1 Title: Self-Efficacy and Risk of Persistent Shoulder Pain: Results of a Classification and 2 Regression Tree (CaRT) Analysis 3 4 Rachel Chester, Mizanur Khondoker, Lee Shepstone, Jeremy Lewis, Christina Jerosch-Herold 5 6 School of Health Sciences, Faculty of Medicine and Health Sciences, University of East Anglia, 7 Norwich, Norfolk NR4 7TJ, UK. 8 Rachel Chester 9 Lecturer in Physiotherapy 10 11 Norwich Medical School, Faculty of Medicine and Health Sciences, University of East Anglia, 12 Norwich, Norfolk NR4 7TJ, UK. 13 Mizanur Khondoker 14 Senior Lecturer in Medical Statistics 15 16 Norwich Medical School, Faculty of Medicine and Health Sciences, University of East Anglia, 17 Norwich, Norfolk NR4 7TJ, UK. 18 Lee Shepstone 19 **Professor of Medical Statistics** 20 21 Department of Allied Health Professions, School of Health and Social Work, University of 22 Hertfordshire, College Lane, Hatfield AL10 9AB, UK. 23 Jeremy Lewis 24 Professor of Musculoskeletal Research 25 26 School of Health Sciences, Faculty of Medicine and Health Sciences, University of East Anglia, 27 Norwich, Norfolk NR4 7TJ, UK. 28 Christina Jerosch-Herold 29 Professor of Rehabilitation Research 30 31 Correspondence to: R Chester r.chester@uea.ac.uk 0044 (0)1603 593571 32 33 Word count: 3112 excluding text box, tables, figures and their legends. 3252 including text box.

1 **Abstract** 2 3 Objectives: To (i) identify predictors of outcome for the physiotherapy management of shoulder pain 4 and (ii) enable clinicians to subgroup people into risk groups for persistent shoulder pain and 5 disability. 6 7 **Methods:** 1030 people aged ≥18 years, referred to physiotherapy for the management of 8 musculoskeletal shoulder pain were recruited. 810 provided data at 6 months for 4 outcomes: 9 Shoulder Pain and Disability Index (SPADI) (total score, pain sub-scale, disability sub-scale) and 10 Quick Disability of the Arm, Shoulder and Hand (QuickDASH). 34 potential prognostic factors were 11 used in this analysis. 12 13 **Results:** Four classification trees (prognostic pathways or decision trees) were created, one for each 14 outcome. The most important predictor was baseline pain and/or disability: higher or lower baseline 15 levels were associated with higher or lower levels at follow up for all outcomes. One additional 16 baseline factor split participants into four subgroups. For the SPADI trees, high pain self-efficacy reduced the likelihood of continued pain and disability. Notably, participants with low baseline pain 17 18 but concomitant low pain self-efficacy had similar outcomes to patients with high baseline pain and 19 high pain self-efficacy. Cut points for defining high and low pain self-efficacy differed according to 20 baseline pain and disability. In the QuickDASH tree, the association between moderate baseline pain 21 and disability with outcome was influenced by patient expectation: participants who expected to 22 recover because of physiotherapy did better than those who expected no benefit. 23 24 Conclusions: Patient expectation and pain self-efficacy are associated with clinical outcome. These 25 clinical elements should be included at the first assessment and a low pain self-efficacy response 26 considered as a target for treatment intervention.

## What are the new findings?

- High levels of pain and disability at baseline are associated with high levels of pain and disability at 6 month follow up. However, this 'predicted' poor outcome is modified to a predicted better outcome if the patient has high pain self-efficacy and a greater expectation of treatment.
- (Pain self-efficacy is the extent or strength of the patient's belief in their ability to complete tasks and reach a desired outcome despite their shoulder pain).
- Low levels of baseline pain and disability are associated with low levels of follow up pain and disability. This predicted better outcome is modified to a predicted poor outcome if the patient has low pain self-efficacy.

# How might it impact on clinical practice in the future?

 We recommend that pain self-efficacy and patient expectation of outcome as a result of physiotherapy treatment should be formally assessed and discussed at the first physiotherapy appointment.

