Author's response to reviews

Title: Costs of shoulder pain in primary care: a prospective cohort study

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Author's response to reviews: see over
REVIEWS FOR MANUSCRIPT
Ton Kuijpers

Reviewer #1: Mattias Ekman

General
This is an interesting and well written paper. The purpose was to determine shoulder pain related costs during 6 months after a first consultation in general practice. The main strength of the paper is that it is based on a prospective bottom-up sample, since top-down studies cannot capture all resource items of relevance in a cost of illness study, or allow for classification of costs according to disease severity etc. Another good thing with the study is the use of a cost diary, which can help reduce the recall bias that may be a problem in retrospective studies.

Major Compulsory Revisions
First, the study does not seem to be naturalistic. The authors state in the methods section that the participating GPs were educated and trained to apply treatment according to the 1999 version of the Dutch guidelines for shoulder disorders issued by the Dutch College of General Practitioners. This has both pros and cons. On the pro side, this means that the treatment is relatively standardized and probably evidence-based. This increases the internal validity of the study. On the con side, this means that the cost of illness study does not necessarily capture treatment as it would actually be performed in Dutch primary care setting. This decreases the external validity of the study, which in turn implies that we cannot be sure that the results can be generalized to GPs who did not participate in the study, and who may not have followed the Dutch guidelines.

It is mentioned in the discussion that the health care costs estimated for shoulder pain in this study were relatively modest, and that this may perhaps be explained by GPs sticking to the relatively inexpensive wait-and-see policy (with pain medication) of the Dutch guidelines. This shows that the authors are aware of the problem, but I would still have liked to see more discussion about the consequences for the generalisability of the results of educating the GPs in using the guidelines.

We agree with the reviewer that if general practitioners outside the study do not apply treatment according to the guideline the results cannot be generalized to these practices. The guideline is not complicated and in line with daily practice (stepwise management regime). The guideline was issued in 1999, a few years before the inclusion of our study started. So, GPs who did not participate in the study could have been influenced by the guideline as much as GPs who participated in the study and followed a course during half a day. We have added some comments in the discussion paragraph on page 11.

Second, the role of co-morbidities of these patients is not clear. Cost of illness studies can measure the total health care costs of patients with the disease, or the health care costs of the disease. In the former case the costs for co-morbidities are included, and in the latter case we try to isolate the costs that are due to the condition under study. I suppose that this study tried to estimate the costs of the disease, and consequently only included costs for shoulder pain. Many of these patients had concomitant musculoskeletal complaints, however, such as pain in
the neck and in the back. How did you ensure separation in the estimation of resources consumed between, for example, shoulder pain and neck pain? And to what extent is such separation meaningful?

In every question of the cost diary we specifically asked ‘…in relation to your shoulder complaints’. In our opinion, excluding patients with concomitant musculoskeletal disorders would have negatively influenced the external validity, because patients in daily practice often do have concomitant musculoskeletal disorders.

Third, how were shoulder complaints defined at inclusion in the study? Could the relatively modest costs be explained by the inclusion of many patients with only mild symptoms? This seems implausible since a majority of the patients had a duration of more than 6 weeks prior to inclusion, but some further comments about the definition of shoulder pain used in this study would nevertheless be clarifying.

We have clarified the definition of shoulder pain in the Methods section on page 6.

**Minor Essential Revisions**

1. Neither in the title nor in the abstract do you mention that the study concerns the Netherlands. This should be mentioned in the abstract at least.

   *We have added this in the title.*

2. In the third line of the data analysis section you refer to the tables without mentioning the specific table numbers.

   *This is because this is the Methods section, in the Results section we refer to Table 2.*

3. You claim that this study is the first to evaluate the overall costs of shoulder pain in primary care, but there may have been studies including pain both in the shoulders and in the neck and the back. Did you make any comparisons with those studies?

   *We conducted a search, but could not find relevant studies, for which a comparison with the results of our study could be made.*

**Discretionary Revisions**

4. In the background you quote one-year prevalence rates for shoulder pain between 5% and 47%. To what extent are such prevalence figures meaningful? Luime et al [1] even write: ‘Health professionals and policymakers who estimate the amount of medical care needed and related costs should be aware of the variations in prevalence rate and the underlying reasons for these differences.’ Almost everybody (as indicated by the upper estimate) has pain in neck or shoulders once in a while, but this does not usually require any medical care (except perhaps rest and self-medication with cheap pain killers). The meaningful prevalence rate in this context is probably the prevalence of people with problems severe enough to require medical care visits and sickness leave. And from an economic viewpoint the chronic and recurrent cases are probably most important.

