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Risk Factors for Hemiplegic Shoulder Pain: A Systematic Review

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1Department of Rehabilitation Medicine, VU University Medical Center, Amsterdam, the Netherlands; 2Institute for Research in Extramural Medicine, Vrije Universiteit, Amsterdam, the Netherlands

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ABSTRACT: A systematic review was conducted to postulate risk factors for hemiplegic shoulder pain in the context of an etiological model and to evaluate the evidence for these risk factors. Studies on risk factors for hemiplegic shoulder pain were identified by a systematic search through MEDLINE and the Cumulative Index of Nursing and Allied Health Literature, and an etiological model was subsequently constructed. To evaluate the evidence for these risk factors, the methodological quality of the studies was rated according to a criteria list. Eleven studies on risk factors for hemiplegic shoulder pain were identified. The relationship between hemiplegic shoulder pain and subluxation and muscle tone has been investigated most frequently, but no consistent findings have been reported. Overhead pulley seems to increase the incidence of hemiplegic shoulder pain. Although much speculation is found in the literature about the etiological processes and risk factors for hemiplegic shoulder pain, there is only weak evidence that an increase in muscle tone increases the occurrence of hemiplegic shoulder pain. Overhead pulley may lead to repeated traumas of the shoulder and cannot be recommended.

KEY WORDS: hemiplegic shoulder pain, risk factors, systematic review

I. INTRODUCTION

Loss of function of the upper extremity is a common problem after stroke. Broekx et al.1 found that 4 years after a stroke, 67% of patients experienced nonuse or disuse of the affected arm as a major problem. The Copenhagen Stroke Study, a population-based follow-up study, reported that 21% of surviving stroke patients had not attained full upper extremity function on discharge from the hospital.2 The function of the hemiplegic upper extremity can further deteriorate as a result of hemiplegic shoulder pain, which is a common complication after stroke.1,3-8 Measures to prevent shoulder pain have been recommended,3,8-11 and various methods of treatment have been described.3,12 During recent decades, at least 16 reviews have been published about the etiology, prevention, and treatment of hemiplegic shoulder pain.3,6,9-11,13-23 None of these reviews has systematically evaluated the evidence for the putative causes and pathways leading to hemiplegic shoulder pain. However, knowledge of the risk factors is important because this will provide indications of how to prevent (an increase in) hemiplegic shoulder pain or how to cure it more effectively. The objectives of this article are to present an etiological model for hemiplegic shoulder pain and to systematically evaluate the available evidence for the risk factors included in the model.
ii. METHODS

A literature search was made using MEDLINE (from 1966 to March 2000) and the Cumulative Index of Nursing and Allied Health Literature (from 1982 to March 2000) using the following keywords: (a) hemiparesis, hemiplegia, stroke, cerebrovascular disorder, cerebrovascular disease, or cerebral accident; (b) shoulder, arm, or upper extremity; and (c) pain. The references of the selected articles were also studied to identify additional eligible studies. Further selection was based on the title and abstract. Cohort and case control studies were included in the systematic review if they met the following inclusion criteria: (a) hemiplegia caused by stroke in the majority of the patients and (b) risk factors for hemiplegic shoulder pain were described separately. Exclusion criteria were (a) pain in the hemiplegic shoulder caused by any obvious reason (e.g., fracture, long top tumor, rheumatoid arthritis) and (b) therapeutic intervention study. Cross-sectional studies were excluded because they lack a clear time relationship between risk factors and hemiplegic shoulder pain. For practical reasons, only studies published in Dutch, English, French, and German were selected.

For the construction of the etiological model, the causes described in the selected articles and the reviews were allocated to one of three categories of impairment after stroke: causes related to impairments in motor function (hemiplegia), causes related to impairments in sensory and cognitive functioning (e.g., sensitivity disorders, aphasia, neglect, visual field defects), and causes related to impairments in the autonomic nervous system (e.g., systematic reflex dystrophy, shoulder-hand syndrome). If an etiological pathway leading to hemiplegic shoulder pain was described, only the primary cause was allocated to the model. Only those causes postulated in the selected articles that were biologically plausible and related to the effects of stroke were included in the model.