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2	Introduction		
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4	Persistent musculoskeletal shoulder pain is common and frequently associated with substantial		
5	disability. Over a period of one month, between 16% and 31% of the general population in the United		
6	Kingdom (UK) will have suffered from musculoskeletal shoulder pain lasting at least 24 hours. 1-3		
7	Experiencing shoulder pain concerns many and accounts for up to three percent of visits to General		
8	Practitioners (GP) annually. <sup>4,5</sup> and up to 48% of people with shoulder pain visit their GP more than		
9	once over a three year period due to ongoing symptoms. <sup>5,6</sup> The most effective treatment is not yet		
10	known; clinical trials comparing surgical and non-surgical management, including exercises		
11	prescribed by physiotherapists report equivocal effects. <sup>7-9</sup> Between 8% and 11% of patients visiting		
12	their GP with shoulder pain are referred to see a physiotherapist at initial consultation, 5,10.11 rising to		
13	18% over a three year period. <sup>5</sup>		
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15	Response to physiotherapy is variable. In a multicentre cohort study of 1030 patients with		
16	musculoskeletal shoulder pain attending physiotherapy, of mean duration 14 months (standard		
17	deviation 28 months), 69% of patients reported complete recovery or being much improved by 6		
18	months follow-up; 17% reported only slight improvement and 14% reported no change or a		
19	worsening of symptoms. 12 A multivariable general linear model (GLM) was used to identify		
20	prognostic factors associated with patient rated pain and disability. 13 Several factors were consistently		
21	associated with a better outcome at 6 months. One limitation of the GLM approach is the difficulty of		
22	practical use, particularly in a clinical setting. All predictor variables within the model are used		
23	simultaneously requiring lengthy calculations (particularly when there are many predictor variables).		
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25	Classification and Regression Tree (CART) Analysis is an alternative method of providing prognostic		
26	guidance. CART analysis considers the predictive value of prognostic factors sequentially, i.e. in a		
27	hierarchy of importance. CART typically results in a simple and readily interpretable decision 'tree'		
28	(or "what if" flow diagram). This can be graphically represented easily and requires no numeric		
29	calculations. 14 This can help guide clinicians to prioritise their initial prognostic assessment to those		
30	factors which are most influential. When modifiable, prognostic factors may become targets for		
31	interventions and inform shared decision making between clinicians and patients. The objective of		
32	these analyses was to provide clinicians with a guide to the most influential factors that predict		
33	outcome for people undergoing management for non-surgical musculoskeletal shoulder pain.		
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Methods

1 Data were available on 1030 people with shoulder pain recruited from primary and secondary care to

2 a multicentre longitudinal cohort study in the East of England, UK, between November 2011 and

3 October 2013. People aged 18 years or over were eligible to participate if they were referred to

4 physiotherapy for the management of musculoskeletal shoulder pain and complained of shoulder or

5 arm pain reproduced on movement of the shoulder. Those presenting with shoulder fractures,

traumatic shoulder dislocations, systemic source of shoulder symptoms, cervical radiculopathy or had

undergone shoulder surgery were excluded. Referral and treatment pathways were unaffected by

participation in the study. The study protocol has been published.<sup>15</sup>

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Outcome variables

11 Two validated patient rated outcome measures were collected at baseline and via postal questionnaire

at six month follow up: the Shoulder Pain and Disability Index (SPADI), 16,17 and Quick Disability of

the Arm, Shoulder and Hand (Quick-DASH). 18 The SPADI is a joint specific questionnaire designed

to measure two domains: shoulder pain and disability. Thirteen items, five comprising a pain subscale

and eight a disability subscale, are scored from zero to ten where zero represents no pain or disability

and ten represents the worst pain imaginable or so difficult it requires help. For this analysis, each

domain carried equal weighting in the overall score. The QuickDASH is an upper limb region specific

18 questionnaire that includes items related to symptoms, daily activities, sleep, social and work

function. Eleven items are scored from one to five where one represents no difficulty and five

represents unable. Each item carries equal weighting in the final score. Scores are converted to a scale

of zero to 100, where zero represents no pain or disability and 100 represents maximum pain and

disability.

