UNIVERSITEIT AMSTERDAM

VU Research Portal

Differences in Patient Characteristics, Number of Treatments, and Recovery Rates Between Referred and Self-referred Patients With Nonspecific Neck Pain in Manual Therapy

Mutsaers, Bert J.; Janssen, Floris J.F.; Koes, Bart W.; Pool-Goudzwaard, Annelies; Verhagen, Arianne P.

Link to publication in VU Research Portal

citation for published version (APA)
Mutsaers, B. J., Janssen, F. J. F., Koes, B. W., Pool-Goudzwaard, A., & Verhagen, A. P. (2020). Differences in Patient Characteristics, Number of Treatments, and Recovery Rates Between Referred and Self-referred Patients With Nonspecific Neck Pain in Manual Therapy: A Secondary Analysis. *Journal of Manipulative and Physiological Therapeutics*, *43*(6), 559-565. https://doi.org/10.1016/j.jmpt.2019.10.008

ORIGINAL RESEARCH

Differences in Patient Characteristics, Number of Treatments, and Recovery Rates Between Referred and Self-referred Patients With Nonspecific Neck Pain in Manual Therapy: A Secondary Analysis

Bert J. Mutsaers, PT, MSc, [a, b, c] Floris J.F. Janssen, MD, [b] Bart W. Koes, PhD, [b, d]
Annelies Pool-Goudswaard, PhD, [a, e] and Arianne P. Verhagen, PhD [b, f]

ABSTRACT

Objective: In various countries, patients can visit a physiotherapist via self-referral. The aims of this study were to evaluate whether there are differences between individuals with nonspecific neck pain who consult a manual therapist via self-referral and those who do so via referral by a physician concerning patient characteristics, number of treatments, and recovery; and whether (self-)referral is associated with recovery.

Methods: This study is part of a prospective cohort study with posttreatment and 12-month follow-up in a Dutch manual-therapy setting. Adult patients with nonspecific neck pain were eligible for participation. Baseline measurements included demographic data and data concerning neck pain. At follow-up, number of treatments, recovery, and satisfaction were assessed. To evaluate differences between the groups, we used the χ^2 test and the independent t test. A logistic regression analysis was used to evaluate the association between referral status and recovery.

Results: In total, 272 manual therapists participated and 1311 patients were included. Of 831 patients whose referral data are available, about half patients consulted a manual therapist by self-referral. The mean number of treatments was 5.4, which did not differ between the 2 groups. We found no differences between the groups concerning age, sex, pain intensity at baseline, or recovery rate. Patients in the self-referral group experienced acute neck pain more frequently, had recurrent complaints more often, and reported less disability compared to the referred group. Referral status was not associated with recovery.

Conclusion: We found several small differences between self-referred and referred patients. (J Manipulative Physiol Ther 2020;43;559-565)

Key Indexing Terms: *Neck Pain; Referral and Consultation*

INTRODUCTION

Neck pain is defined as pain in the neck that lasts at least 1 day.[1] It is the sixth leading global cause of disability, ranking among the top 10 causes of disability worldwide.[2] The mean point prevalence of nonspecific neck pain is 14%, the mean 1-year prevalence is 26%, and the 1-year incidence ranges from 10% to 21%.[3] In the Netherlands, costs associated with neck pain represent 1% of health care expenditures and the number of people experiencing neck pain are predicted to increase to 50% by 2040.[4] The prognosis of patients presenting with an acute episode of neck pain in

[a] Institute for Master Education in Manual Therapy, SOMT University of Physiotherapy, Amersfoort, the Netherlands.

[b] Department of General Practice, Erasmus University Medical Center, Rotterdam, the Netherlands.

[c] Department of Physiotherapy, Avans University of Applied Sciences, Breda, the Netherlands.

[d] Center for Muscle and Joint Health, University of Southern Denmark, Odense, Denmark.

[e] Faculty of Human Movement Sciences, Vrije Universiteit Amsterdam, Amsterdam, the Netherlands.

[f] Discipline of Physiotherapy, University of Technology Sydney, Sydney, Australia.

