rajaratnam[12] proposed use of all three tests to identify those at risk of early pain at rest. Results from this study support use of these tests as a screening tool beyond the early phase, with evidence that positive objective results double a patient's probability of developing future hemiplegic shoulder pain. At both follow up points, the passive external rotation test and modified Neer test recorded greater number of positive results than the passive hand-behind-neck. Passive external rotation findings on follow up were greater than subjective report of pain alone (21% reported pain at 4 months, 25% recorded positive external rotation test; 21% reported pain at 12 months, 22% recorded positive passive external rotation test). Remaining objective tests did not provide results higher than subjective pain result, but it must be considered that the variety of movements

covered by the use of all three of these tests provides a more thorough screening tool. The tests used are simple to perform, easy to teach in a reproducible manner, and time and cost efficient in the context of incorporation into standardised protocols. Whilst it is well established that transfer of evidence into clinical practice is significantly delayed, the use of such a simple screening assessment can be hoped to be easily implemented within a field of medicine at ease with joint assessment and manual handling.

The use of screening assessments should not replace more in-depth diagnostic assessments of patients with verified hemiplegic shoulder pain. The increasing body of research exploring the contribution and overlap of neuropathic as well as nociceptive pain mechanisms[8, 23, 24] highlights the importance of careful assessment beyond the musculoskeletal paradigm covered by the outlined objective measures. As such, the assessment outlined is supported as a screening tool, rather than a diagnostic tool. More in depth assessment is required to ascertain potential contributors to active pain, and should consider specific spasticity measures and comprehensive pain history. Screening in this study is perhaps of more utility to identify those not subjectively reporting pain at rest but potentially experiencing pain with range of movements beyond their active range. The data supports that positive objective tests double the risk of future development of pain. It must also be highlighted that the paucity of evidence-based treatment options currently available means that successful screening does not yet yield significant benefit to the patient group. A focus on effective treatment options is required in order to make best use of screening within an assessment and management protocol.

Study Limitations:

The study was limited by some loss of patient data due to early death or delay in ascertainment which reduced the ability to achieve timely or prospective assessment. As highlighted in the parent study[14], ascertainment may have been incomplete despite intensive efforts. In addition, there was variable loss of data at the follow up assessments. Finally, we did not account for spasticity in our assessments, which could have affected passive range and pain reports.

Strengths of our study include the use of 'ideal' methodology[13] to avoid selection bias and the prospective assessments available for analyses.

Conclusion:

Close to 30% of people develop pain in the first year after stroke, with peak incidence at 4 months. Comparison with an earlier population study[5] shows that, despite increased focus on evidence-based treatments in stroke, over 7 years no reduction in frequency of this common complication stroke has been shown. Systematic use of clinical assessments are useful in identifying people at risk of shoulder pain. As the disorder is most common and severe after hospital discharge, targeted protocols including predictive objective measures may facilitate improved identification and management. Further research is required to elucidate a practical range of preventative and treatment options for this condition.

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Conflicts / Disclosures:

None to declare

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Figure Legend:

Figure 2: Frequency of Hemiplegic Shoulder Pain

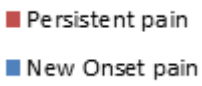

Figure 3: Factors aggravating shoulder pain over 12 months