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Baseline predictor variables:

25 Data for potential prognostic factors were collected prior to and during the participant's first

physiotherapy appointment using bespoke questionnaires and clinical record forms. These variables

included demographics, patient expectations and beliefs, lifestyle, general health, work, shoulder

history and presentation, and clinical examination findings. 15 All factors statistically associated with

outcome ( $p \le 0.05$ ) in at least one of the multivariable linear models from our previous analysis <sup>13</sup> were

included in the CART analysis. In addition, baseline factors measured, but not found to be statistically

significant, were entered if reported as a significant prognostic factor for outcome in reviews of other

musculoskeletal studies. 19,20 A description of the variables included in the CART analysis are detailed

in Supplementary file 1.

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Patient involvement

Patient and public representatives were involved in the design of the study, in particular, details

associated with the timing and procedures for recruiting and follow up of participants, and the design

1 and layout of questionnaires for data collection. A lay version of results, designed with patient and 2 public representatives, were disseminated to all study participants who at their final data collection 3 replied that they would like a copy. Patients were not involved in the actual recruitment or conduct of 4 the study. 5 6 Statistical analysis 7 We used regression trees algorithms, a sub-class of Classification And Regression Tree (CART), <sup>21</sup> for 8 continuous outcome variables, in our analyses. CART uses a recursive partitioning of the study 9 sample to produce sub-groups as homogenous as possible with respect to the outcome of interest. This 10 partitioning is based upon a binary split of each predictor variable. It is a more flexible approach for 11 uncovering complex variable relationships than traditional linear modelling as it does not rely on any 12 functional relationship between the outcome and predictor variables, nor does it require any 13 distributional assumption regarding the outcome variable. CART is also less sensitive to outlying 14 data, well suited for a large number of predictor variables and therefore offers a suitable alternative 15 for building prediction models where the relationships among variables are unspecified and existing 16 parametric statistical methods are not suitable to guide the model building.<sup>22</sup> The prediction accuracy 17 of CART is comparable with parametric regression models and it can be more accurate when the 18 relationship between the outcome and predictor variables is non-linear.<sup>21</sup> Furthermore, the partitioning 19 in CART can be represented graphically as an easily interpretable decision tree<sup>14</sup> that may then be 20 used to inform clinical practice. 21 22 We constructed four regression trees for each of the four outcome variables, i.e. SPADI overall score, 23 SPADI pain subscore, SPADI disability subscore and QuckDASH score respectively. We used the R 24 (R Core Team, 2015) package rpart.<sup>23</sup> For each of the 6-month outcome variables the respective 25 baseline score was included within the list of predictor variables. Including the respective baseline 26 scores, the number of predictor variables entered in different models ranged between 32 and 34. The 27 pain and disability sub-scales were not included in the total SPADI tree analysis therefore using only 28 32 variables (see Supplementary file 1 for a complete list and definitions of variables). 29 30 The procedure for building a regression tree in rpart is performed as follows:<sup>24</sup> 31 Building a tree: First, the predictor variable is found which best splits the sample into two sub-groups. 32 The 'best' is defined as the split that maximises the between groups sum-of-squares (or, equivalently, 33 minimises the within-group error sum-of-squares). This process is applied separately to each sub-34 group recursively until the subgroups either reach a minimum size (set to 7 in our analysis, the default 35 in rpart) or until no improvement can be made in the model fit.

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second stage of the procedure consists of using cross-validation to trim back the full tree. We used 10-

Pruning the tree: The resultant model is typically too complex and likely to over-fit the data. The

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1 fold cross-validation to evaluate model fit at a series of model complexities and chose the optimal 2 (pruned) tree by inspecting the plot of the cross-validated error against model complexity. Statistical 3 analyses were completed by a statistician without prior knowledge of the clinical area or results of 4 earlier analyses. Based on the cross validated predicted residual error sum of squares (PRESS) 5 statistic, these trees provided the best predicting models. Retention of more baseline variables within 6 the trees did not improve their predictive capacity. See supplementary file 2 for the plot for cross-7 validated errors versus model complexity for SPADI (total score) at 6 months. 8 9 Estimating a model and evaluating prediction accuracy on the same dataset generally over-estimates 10 model performance. It is, therefore, recommended to evaluate the model performance on an 11 independent dataset (i.e. a dataset that was not used to estimate the model). In the absence of an 12 independent test dataset for model evaluation, cross-validation approach is an alternative way to 13 create independent datasets for model assessment by holding apart a small portion of the sample for 14 model evaluation. More specifically, 10-fold cross-validation involves randomly splitting the data into 10 parts of similar size, holding aside one part  $(1/10^{th})$  of the whole sample) for testing and using the 15 16 rest (9/10th) for model estimation. The process is repeated 10 times, meaning that each of the 10 folds 17 is used as independent test set for model evaluation. Overall model performance is typically assessed 18 by calculating prediction errors on each of the 10 folds and averaging across all of these results. 19 20 Cross-validation is a widely used and acceptable way of validating model accuracy/performance and 21 we used this approach to select the optimal CART model, i.e., to select the variables that are most 22 predictive of the respective outcome variables (and also to remove those not contribution enough to 23 the prediction model). The results of this validation process ensures that our selected CART models 24 are optimal, despite not having a separate independent validation dataset.