Corresponding author: Arianne P. Verhagen, PhD, University of Technology Sydney, Graduate School of Health, Discipline of Physiotherapy, Ultimo 2007, Sydney, Australia..
(e-mail: *Arianne.verhagen@uts.edu.au*).

primary care is poor, as 47% still experience symptoms after 1 year.[5]

Patient self-referral, or direct access, means that patients can be examined, evaluated, and treated by physiotherapists without the requirement of a physician's referral. Since January 2006, patients in the Netherlands can consult a physiotherapist or manual therapist without referral. This decision was evaluated 5 years later using data from a longitudinal database registry in Dutch primary care.[6] It was found that the number of individuals with musculoskeletal disorders who consulted a physiotherapist using self-referral increased from 27.8% in 2006 to 44.2% in 2010 and 56% in 2017.[7] Furthermore, a slight difference was found between referred and self-referred patients in the number of treatments. Self-referred patients needed on average 3 treatment sessions fewer than referred patients, about 10 versus 13.[6] A recent systematic review found that self-referred patients needed fewer physiotherapy treatments and visits to physicians, less imaging performed, and fewer nonsteroidal anti-inflammatory drugs and secondary care referrals.[8] The self-referred patients were quite often younger, with a higher level of education, and mostly they presented a less severe clinical condition and a more acute complaint. The systematic review suggests that self-referral to physiotherapy is feasible, safe, and cost-efficient.[8]

Manual therapy (or musculoskeletal physiotherapy) is considered a specialized physiotherapy treatment in the Netherlands. Manual therapists focus predominantly on spinal complaints and frequently perform spinal manipulations and mobilizations aimed at reducing the time to recovery.[9] It remains unclear whether the differences found between referred and self-referred patients in physiotherapy also hold in manual-therapy practice.

Therefore, this study aims to evaluate whether there are differences (2-tailed) between individuals with nonspecific neck pain who consult a manual therapist via self-referral and those who do so via referral by a physician concerning patient characteristics, number of treatments, and recovery; and whether (self-)referral is associated with recovery after treatment.

METHODS

Design

This study is part of a prospective cohort study (the Amersfoorts Nekonderzoek of the Master Manuele therapie Opleiding [ANiMO]) of individuals with neck pain consulting a manual therapist, with posttreatment and 12-month follow-up. Ethical approval was obtained from the Medical Ethical Committee (MEC-2007-359) of the Erasmus University Medical Center, Rotterdam, the Netherlands.

Participants

Manual Therapists.   In total, 279 manual therapists (MTs) attending an educational program were asked to participate in this study; all of them participated as part of the course. All therapists were licensed MTs registered by the Royal Dutch Society for Physical Therapy. They were all working in primary or secondary health care settings. We consider this a random sample of Dutch MTs, as all MTs have to follow this educational program to keep their license.

Patients.   All participating MTs were asked to include at least 5 patients aged 18 years and over who consulted them for neck pain between November 2008 and April 2009. Excluded were all patients with known self-reported specific causes of neck pain (eg, known vascular or neurological disorders, neoplasms, rheumatic conditions, referred pain from internal organs).

Baseline Measurement

Manual Therapists.   Sociodemographic and professional data were collected at baseline and comprised sex, age, occupational setting, number of hours at work, and number of years of experience with the management of patients with nonspecific neck pain. Furthermore, during each treatment session the MTs registered in the patient's treatment diary the number of treatments, their process of clinical reasoning, and the chosen treatment modalities. MTs gathered this data independently from the patient.