   *We agree with the reviewer. This is why we decided not explicitly extrapolate our results to a wider population. We have added a recent reference on page 5 which reports on prevalence rates for a primary care population (Bot, 2005).*
5. You only present the costs by follow-up period in weeks and by duration of symptoms. Did you also consider presenting the costs by level of pain or disability? We considered presenting the costs for patients with a worse prognosis according to a prediction rule we have recently developed (Kuijpers et al., Pain, 2006). However, further research is needed before cut-off points for a worse prognosis can be set reliably. In this study we aimed to study the overall costs of shoulder pain, without selecting subgroups. We think that in a latter stage, when results become available that enable a reliable definition of relevant subgroups, these subgroup analyses can be of importance.

6. There was no estimate of lower productivity at work as a result of shoulder pain. This is also mentioned by the authors in the discussion. It has to be said, however, that it is a matter of dispute to what extent it is possible to measure accurately the costs of lower productivity at work. This could have been discussed in more detail. Our study does not provide information on the actual loss of productivity among those who kept on working regardless of the shoulder pain (“work presenteeism”), or costs related to hiring and training replacements, spill-over effects to co-workers, and compensatory mechanisms that may reduce or increase the overall productivity loss (Jacob-Tacken KHM, Koopmanschap MA, Jan Meerding W, Severens JL. Correcting for compensating mechanisms related to productivity costs in economic evaluations of health care programmes. Health Econ 2005; 14(5):435-443). Clearly, a more adequate estimation of production loss is needed to improve the meaningfulness of future studies.

7. Maybe you should point out in Table 1 that the direct health care costs are per visit (except hospitalisations) and that direct non-health care costs and indirect costs are measured per hour. This is mentioned in the text, but many readers look first at headings and tables before reading the text. We have added this suggestion to Table 1.
Reviewer #2: Jens Ivar Brox

Major Compulsory Revisions
My first impression was that the study is well designed, including a properly defined population, using accurate outcome measures, and the other advantages include a large sample with sufficient follow up, and a nicely presented manuscript. However, the population is poorly defined including any shoulder pain, excluding only severe physical and mental conditions, diagnosed by any of 103 general practitioners. Even more important, patients who had taken sick-leave for other reasons in the previous 2 months, most likely musculoskeletal pain related to their shoulder pain, were included. Since the majority of the patients included reported concomitant musculoskeletal pain from other regions, sick leave for other reasons is a major confounder, and additional analysis should be performed excluding patients on sick leave. The results reported may overestimate the costs after first consultation for shoulder pain. This is particularly important because 12% of the patients contribute to 74% of the costs. We have added some information about the definition of shoulder pain (see comment reviewer #1). We solely measured ‘Sick leave in the previous 2 months due to shoulder pain’, which means that sick leave for other reasons did not confound our results. We think we have not been totally clear on this point and have added a comment to Table 2.

A second problem is that the outcomes reported are poorly defined. What is the interpretation of persistent symptoms? Just usual acceptable discomfort or considerable pain and disability? As presented, 46% is not meaningful. We have added some information regarding this point on page 8.

The questionnaire used to assess costs seems not validated. In addition rough estimates are given for unit costs. The most accurate costs are probably related to direct health costs assuming that reliable answers are given. The figures given in the tables leave the impression of accurately measured costs while measurements may not be reliable. The cost diary we used was tested and validated bij Goossens et al (Goossens ME, Rutten-van Molken MP, Vlaeyen JW, van der Linden SM. The cost diary: a method to measure direct and indirect costs in cost-effectiveness research. J Clin Epidemiol. 2000 Jul;53(7):688-95.). See page 6.

The classification of non-health costs with health care costs is also questionable. To me alternative therapies and over the counter medication should be classified as health-care costs. Using this classification 75% of total costs are indirect costs, but the percentage may be lower if patients on sick leave at baseline are excluded. Indeed, OTC medication and alternative therapies are health care costs, but at the start of this study costs were categorized into direct and indirect costs. Nowadays a separation is made between costs in the health care sector, costs in other sectors, patient and family costs, and costs of production losses.

Minor Essential Revisions
Abstract: Conclusion: Keep the first sentence, omit the next sentences. We have omitted these sentences.