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#### Results

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One thousand and fifty-five participants were assessed by physiotherapists and subsequently recruited and consented into the study. One thousand and thirty participants were found to be eligible for the study and provided adequate baseline data. There were no potential prognostic factors at baseline for which more than 2% of data were missing. Eight hundred and eleven participants (79%) provided outcome data at 6 month follow up. One participant was excluded due to incomplete outcome data. See flow diagram in figure 1.

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Figure 1: STROBE flow diagram. Participant recruitment and follow-up

1 All participants providing complete outcome data at six months were included in the CART analysis 2 for the SPADI and QuickDASH (n=810). There were differences between those participants who 3 provided complete data at six months and those who did not. Completers were older by a mean of 10 4 years, had greater pain self-efficacy by a mean of almost 4 out of a possible 60 points, were almost 5 twice as likely to exercise, and had a two-fold lower likelihood to report anxiety or depression. A 6 summary description of baseline characteristics for all participants' (n=1030) for each of the 34 7 variables entered into the CART analysis are provided in supplementary file 3. 8 9 Figures 2-5 represent the resulting pruned regression trees for the total SPADI, SPADI pain subscale, 10 SPADI disability sub scale, and QuickDASH at six months follow up. Three variables were identified 11 as important predictors of six-month outcomes: 1) baseline pain or disability levels, 2) pain self-12 efficacy and 3) patient expectation of "change as a result of physiotherapy treatment". All three 13 variables were collected prior to the participant's first physiotherapy attendance. Pain self-efficacy is 14 the extent or strength of the patient's belief in their ability to complete tasks and reach a desired outcome despite their shoulder pain.<sup>25</sup> Pain self-efficacy was measured using the pain self-efficacy 15 16 questionnaire (PSEQ)<sup>26</sup> which comprises of 10 items rated 0 to 6, zero representing minimum pain 17 self-efficacy and 6 representing maximum pain-self efficacy. The total score is out of 60, a higher 18 score representing higher pain self-efficacy. Patient expectation of change was collected in response 19 to the following question "How much do you expect your shoulder problem to change as a result of 20 physiotherapy treatment" and was measured on a 7 point Likert scale ranging from "completely 21 recover" to "worse than ever". 15 22 23 The first 'node' (at the top of the trees) represents the sample (i.e. all 810 participants). This then 24 divides into two, based on cut-off values for baseline pain or disability (SPADI, SPADI subscale 25 score or QuickDASH). The baseline score was therefore considered the most important variable in 26 predicting the respective six-month outcome. In addition to baseline pain or disability, each pruned 27 regression tree retained only one other variable of the 34 variables considered: baseline pain self-28 efficacy or patient expectation. Either pain self-efficacy or patient expectation led to classification of 29 participants into four subgroups. The number of participants in these subgroups ranged from 48 to 30 487. 31

Figure 2: Regression tree for total SPADI score

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Explanatory legend: Cut off points for the SPADI and PSEQ have been rounded up or down to whole numbers. The 4 boxplots at the bottom of the figure illustrate the distribution of total SPADI scores at 6 month follow up. The median SPADI score at 6 month follow up, (represented by the horizontal line