Patients.   All patients filled in a baseline questionnaire independently including age, sex, pain intensity (using the Numeric Rating Scale [NRS]), duration of complaint (acute, subacute, or chronic), recurrent complaints (yes/no), medication use (yes/no), work status (yes/no), disability (using the Neck Disability Index [NDI] and Neck Bournemouth Questionnaire [NBQ]), fear avoidance (using the Fear-Avoidance Beliefs Questionnaire [FABQ]), and whether they had previous experience consulting a MT (yes/no).[10-14]

The NRS measures momentary pain intensity; it is an 11-point scale ranging from 0 (no pain) to 10 (unbearable pain). The NDI is a questionnaire consisting of 10 items that deal with the limitation caused by the complaint, both in work-related and non−work-related activities. For each item, the degree of limitation is determined from 0 (no limit) to 5 (huge constraint). All scores are added up and converted to percentages reflecting the degree of disability. The NBQ highlights the biopsychosocial dimensions of pain; behavior and environment affect the development, progress, and perception of pain. The NBQ is a questionnaire consisting of 7 items in which each item can be displayed on an 11-point scale ranging from 0 to 10, with higher scores indicating more pain or limitation for the given activity. Ultimately, the total score is calculated by taking the sum of the 7 items in a range of 0 to 70. The FABQ measures the extent to which physical activities (FABQ-PA) and work-related activities (FABQ-W) affect the pain. The questionnaire consists of 16 items, each measured on a 7-point scale (ranging from 0-6) indicating the extent to which it affects the pain. The first 5 questions

relate to the extent the physical activity affects the pain, with a total FABQ-PA score ranging from 0 to 30. The remaining 11 questions are related to the degree to which work influences the pain, with a total FABQ-W score ranging from 0 to 66. The higher the score, the more the activities influence the pain.

Posttreatment Measurement

Manual Therapists.   At the end of the treatment episode, the MT assessed the number of treatment sessions and reported in the treatment diary the reason for stopping the treatment episode.

Patients.   At the end of the treatment episode, patients completed a posttreatment questionnaire including the NRS, NDI, NBQ, and FABQ. Recovery of the complaint and treatment satisfaction were both measured using the Global Perceived Effect (GPE) scale.[15-17] The GPE-recovery scale asks the patient to rate, on a 7-point numerical scale, how much their condition has improved or deteriorated since baseline; it ranges from totally recovered to worse than ever. The GPE-satisfaction scale indicates, on a 7-point numerical scale, how satisfied the patient is with the received treatment. For this question the scale ranges from absolutely satisfied to absolutely not satisfied.

All patient data were gathered using paper-based questionnaires. A research assistant entered the data in SPSS statistical software and we performed a random 10% check for mistakes. To collect the data from the MTs, a custom-made digital survey was carried out. Personal log-in codes were provided per MT during the educational program. MTs had access only to their own data. Only the principal investigator had access to all personalized data, and MTs' data were recoded as numbers. All analyses were performed on coded data.

Analysis

To summarize the baseline data, we used descriptive statistics. We present data on the total group and the self-referral and referral groups. The duration of the complaint was divided into acute (0-6 weeks), subacute (6 weeks to 3 months), and chronic (longer than 3 months). The recovery data were dichotomized into "recovered" (scores: "completely recovered" and "much improved") and "not recovered"; and for satisfaction, into "satisfied" (scores: "absolutely satisfied" and "very satisfied") and "not satisfied."

Next, the difference between the self-referral and referral groups at baseline was tested. For the dichotomous variables, we used the χ^2 test, and for the continuous variables, we used the independent *t* test. We checked whether the continuous data were normally distributed using the Shapiro-Wilk test. In case of data that were not normally distributed, we used a nonparametric test (Mann-Whitney U test) for assessing (median) differences.

Lastly, we evaluated whether referral is 1 of the predictors of recovery in a logistic regression model, using backward Wald regression. Predictors were selected based on the literature (age, gender, duration of complaint, recurrent complaints, pain [NRS], and function [NDI]).[3,5] Some extra predictors were added to explore their association with recovery (referral, number of treatments, and previous experience [expectancy of the patient]). In the selection we aimed to comply with at least 10 predictors per case in the smallest group, meaning a maximum of 9 predictors. We checked a priori multicollinearity between the predictors using the correlation matrix. All analyses were done in SPSS 24.

Results

Participants

Manual Therapists.   In total, 272 MTs participated, including 1 to 5 patients each. The MTs provided data on the number of treatments for 1090 patients, and data on referral for 831 (76.2%) of them; for 259 patients, data on referral were missing.