The methodological quality of the selected studies was rated by the first author according to a criteria list that is presented in the Appendix. This list was derived from similar criteria lists and focuses on valid measurements of both the risk factors and the hemiplegic shoulder pain, the evaluation of the time relationship between these variables, and the statistical analysis. The number of items on the list of criteria was reduced to 12 (maximum attainable score). Data on study design, study population, and risk factors were extracted. If possible, odds ratios (ORs), risk ratios (RRs), and 95% confidence intervals (95% CIs) were calculated. Risk factors for which only correlation coefficients were given were not considered to be informative for the identification of etiological relationships. A significant correlation coefficient only indicates a linear association between variables, and the results depend on the units used. Correlation coefficients cannot be used for prediction of the dependent variable (hemiplegic shoulder pain) by the independent variable (risk factor).

III. RESULTS

A. Selected Studies

Nine articles and two abstracts that were retrieved described studies on risk factors for hemiplegic shoulder pain (Table 1). The study populations varied from 21 to 219 patients. Most studies included only patients with stroke, but one study also included patients with head injuries or tumors leading to hemiplegia. One other study did not report the cause of hemiplegia. Nine studies had cohort designs (including one clinical trial), and two studies had case control designs.

B. Etiological Model

The etiological model for hemiplegic shoulder pain is shown in Figure 1. Motor function impairments after stroke may lead to flaccidity or spasticity of the upper extremity. Both are mentioned as risk factors for hemiplegic shoulder pain: Flaccidity may cause stress on the soft tissues around the shoulder (capsule, ligaments, nerves, plexus brachialis), which might result in hemiplegic shoulder pain, and can also lead to hemiplegic shoulder pain via glenohumeral subluxation. In some cases, no further explanation was given. Spasticity is considered to be a risk factor by several authors; it may lead to pain due to an unspecified lesion, or a contracture of the shoulder, or no...
# TABLE 1
Description of Study Population, Study Design, Definition of Hemiplegic Shoulder Pain, and Risk Factors for Hemiplegic Shoulder Pain