1 dissecting the box), is lowest (better outcome) in the subgroup represented by the box furthest left and 2 highest (poorer outcome) in the subgroup represented by the box furthest right. 3 4 Figure 3: Regression tree for SPADI Pain Subscale score 5 Explanatory legend: Explanatory legend: Cut off points for the SPADI Pain Subscale scores and 6 PSEO have been rounded up or down to whole numbers. The 4 boxplots at the bottom of the figure 7 illustrate the distribution of total SPADI Pain Subscale scores at 6 month follow up. The median 8 SPADI Pain Subscale score at 6 month follow up, (represented by the horizontal line dissecting the 9 box), is lowest (better outcome) in the subgroup represented by the box furthest left and highest 10 (poorer outcome) in the subgroup represented by the box furthest right. 11 Figure 4: Regression tree for SPADI Disability Subscale Score 12 13 Explanatory legend: Explanatory legend: Cut off points for the SPADI Disability 14 Subscale scores and PSEQ have been rounded up or down to whole numbers. The 4 boxplots at the 15 bottom of the figure illustrate the distribution of total SPADI Disability Subscale scores at 6 month 16 follow up. The median SPADI Disability Subscale score at 6 month follow up, (represented by the 17 horizontal line dissecting the box), is lowest (better outcome) in the subgroup represented by the box 18 furthest left and highest (poorer outcome) in the subgroup represented by the box furthest right. 19 20 The cut point for baseline SPADI scores (total, pain and disability sub-scores) at the first node of each 21 tree ranged from 62 to 75. When sub-dividing patients with lower baseline SPADI or baseline SPADI 22 pain sub scores into two groups using baseline pain self-efficacy scores, the cut off for the PSEQ was 23 40 and 41 respectively. When sub-dividing patients with higher baseline SPADI pain or disability 24 scores into two groups using pain self-efficacy scores, the cut off point for the PSEQ was consistently 25 48. 26 27 Figure 5: Regression Tree for QuickDASH 28

Explanatory legend: Cut off points for the QuickDASH scores have been rounded up or down to whole numbers. The 4 boxplots at the bottom of the figure illustrate the distribution of QuickDASH scores at 6 month follow up. The median QuickDASH score at 6 month follow up, (represented by the horizontal line dissecting the box), is lowest (better outcome) in the subgroup represented by the box furthest left and highest (poorer outcome) in the subgroup represented by the box furthest right.

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1 Table 1 and figures 2-4 show that the size of any subgroup with low pain self-efficacy ranged from 2 16% (n=127) to 20% (n=161) of participants in the cohort. The SPADI pain tree includes two 3 subgroups with low pain self-efficacy and this constitutes as 36% (n=288) of the cohort. The 4 discrimination between median outcome scores associated with different pain self-efficacy scores 5 differs between trees and baseline pain and/or disability. For example, the median difference in 6 subgroups is most marked for participants with high baseline SPADI pain subscores (≥75) and least 7 for participants with lower baseline total SPADI scores (<68). Twenty three percent (n=239) of the 8 cohort had a baseline of 41-59 on the QuickDASH, their outcomes were differentiated by their 9 expectation of "change as a result of physiotherapy treatment": the median difference between

subgroups at outcome being 23/100 on the QuickDASH.

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Table 1: Median (interquartile range) outcome (SPADI total, SPADI pain subscale, SPADI disability subscale and QuickDASH) for each subgroup for each tree.

#### SPADI tree at 6 months

Baseline	Number (%)	Median	IQR
<68 SPADI,≥40 PSEQ	487 (60)	9	3 to 23
<68 SPADI, <40 PSEQ	140 (17)	25	10 to 49
68 to 81 SPADI	135 (17)	36	13 to 60
≥82 SPADI	48 (6)	66	27 to 80
SPADI Pain tree at 6 months			
Baseline	Number (%)	Median	IQR
<75 SPADI Pain ,≥41 PSEQ	474 (58)	12	4 to 27
<75 SPADI Pain, <41 PSEQ	161 (20)	30	12 to 56
≥75 SPADI Pain, ≥48 PSEQ	48 (6)	20	12 to 56
≥75 SPADI Pain, <48 PSEQ	127 (16)	56	26 to 77
SPADI disability tree at 6 months			
Baseline	Number (%)	Median	IQR
<42 SPADI Disability	404 (50)	5	1 to 13
42 to 61 SPADI Disability	203 (25)	15	5 to 39
≥62 SPADI Disability, ≥48 PSEQ	48 (6)	13	7 to 36
≥62 SPADI Disability, <48 PSEQ	155 (19)	44	18 to 69
QuickDASH tree at 6 months			
Baseline	Number (%)	Median	IQR
<41 QuickDASH	474 (59)	9	2 to 18
41 to 59 QuickDASH, Pt expectation: CR or much improved	180 (22)	18	7 to 36