Patients.   In total, 1311 patients are included in the cohort, of which 1190 provided data at baseline. The mean age of the patients was 44.7 years, and 69.4% were female (Table 1). Almost half of the patients had chronic complaints (47.9%), and more than half mentioned that their complaints were recurrent (66.9%). The average pain intensity was moderate (4.8 on the 11-point NRS), as was the average disability measured with the NDI and NBQ (Table 1). Not all continuous data were normally distributed.

Follow-up

After treatment, 747 patients (62.8%) provided data, with the majority stating themselves to be recovered (61.6%) and satisfied with the treatment (71.2%; Table 1). The mean (SD) number of treatments was 5.4 (2.6). The range of number of treatments was 1 to 32, with a median of 5 (Fig 1).

Referral

Of all 831 patients with information on referral, 413 (49.7%) consulted the MT via self-referral, 372 (44.8%) were referred to the MT by their general practitioner, 45 by a medical specialist, and 2 by their company doctor. Table 1 presents the differences at baseline and follow-up between the self-referral and referral groups. Overall, most baseline variables are comparable. About one-third of the people had previously been to a MT, but the number was slightly lower in the self-referral group compared with the referred patients (mean difference = 5.7%). In the self-referral group, patients on average had acute complaints more often (mean

Table 1. *Patient Characteristics*

Characteristic	Self-referral (n = 413)	Referral (n = 418)	Total (n = 1311)
Baseline			
Age, y (n = 1170), mean (SD)/median	44.5 (13.6)/44	46.2 (14.5)/46	44.7 (13.7)/44
Sex (n = 1186), male (%)/female (%)[a]	116 (30.5)/296 (69.5)	123 (31.5)/295 (68.5)	363 (30.6)/768 (69.4)
Pain intensity (NRS 0-10; n = 1183), mean (SD)/median	4.7 (2.1)/5	4.9 (2.1)/5	4.8 (2.1)/5
Duration of the complaint (n = 1071), number (%)[a]			
Acute (<6 weeks)	149 (42.8)	126 (35.5)*	420 (39.2)
Subacute (6 weeks to 3 months)	45 (12.9)	40 (11.3)	138 (12.9)
Chronic (>3 months)	154 (44.3)	189 (53.2)*	513 (47.9)
Recurrent complaint (n = 1129), yes (%)[a]	256 (70.3)	227 (60.9)*	755 (66.9)
Use of medication (n = 1190), yes (%)[a]	173 (45.4)	202 (51.5)	560 (51.6)
Work status (n = 1163), yes (%)[a]	291 (77.8)	278 (72.4)	896 (77)
Smoking (n = 1190), yes (%)[a]	92 (24.1)	97 (24.7)	300 (25.2)
NDI (0-100; n = 1096), mean (SD)/median	10.7 (8.8)/4	13.6 (10.6)/6*	12.3 (9.7)/12
Previous experience with MT (n = 1169), yes (%)[a]	126 (33.3)	150 (39)	407 (34.8)
NBQ (0-70; n = 1171), mean (SD)/median	26.7 (12.2)/26	28.8 (13.4)/28*	28.3 (12.9)/28
FABQ-PA (0-30; n = 1103), mean (SD)/median	12.3 (7.4)/13	13.9 (7.5)/14*	13.2 (7.3)/13
FABQ-W (0-66; n = 1129), mean (SD)/median	11.9 (11.3)/10	15.3 (13.6)/12	13.4 (12.2)/13
After treatment			
Number of treatments (n = 1092), mean (SD)	5.3 (2.5)	5.6 (2.5)	5.4 (2.6)
Recovery (n = 730), yes (%)[a]	158 (64)	141 (58.3)	450 (61.6)
Satisfied (n = 747), yes (%)[a]	179 (71)	176 (71.5)	532 (71.2)

Owing to missing data, percentages may not total 100%.
FABQ-PA, Fear-Avoidance Beliefs Questionnaire, Physical Activities; *FABQ-W*, Fear-Avoidance Beliefs Questionnaire, Work-Related Activities; *NBQ*, Neck Bournemouth Questionnaire; *NDI*, Neck Disability Index; *NRS*, Numeric Rating Scale; *SD*, standard deviation.
[a] χ^2 test used;
* $P < .05$.

difference = 8.9%), experienced less disability (mean difference = 2.9 on a 0-100 scale), and experienced recurrent complaints more often (mean difference = 9.6%). These differences are small but reached statistical significance.