<table>
<thead>
<tr>
<th>Study (MS)</th>
<th>Number (at start and end of follow-up) and description of patients</th>
<th>Design, time of measurement, follow-up</th>
<th>Definition HSP, methods of measurement</th>
<th>Risk factors</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bohannon&lt;sup&gt;29&lt;/sup&gt; (4)</td>
<td>30/30; patients with CVA admitted for further rehabilitation; age 68.5 years (SD = 10.5); 13 male, 17 female; time since onset of stroke at beginning 30.9 days (SD = 15.3), at discharge 66.6 days (SD = 30); side of hemiplegia 22 left, 8 right</td>
<td>CS, on admission and at discharge</td>
<td>Pain during slow external rotation; modified Ritchie articular index: 0 = no tenderness; 1 = complaint of pain; 2 = complaint of pain and wince; 3 = complaint of pain, wince, and withdrawal</td>
<td>Subluxation (palpation) RR = 1.17 (0.98 to (category 0 vs. 1 to 3)</td>
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<tr>
<td>Crossen-Sills and Schenkman&lt;sup&gt;25&lt;/sup&gt; (0)</td>
<td>21 men admitted to rehabilitation hospital following CVA; no further description given</td>
<td>CS, on admission, 3 weeks later, and at discharge</td>
<td>No definition given</td>
<td>Subluxation NC</td>
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<tr>
<td>Feys et al.&lt;sup&gt;36&lt;/sup&gt; (1)</td>
<td>75 patients; mean age 66 years; 47 male, 28 female; side of hemiplegia 50 left, 25 right</td>
<td>CS, twice in first 2 months</td>
<td>The following different shoulder problems are described: inferior subluxation, SHS, capsulitis, impingement, shoulder pain at rest and during passive movements, measured by clinical tests and/or technical investigations</td>
<td>Tactile sensibility, muscle tone Sensibility loss contributed in prediction of pain at rest (OR not given), NC</td>
</tr>
<tr>
<td>Jespersen et al.&lt;sup&gt;4&lt;/sup&gt; (3)</td>
<td>173/164 (exclusions dysphasia [1], pain before CVA [7], trauma [1]); patients with stroke admitted to hospital; 83 male, 90 female; time between stroke and HSP median = 54.5 days (range 8 to 240); dysphasia at admission (52), neglect on admission (40)</td>
<td>CS, weekly rounds, and 6-month control visit</td>
<td>Spontaneous complaints of pain during the weekly round; unclear whether pain was asked about during the 6-month control visit; in total, 38 patients complained about shoulder pain, 11 after discharge; 2 had central poststroke pain, 4 clinically rotator cuff tears, 1 SBD</td>
<td>Neglect OR not given but reported as NS</td>
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(Table continues on next page)
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<tr>
<td>Kumar et al.30 (7)</td>
<td>28 patients with cerebral infarction, admitted for stroke rehabilitation; age 64.9 years (SD = 8.2, range 43 to 90); 27 male, 1 female; time between onset of stroke and treatment 14.5 days (SD = 2.52, range 10 to 20); side of hemiplegia 12 left, 16 right</td>
<td>CS, with 3 exercise groups: OP, SB, passive ROM; follow up 3 months</td>
<td>No definition given; patients reported verbal or nonverbal pain (yes or no): 7 patients developed shoulder pain, including 1 with SRD</td>
<td>OP, SB, passive ROM, subluxation (palpation, X-ray)</td>
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<td>van Ouwenaller et al.3 (3)</td>
<td>345(7)/219 patients with hemiplegia based on CVA (79%), head injury (14%), tumors (7%); 166 male, age 47 years (range 19 to 78), flaccid 33, spastic 133; 53 female, age 46 years (range 18 to 74), flaccid 11, spastic 42</td>
<td>CC, changes in the shoulder were noted during the regular rehabilitation program; time relationship is not clear; follow-up at least 6 months (mean = 11 months)</td>
<td>Pain in hemiplegic shoulder based on subluxation, tendinitis, SRD, or SRD + subluxation; in total, 157 patients with HSP</td>
<td>Muscle tone (myotatic reflex); subluxation (X-ray shoulder?); RSD: criteria described; tendinitis (?)</td>
</tr>
<tr>
<td>Poulin de Courval et al.31 (5)</td>
<td>94 patients with hemispheric stroke; age 68 years (SD = 9.1); 51 male, 43 female; time since onset of stroke 40 days (SD = 29); time between stroke and admission 3 to 5 weeks; side of lesion in cerebro 36 right, 58 left</td>
<td>CC</td>
<td>No definition given; examined with anamnesis and physical examination; in total, 45 patients with HSP</td>
<td>Hemineglect (Alberts test, Benton test, copying drawings); sensitivity (pin prick); muscle tone (physical examination); subluxation (X-ray)</td>
</tr>
</tbody>
</table>

TABLE 1 (continued)

Description of Study Population, Study Design, Definition of Hemiplegic Shoulder Pain, and Risk Factors for Hemiplegic Shoulder Pain