Page 7: Explain friction costs better. Omit the sentence starting with: "Measures..’ We have explained the friction cost method more extensively on page 7.
Page 7: Why use multiple logistic regression to evaluate baseline differences? Why just not present the table without statistical analysis or use multiple logistic regression to test differences at 6 months.

The analysis was carried out to compare the characteristics of patients with incomplete cost diaries to patients with complete cost diaries, that is, to explore the risk of selective non-response to the cost diaries. We were interested to see which patient characteristics were most strongly associated with the probability of an incomplete dataset; hence the multiple logistic regression analysis. Testing differences at six months would not provide us with this information. Although, there appears no direct clinical meaning in this test result, which may arise from relatively low power, we add the results of such regression analysis to explore the magnitude of a difference in costs for those with and without complete data, because we think this still may be informative.

Page 8: omit as well.

Unfortunately, we do not understand what the reviewer wants us to omit.

Page 9: Hospitalization reflect extra costs in treatment of the shoulder (See Brox et al BMJ, 1993)

Brox et al stated in 1993 that “Treatment costs were higher for those given surgery (720 pounds v 390 pounds).” This is in line with our findings. We identified one patient in our study that underwent surgery for his shoulder complaints, and also generated extra costs (€2715).

Page 9: Rewrite after performing additional analysis.

See our response above to Major Compulsory Revisions.

Page 11: Poor prognosis is not well defined since persistent symptoms are not well defined

We have added some information on this point on page 8.

Page 12: The effectiveness of manipulative therapy. Why not report costs for manipulative therapy which is more relevant than for the control group. As I understand this paper the clinical differences could be questioned and the extra costs are 3.8 consultations by the manual therapist?!

We chose to compare our results to the control group because these patients received more or less the same treatment: usual care according to the national GP guideline for shoulder pain.

Reviewer #3: Elaine Harkness

General

This study uses a prospective design to determine the economic costs of shoulder pain in individuals who consulted their primary care physician. The literature in relation to health care and lost productivity costs of musculoskeletal pain in general is limited. Other studies are limited in that they have used prevalence data and linked this to a number of other data sources, thus the estimates are implicit on a number of assumptions. This study has been well conducted and provides evidence to suggest that the cost of shoulder pain within a Dutch primary care setting was relatively low.
Major Compulsory Revisions

Discussion

Limitations of the use of cost diaries should be discussed e.g. compliance and validity. Did the authors try to determine the validity of the cost diaries in any way e.g. by comparing reported and actual visits to general practitioners. In addition, the external validity of the study should be discussed.

The cost diary was tested by Goossens et al, 2000 (see page 6). We have added some comments on the generalisability of our results on page 10).

Minor Essential Revisions

Medication costs (prescriptions & over-the-counter) and out-of-pocket expenses are not included in the unit costs in Table 1.

This is correct, prices for over the counter medication and out-of-pocket expenses were reported by the patient. As stated on page 7, costs of medication on prescription were based on the prices provided by the Royal Dutch Society for Pharmacy.

Discretionary Revisions

Title
I feel the title is a little misleading as it is costs of shoulder in those presenting in primary care, not just costs in relation to primary care.

We add ‘consulters’ in the title.

Methods

Where costs for consultations to other primary care professionals (e.g. nurses) considered? Costs the cost information is a little confusing, it sounds as though patients were asked to complete the costs for each of these factors, when in fact it appears that patients were asked report the number of visits to various health care professionals, hours of help from others and days of sick leave etc. and that these were then applied to the unit costs in Table 1. The exception seems to be the costs for alternative therapists. If this is the case it needs to be clarified in the text and more information should be given on how unit costs were obtained.

A little more detail on the friction method would be helpful.

We have added some comments on the friction cost method on page 7 in the methods section. We also considered costs for other primary care professionals (e.g. allied health care professionals such as physiotherapists and manual therapists). In the Netherlands there are no nurses involved in the management of shoulder pain in primary care.

Data analysis

Were the analyses for patients with persistent symptoms based on self-report?

We have added some information on this subject on page 8.

Results

Include the numbers of patients who were eligible for the study but were excluded and the reasons why. The authors comment in the discussion that those patients with more serious injury (fractures, dislocation or previous surgery) were excluded from the current study but may generate substantial costs, therefore it would be interesting to know what proportion were excluded for this reason.

Unfortunately we do not have data that may gain insight in the proportion of patients who were excluded for these reasons.
(END OF REVIEWS)