41 to 59 QuickDASH, Pt expectation: SI, same or worse 59 (7) 41 25 to 52 ≥60 QuickDASH 97 (12) 45 27 to 61

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## Validation

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External validation of the results was not possible as we were unable to identify an external dataset containing the same or similar variables. We have, however, conducted an informal internal validation of the results by partitioning the QuickDASH outcome data based on the classifications of the SPADI regression trees and comparing the distribution of QuickDASH outcome within each sub-group with that of the SPADI outcomes. Comparison of QuickDASH distributions corresponding to the total SPADI, SPADI pain and SPADI disability trees are displayed in Supplementary files 4, 5 and 6 respectively. The similarity of the pattern distributions of the SPADI outcome on the left and

QuickDASH outcome on the right demonstrate the replicability of the SPADI tree.

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## Discussion

The objective of these analyses was to identify important predictors of outcome for patients presenting with non-surgically managed musculoskeletal shoulder pain. We identified that only three of 34 baseline variables considered in the classification trees were predictive of outcome. These were i) baseline pain or disability measured by the SPADI or QuickDASH, ii) pain self-efficacy measured by the PSEO, <sup>26</sup> and iii) patient's expectation of "change as a result of physiotherapy treatment", measured on a 7-point Likert scale. 15 As expected, there was a positive association between pain and disability at baseline and at six month follow up, i.e. those with higher scores at baseline tended to have higher scores at follow-up. However, in all three SPADI classification trees higher pain selfefficacy influenced this relationship: for patients with high baseline pain or disability (cut off points 75 and 62 respectively), higher pain self-efficacy (PSEQ≥48) reduced the likelihood of continued high levels of pain and disability at six-month follow up. Between 16 and 19% of participants were at risk of continued high levels of pain and disability (measured by SPADI pain and disability subscores) at 6 months due to i) high baseline pain and disability and ii) low pain self-efficacy. For patients with moderate levels of baseline pain and disability measured with the QuickDASH (41 to 59), the association was influenced by patient expectation: participants who expected to completely recover or much improve as a result of physiotherapy did better than patients who expected to only slightly improve, stay the same or worsen. Participants at risk of continued high levels of pain and disability at six-month follow up due to a lower expectation of recovery constituted 7% of our cohort at six month follow up.

1 Pain self-efficacy also influenced outcome for patients with low levels of baseline pain and disability:

2 for patients with low baseline SPADI and SPADI pain scores (<68 and <75 respectively), low pain

3 self-efficacy (PSEQ<40 and <41 respectively) increased the likelihood of persistent pain. Perhaps

surprisingly, patients reporting low baseline pain but low pain self-efficacy (n=161, 20% of cohort)

had a similar or worse outcome on the SPADI pain subscale to patients with high baseline pain but

high pain self-efficacy (n=48, 6% of cohort).

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Our regression tree analyses provide a useful and simple clinical guide, highlighting the influence of

9 patient beliefs and expectations of treatment on outcome, irrespective of baseline pain and disability.

Whilst these finding are consistent with those from the GLM analysis, <sup>13</sup> the CART analysis selects

variables based on prediction power rather than statistical significance or p values. Variables are

included in order of importance; the most predictive variable is included first, the analysis then

searches for the second most important variable among the rest, and so on. The prediction error curve

estimated using cross-validation gives a clear indication at what point in the selection process the

additional predictors are not contributing enough to the prediction model. The prediction based

variable selection combined with cross-validation for assessing model performance ensures that only

the relevant and most predictive variables are included in the optimal model.

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CART analysis has advantages over traditional regression modelling in that it does not require a specified distribution of outcome data or a large sample size<sup>27</sup>. In terms of predictive power CART analysis is comparable to traditional modelling.<sup>21</sup> However CART does have limitations. Defining subgroups based on data driven cut-points for continuous measures (i.e. the PSEQ) is subject to sampling variability, but the CART methodology does not provide a measure of uncertainty (e.g., standard errors or confidence intervals) associated with the cut-off points. A different cut off point may be selected in a different sample, but it was not our intention to provide a ready to deploy clinical tool with definitive cut-off points at this stage. We rather aimed to demonstrate that an easily interpretable prediction tool with potential for clinical applications can be developed which can be further examined in bigger and external cohorts to derive more generalisable cut-offs. However, use of cross-validation approach for model selection should make the derived models sufficiently robust at least for the population represented by the study cohort. Also, being a multicentre study with broad eligibility criteria increases the generalisability of the results to the wide range of patients and

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With regards to non-surgically managed shoulder pain, this study is one of only two known using a CART analysis to investigate the hierarchy of predictive factors associated with outcome. Vergouw et

presentations of shoulder pain commonly seen by physiotherapists within primary and secondary care.