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<td>Sample Description</td>
<td>Methodology</td>
<td>Evaluation Criteria</td>
<td>Results</td>
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<td>Roy et al. (3)</td>
<td>83/76 (2 died, 5 dropouts), age median = 73 years (range 41 to 99); 37 male, 39 female; time since onset of stroke median = 7 days (range 1 to 29); hemiplegia on dominant side 37, nondominant side 38; shoulder pain before stroke 9</td>
<td>CS, follow-up 12 weeks</td>
<td>No definition given; measured with vertical VAS, severity not mentioned; in total, 55 patients HSP ever</td>
<td>Malalignment (X-ray, Van Langenbeke criteria); subluxation (same); muscle tone (Asworth); RSD (Kozin criteria) OR 2.86 (pain at rest) 4.02 (for movement pain) (NS, no CI given); 11.02 (pain at rest), 4.24 (for movement pain) (NS, no CI given); NC; NC</td>
</tr>
<tr>
<td>Savage and Robertson (3)</td>
<td>26 patients with vascular disease (infarction or hemorrhage); age 69.8 years (range 40 to 87); 16 male, 10 female; time since onset CVA 4 weeks to 4 months; first CVA, no previous shoulder problems</td>
<td>CS, at start and at end; follow-up 4 weeks</td>
<td>No definition given; suspicion of SHS referred to rheumatologist (13 cases of SHS diagnosed); pain was measured daily by PT score 1 to 3</td>
<td>Subluxation (X-ray); SHS; muscle tone (physical examination) NC, only p &lt; .02 reported; NC; NC</td>
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<tr>
<td>Shai et al. (4)</td>
<td>50 consecutive patients with hemiplegia, of whom 40 with X-ray, of whom 33 in analyses</td>
<td>CC</td>
<td>Disabling shoulder pain (+ or –), no further description given</td>
<td>Subluxation (X-ray rated 1 to 3) Normal X-ray 3/14 chronic pain (not described); with V-shaped widening 8/10 chronic pain; with subluxation 6/9 chronic pain; OR V-shaped vs. not V-shaped 6.2 (4.4 to 8.0); RR V-shaped vs. normal 14.7 (12.7 to 16.7)</td>
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<tr>
<td>Wanklyn et al. (3)</td>
<td>124/108; age 71 years (range 60 to 89); 57 male, 51 female; side of hemiplegia 57 left, 46 right (5 multiple lesions); length of stay 62 days (range 5 to 289)</td>
<td>CS, measurement at discharge (t₁), 8 weeks (t₂) and 6 months after discharge (t₃); follow-up 6 months</td>
<td>Pain in hemiplegic shoulder measured with a questionnaire (no references); t₁ 39 HSP, t₂ 59 HSP, t₃ 36 HSP</td>
<td>Subluxation (palpation) NC</td>
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</table>

Note: MS, methodological score; HSP, hemiplegic shoulder pain; RR, rate ratio; OR, odds ratio; NC, not computable; CVA, cerebrovascular accident; CS, cohort study; SHS, shoulder-hand syndrome; SRD, sympathetic reflex dystrophy; NS not significant; OP, overhead pulley; SB, skateboard; ROM range of motion; CC, case-control study; LOS, length of stay; VAS, visual analogue scale; CI, confidence interval.

a Palpation and if clinically no subluxation an additional X-ray.
Impairments in sensory and cognitive functioning may increase the risk of hemiplegic shoulder pain. According to Poulin de Courval et al., neglect increases the risk of repeated traumas of the shoulder, and this may result in lesions of the surrounding soft tissues, leading to capsulitis, bursitis, tendinitis, rotator cuff tear, or rupture, which may result in hemiplegic shoulder pain. Other causes of repeated traumas are malpositioning of the patient or mishandling of the upper extremity, either during transfer or during too vigorous exercise therapy (not included in the model). Impairments in the autonomic nervous system may lead to shoulder-hand syndrome or sympathetic reflex dystrophy, and these conditions may cause hemiplegic shoulder pain. Central pain and thalamic pain are not included in the model.
model. In addition to the risk factors that are clearly related to stroke and are biological plausible, many other risk factors are also mentioned, such as age, weight, muscle strength, range of motion (ROM) of the shoulder, affected side, gender, Barthel Index score, metastasis, prestroke degenerative disorders of the shoulder, severity of stroke, depression, and time since onset.

C. Risk Factors

As mentioned before, the risk factors for which only correlation coefficients were given were excluded from Table 1. Details of the methodological scores for the individual studies are presented in Table 2, showing the total scores, varying from 0 to 7. The evidence for the various risk factors is summarized in Table 3. The relationship between subluxation and hemiplegic shoulder pain was investigated in nine studies, but statistically significant results were only found in one study. Increased muscle tone was found to be positively related to hemiplegic shoulder pain in two studies, in three other studies, no RR or OR could be computed. Exercising the passive ROM with overhead pulley (leading to repeated traumas of the shoulder) was found to increase the risk of the occurrence of hemiplegic shoulder pain. Subluxation seems to be an effect modifier of this relationship. The RR for patients with subluxation is 10 (95% CI, 1.6 to 64.2), and for patients without subluxation, the RR is 4 (95% CI, 0.5 to 34.2).