At follow-up, the number of treatments was comparable between the groups. Most patients were satisfied with their treatment, and there was a slight difference in recovery in favor of the self-referral group (mean difference = 5.7%).

For the regression analysis all correlations were below 0.46, so no multicollinearity was present. Furthermore, referral was not a predictor for recovery when evaluated in a prognostic model (Table 2). The explained variance of the model was low at 7.2%. This model showed that male patients with an acute complaint and low disability at baseline have the best chance to recover.

Discussion

Main Findings

In Dutch manual-therapy practice, about half of patients consult the MT via self-referral. This group of patients
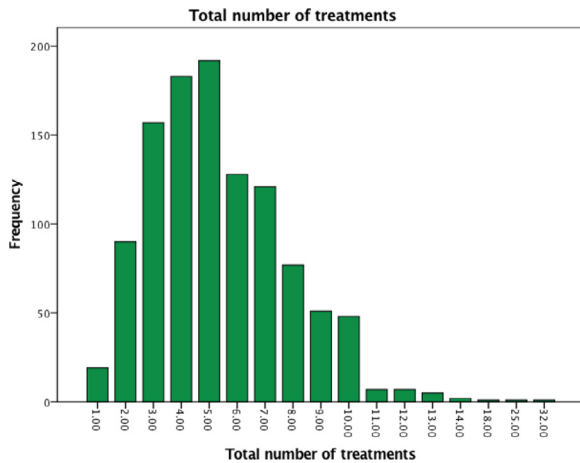
Fig 1. *Treatment frequency in number of times treated.*

more often has acute and recurrent complaints and less disability compared with referred patients. These differences are small, all below 10%.

Comparison With the Literature

Our finding on the percentage of self-referrals is consistent with findings from a longitudinal database registry in Dutch primary care (NPCD).[6] In contrast with other studies, we found no difference in number of treatment sessions, age, or sex between self-referred and referred patients.[8] We

found, for example, no differences in treatment numbers compared with direct access in physiotherapy, which might lead to the assumption that direct access might not impact health care costs as much as in physiotherapy. Like the findings in the systematic review, we also found that self-referred patients more often presented to the MT with acute complaints. In comparison to referred patients, self-referred patients reported slightly less often that they had previous experience with an MT. This has not been evaluated before, but our assumption was that if patients had a good experience with treatment by an MT, they would probably more frequently self-refer to the MT for new or recurrent complaints. This assumption does not hold in our data.

When compared with the NPCD, the average number of treatments in our study was much lower.[6] We found an average of 5.4 treatment sessions, compared with 10-13 in the NPCD. Our finding is comparable with the findings in a recent randomized clinical trial,[9] where the average number of manual-therapy treatments was 6.1 and the average number of physiotherapy treatments was 10. It might be that because of the low number of treatments, we were unable to find a difference between referred and self-referred patients.

Strengths and Limitations

This is 1 of the largest prospective cohort studies in individuals with nonspecific neck pain. A limitation of this study is the amount of missing data. Data come from

Table 2. *Prediction of Recovery*

Variable	Beta	OR (95% CI), Complete Model	OR (95% CI), Model Based on Backward Wald Regression
Number of treatments (continuous)	−0.03	0.97 (0.88-1.06)	
Referral (yes)	−0.15	0.86 (0.56-1.34)	
Age (continuous)	0.005	1.0 (0.99-1.02)	
Gender (male)	0.44	1.62 (1.0-2.62)	1.54 (0.97-2.46)
Recurrent complaint (yes)	−0.20	0.82 (0.51-1.34)	
Expectancy (yes)	−0.02	0.98 (0.61-1.58)	
Pain intensity (continuous)	−0.08	0.92 (0.81-1.04)	
NDI sum score (continuous)	−0.07	0.95 (0.90-1.0)	0.93 (0.89-0.97)
Duration of complaint (acute)	0.60	1.88 (1.16-3.05)	1.83 (1.15-2.91)
Performance measures			
Constant		0.740	0.367
Explained variance		8.6%	7.2%
Hosmer-Lemeshow test		*P* = .99	*P* = .764

NDI, Neck Disability Index; *OR*, odds ratio.