This is further supported by similar patterns of the distributions of the QuickDASH outcome based on

classification of participants using the SPADI trees in our informal internal validation.

1 al<sup>28</sup> compared the results of CART and logistic linear regression for 587 patients with musculoskeletal 2 shoulder pain attending General Practice in the Netherlands, however, they did not include patient 3 expectation of change as a result of physiotherapy and pain self-efficacy. A positive association 4 between patient expectation and outcome has been consistently reported for a range of health 5 conditions, <sup>29-31</sup> although ours is the first to investigate patient expectation of outcome in nonsurgically managed shoulder pain. The association between pain self-efficacy and chronic non-cancer 6 7 pain has also been consistently reported for a range of health conditions.<sup>32</sup> Ours is one of only two 8 studies to investigate self-efficacy in non-surgically management for shoulder pain. A randomised 9 controlled trial of 102 participants, <sup>33</sup> did not find an association between baseline pain self-efficacy 10 and the outcome of supervised exercise or radial extracorporeal shockwave therapy. 11 Based on our findings that pain self-efficacy and patient expectation are important predictors of 12 outcome we recommend that they be formally assessed in all patients with musculoskeletal pain. 13 There is currently no standardised method of measuring patient expectation and we therefore 14 recommend using a patient rated Likert scale that includes a worsening as well as improvement of 15 shoulder pain.<sup>29</sup> There are several validated measurement tools for pain self-efficacy and for the busy 16 clinician we recommend using shortened patient rated versions such as the PSEQ-2<sup>34</sup> comprising two 17 items. Standardised questionnaires like the PSEQ-2 and a single question on expectation of outcome 18 provide an opportunity to openly discuss patient beliefs and expectations which healthcare 19 practitioners may find challenging otherwise. 35, 36 Such patient-clinician dialogues around the 20 potential impact of expectations and beliefs further supports shared decision-making. Our results 21 suggest that cut points will vary according to baseline pain and disability and therefore the use of 22 specific cut-points for stratification is not justified. Further research is also needed to validate our 23 point estimates in an external cohort. 24 It is plausible that patient expectation and pain self-efficacy are mediating factors.<sup>29</sup> Adherence to 25 non-surgical management is reportedly low.<sup>37</sup> The therapeutic effect of a home exercise and/or self-26 management programme cannot be realised if not enacted by the patient.<sup>38</sup> One of the suggested 27 mechanisms by which higher patient expectation is associated with outcome is through an increased 28 motivation to engage and adhere to an intervention that participants believe will have a beneficial 29 outcome.<sup>39</sup> 30 Although not previously reported for those experiencing shoulder pain, high self-efficacy has been shown to be significantly associated with greater exercise adherence 40-42 as well as other health 31 behaviours such as physical activity<sup>43-45</sup> and taking medications as prescribed.<sup>46</sup> A consistent and 32 33 statistically significant association between all three factors; changes in self-efficacy, adherence and

- 1 outcome, has yet to be demonstrated. Further studies are needed to explore if moderating self-efficacy
- 2 affects outcome.
- 3 Further development and testing of, educational interventions targeting healthcare practitioners with
- 4 strategies to increase patients' pain self-efficacy and expectations of treatment is needed. A number of
- 5 promising interventions exist for increasing patients' self-efficacy and include positive feedback on
- 6 performance, observation of mastery in others, graded activity, identifying realistic goals for which
- 7 the patient is likely to succeed and selecting tasks and activities relevant to the patient. <sup>32,43</sup> Variability
- 8 in reported effectiveness suggests that the purpose, content and delivery may need to be tailored to
- 9 each patient, requiring a person centred approach.