### Table 2

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<th>Study item</th>
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<td>Shai et al.</td>
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Note: UC, unclear.

* See the Appendix for descriptions of the items.

### Table 3

<table>
<thead>
<tr>
<th>Risk Factor</th>
<th>Number of Studies</th>
<th>Significant</th>
<th>Nonsignificant</th>
<th>Inconclusive*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Subluxation</td>
<td>9</td>
<td>1</td>
<td>3</td>
<td>5</td>
</tr>
<tr>
<td>Increased muscle tone</td>
<td>5</td>
<td>2</td>
<td>0</td>
<td>3</td>
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<tr>
<td>Loss of sensibility/neglect</td>
<td>3</td>
<td>0</td>
<td>2</td>
<td>1</td>
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<tr>
<td>Repeated trauma</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>SHS/SRD</td>
<td>3</td>
<td>0</td>
<td>0</td>
<td>3</td>
</tr>
</tbody>
</table>

Note: SHS, shoulder-hand syndrome; SRD, sympathetic reflex dystrophy.

* Inconclusive means statistics (risk ratio or odds ratio) not given and not computable.
IV. DISCUSSION

A model always simplifies reality, and this model for hemiplegic shoulder pain is no exception. It was decided to include only causes that were described in the selected studies and that, in the opinion of the authors, were biologically plausible and clearly related to stroke. For example, depression was not included, although it is possible that depression may lead to immobilization, which may result in changes in the soft tissues of the shoulder (frozen shoulder) and therefore cause pain. In spite of these limitations, it is thought that this model may help structure the main etiological mechanisms contributing to hemiplegic shoulder pain. On the basis of this model, a systematic review was conducted to evaluate the evidence for the postulated risk factors.

A. Role of Various Risk Factors

Combining Tables 1 and 3, it might be concluded that subluxation is not a risk factor for hemiplegic shoulder pain. The only positive study was a retrospective case control study. In two studies, increased muscle tone was found to increase the risk of hemiplegic shoulder pain; three other studies provided no data on this relationship. From these findings, the conclusion could be drawn that increased muscle tone may increase the risk of hemiplegic shoulder pain. Jespersen et al. reported an OR of 2.3 (95% CI, 1.1 to 5.1) for muscle power (total paralysis vs. nontotal paralysis), indicating that total paralysis increases the risk of shoulder pain. In patients with hemiplegia, muscle power is closely related to muscle tone. Disorders in either are related to poor motor function. To keep the model simple, it was decided to include only muscle tone. Neglect may increase the risk of repeated traumas, with resulting soft tissue lesions. However, none of the studies found a relationship between neglect (or sensory disorders) and hemiplegic shoulder pain. A major shortcoming of these studies may be that the measurement of neglect was too late: on average 40 days after stroke or at least 4 weeks after stroke. The natural course of neglect is that it is most severe immediately after stroke, usually followed by (partial) recovery.

Some etiological pathways seem quite obvious; for example, repeated traumas of the shoulder capsule may lead to capsulitis, which causes pain, or disturbances of movement in the shoulder joint may result in impingement of a muscle tendon, also causing pain. For other risk factors, the causal mechanism is not so obvious. Shoulder-hand syndrome and sympathetic reflex dystrophy may lead to pain in the hemiplegic shoulder, but the exact mechanisms are still unclear. Spasticity is mentioned as a possible risk factor for hemiplegic shoulder pain, but it remains unclear as to how increased muscle tone leads to pain. One suggestion is that it causes pain by producing long-lasting severe traction on the periost at the site of the muscle insertion.

B. Definition and Measurement of Hemiplegic Shoulder Pain

Most studies gave some definition of hemiplegic shoulder pain or the method used to measure the pain (Table 1) or described the disorders that were investigated. One abstract gave no description of the measurement of hemiplegic shoulder pain. However, there is no consensus on the definition of hemiplegic shoulder pain or the most appropriate method of measurement. Only a minority of the studies measured the severity of the pain, and no results were presented concerning the relationship between the severity of pain and the degree of a risk factor. A “dose-response” relationship could be a strong indication that the risk factor is an important etiological factor.