## Conclusion

- 12 This is the first known study to subgroup people with shoulder pain of musculoskeletal origin
- 13 attending physiotherapy into risk groups for persistent pain and disability based on a range of baseline
- personal, clinical, activity, and participatory variables. This multicentre study provides evidence that
- for a given baseline measure of shoulder pain and disability, pain self-efficacy and patient expectation
- of change as a result of physiotherapy, are the most influential predictors of patient rated outcome at
- six month follow up. Additionally, this is the first study to demonstrate that for people with shoulder
- pain higher pain self-efficacy reduced the likelihood of continued high levels of pain and disability at
- six-month follow up, for those with high baseline pain or disability. The likelihood of persistent pain
- increased in the subgroup that were categorised as having low levels of baseline pain and disability
- 21 and concomitant low pain self-efficacy. Of importance those identified as having low baseline pain
- and low self-efficacy had similar or worse outcome on the SPADI pain subscale to those with high
- baseline pain and high pain self-efficacy.

- 25 Although our findings are applicable to people referred to physiotherapy for the management of
- shoulder pain of any duration and in primary and secondary care, they are likely to be applicable
- beyond this group.
- 28 Based on our findings we suggest that pain self-efficacy and patient expectation should be formally
- assessed and discussed at the first physiotherapy appointment. Further research should investigate
- 30 whether these factors can be targeted and modified by therapeutic interventions and improve patient
- 31 outcomes.
- 32 **Declaration of competing interests:** All authors have completed the Unified Competing Interest
- form at www.icmje.org/coi disclosure.pdf (available on request from the corresponding author) and
- declare that (1) RC had support from a National Institute of Health Clinical Doctoral Research
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1 have an interest in the submitted work in the previous 3 years; (3) the authors' spouses, partners, or 2 children have no financial relationships that may be relevant to the submitted work; and (4) the 3 authors have no non-financial interests that may be relevant to the submitted work." 4 5 Contributors: RC conceived the study, RC, LS, JL and CJH authors contributed to the design of the 6 study. RC directed the study and managed data acquisition. RC had full access to all of the data in the 7 study and takes responsibility for the integrity of the data. MK undertook and LS provided expert 8 advice on the statistical analyses. RC and MK drafted the manuscript. LS, JL and CJH provided 9 additional important intellectual and substantial scientific input throughout the study and on all 10 redrafts of the report. 11 12 Ethics approval: This was obtained from the National Research Ethics Service, East of England -13 Norfolk, UK in July 2011 (reference 11/EE/0212). 14 15 Funding: RC was funded by the National Institute for Health Research (NIHR CAT CDRF 10–008). 16 CJH was funded by the National Institute for Health Research (NIHR- SRF-2012-05-119). The 17 funders of the study had no role in study design, data collection, data analysis, data interpretation, 18 writing of the report or decision to submit the article for publication. The corresponding author had 19 full access to all the data in the study and had final responsibility for the decision to submit for 20 publication. The views and opinions expressed therein are those of the authors and do not necessarily 21 reflect those of the NIHR, NHS or the Department of Health. 22 23 Transparency declaration: The lead author (the manuscript's guarantor) affirms that this manuscript 24 is an honest, accurate, and transparent account of the study being reported; that no important aspects 25 of the study have been omitted; and that any discrepancies from the study as planned (and, if relevant, 26 registered) have been explained. 27 28 This article distributed in accordance with the Creative Commons Attribution Non Commercial (CC 29 BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-30 commercially, and license their derivative works on different terms, provided the original work is 31 properly cited and the use is non-commercial. See: http://creativecommons.org/licenses/by-nc/4.0. 32 33 Data sharing: No additional data available. 34 35 **Patient involvement:** Patient and public representatives were involved in the design of the study, in 36 particular, details associated with the timing and procedures for recruiting and follow up of 37 participants, and the design and layout of questionnaires for data collection. A lay version of results,

designed with patient and public representatives, were disseminated to all study participants who at

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- 1 Explanatory legend: Cut off points for the QuickDASH scores have been rounded up or down to
- whole numbers. The 4 boxplots at the bottom of the figure illustrate the distribution of QuickDASH
- 3 scores at 6 month follow up. The median QuickDASH score at 6 month follow up, (represented by the
- 4 horizontal line dissecting the box), is lowest (better outcome) in the subgroup represented by the box
- 5 furthest left and highest (poorer outcome) in the subgroup represented by the box furthest right.