C. Selection of the Studies

As mentioned before, at least 16 reviews on hemiplegic shoulder pain have been published. Many references contained in these reviews could not be included in the present systematic review because the time relationship between the risk factor and hemiplegic shoulder pain was not clear. Only 11 studies were identified that investigated the role of one or more risk factors in the occurrence of hemiplegic shoulder pain. The methodological quality of the studies was
low: Only Kumar et al.\textsuperscript{30} scored more than 50% of the maximum attainable score. Poulin de Courval et al.\textsuperscript{31} were the only investigators who intended a priori to study the influence of one risk factor (neglect) on hemiplegic shoulder pain. In all other studies, the presentation of the results suggests that the study of the relationships was not preplanned. The statistics applied seem to support this impression: Correlation coefficients test only whether a linear correlation exists between two variables, and the results depend on the units used. Correlation coefficients cannot be used to estimate causal relationships.

V. SUMMARY AND CONCLUSIONS

The conclusion of this review is that there is some evidence that overhead pulley should not be applied for passive movement of the hemiplegic shoulder. Although the relationship between subluxation and hemiplegic shoulder pain was investigated in nine studies, the existence of this relationship is still unclear. Increased muscle tone seems to increase hemiplegic shoulder pain, but this relationship lacks conclusive evidence. Imprudent handling and mishandling of the shoulder, resulting in damage to the shoulder, are often mentioned as causes for hemiplegic shoulder pain,\textsuperscript{3,9,11,15,23} but none of the studies presented any results concerning these risk factors, except for the effect of overhead pulleys, which can be considered as a method of passive ROM exercise that leads to repeated traumas of the shoulder. Further research on risk factors for hemiplegic shoulder pain is recommended, with an emphasis on these risk factors, which can be modified during rehabilitation and in daily life.

REFERENCES

23. van Langenberghe HVK, Partridge CJ, Edwards MS,


APPENDIX: Criteria List for Methodological Quality

Study population
1. Positive if the main features of the study population are described (sampling frame and distribution of the population according to age, sex).
2a. Positive if cases and controls are drawn from the same population and a clear definition of cases and controls was stated (CC).
2b. Positive if participants with hemiplegic shoulder pain at baseline (or at start of the follow-up) are excluded (CS).
3. Positive if the participation rate is at least 80% of the potential participants (hemiplegics) or if the participation rate is 60% to 80% and nonresponse is not selective (data presented). Explanation: A number of consecutive patients without further information on method of selection should be rated as unclear.
4. Positive if the response at main moment of follow-up is at least 80% of the survivors or if the nonresponse is not selective (data presented).

Exposure assessment
5. Positive if the method for measuring (main) risk factors is valid for at least half of the (main) risk factors, and the same for all participants. Main factors are those factors that are of main interest according to the authors, or the factors for which results are presented. Valid means that results of a validation study are presented or that references are added to a validation study.
6. Positive if data on prestroke shoulder disorders are collected and presented.
7. Positive if the exposure assessment is blinded to disease status. If exposure assessment and disease status are measured at the same moment (and/or by the same person), this item should be rated negative.
8. Positive if the exposure is assessed at a time prior to the occurrence of the hemiplegic shoulder pain, and this exposure is used in the analyses. If this is not clearly described, this item should be rated as unclear.

Outcome assessment
9. Positive if the method for assessing shoulder pain is valid. This should be the assessment used in the analyses. Valid means that results of a validation study are presented or that references are added to a validation study.

Analysis and data presentation
10. Positive if the statistical model used was appropriate for the outcome studied and the measures of association estimated with this model were presented (including confidence intervals). Appropriate tests (depending on the type of variable): linear regression, logistic regression, odds ratios (univariate/multivariate).
11. Positive if the analyses are controlled for confounding or effect modification.
12. Positive if the number of cases in the multivariate analysis was at least 10 times the number of independent variables in the analysis.

Note: +, positive; −, negative; ?, unclear; CC, case control study; CS, cohort study.