2 different sources: the MTs' treatment diaries and the patients themselves at baseline and follow-up. For several patients we had only treatment data from the MT; these patients, although they filled out an informed consent, did not complete any questionnaire. At baseline some patients did not fill in all questions, for instance on age and gender; others did not fill in the questionnaires on disability or fear avoidance. In addition, we suffered a loss to follow-up of 37.2%. This nonresponse leads to incomplete data, and estimates are less precise and statistical analysis has less power. If the dropout is selective, the nonresponse can lead to a systematic distortion of the results, but we have no indication of selective dropout in this cohort.

Conclusion

This study showed that there were several statistically significant but small differences between the self-referral and referral groups. In general, self-referred patients reported less disability and more often recurrent and acute complaints when consulting an MT. Self-referred patients had similar average numbers of treatment sessions and recovery rates to referred patients.

Acknowledgments

Funding Sources and Conflicts of Interest

Contributorship Information

Concept development (provided idea for the research): B.W.K., A.P.-G., A.P.V.
Design (planned the methods to generate the results): A.P.V.
Supervision (provided oversight, responsible for organization and implementation, writing of the manuscript): B.W.K., A.P.-G., A.P.V.
Data collection/processing (responsible for experiments, patient management, organization, or reporting data): B.J.M., F.J.F.J.
Analysis/interpretation (responsible for statistical analysis, evaluation, and presentation of the results): B.J.M., F.J.F.J., A.P.V.

Literature search (performed the literature search): B.J.M., F.J.F.J.
Writing (responsible for writing a substantive part of the manuscript): B.J.M., F.J.F.J., B.W.K., A.P.-G., A.P.V.
Critical review (revised manuscript for intellectual content, this does not relate to spelling and grammar checking): B.J.M., F.J.F.J., B.W.K., A.P.-G., A.P.V.

Practical Applications
- At baseline, self-referred patients had more acute complaints and less disability compared to referred patients.
- Baseline differences between referred and self-referred patients were not related to recovery rate.
- Self-referred patients did not differ from referred patients concerning age, gender, pain intensity, or number of treatments.

References

1. Hoy D, March L, Woolf A, et al. The global burden of neck pain: estimates from the Global Burden of Disease 2010 study. *Ann Rheum Dis*. 2014;73(7):1309-1315.
2. GBD 2017 DALYs and HALE Collaborators. Global, regional, and national disability-adjusted life-years (DALYs) for 359 diseases and injuries and healthy life expectancy (HALE) for 195 countries and territories, 1990-2017: a systematic analysis for the Global Burden of Disease Study 2017. *Lancet*. 2018;392(10159):1859-1922.
3. Hoy DG, Protani M, De R, Buchbinder R. The epidemiology of neck pain. *Best Pract Res Clin Rheumatol*. 2010;24 (6):783-792.
4. Borghouts JAJ, Koes BW, Vondeling H, Bouter LM. Cost-of-illness of neck pain in The Netherlands in 1996. *Pain*. 1999;80(3):629-636.
5. Vos CJ, Verhagen AP, Passchier J, Koes BW. Clinical course and prognostic factors in acute neck pain: an inception cohort study in general practice. *Pain Med*. 2008;9(5):572-580.
6. Swinkels ICS, Kooijman MK, Spreeuwenberg PM, et al. An overview of 5 years of patient self-referral for physical therapy in the Netherlands. *Phys Ther*. 2014;94(12):1785-1795.
7. Nivel. Cijfers Fysiotherapeuten. Available at: https://www.nivel.nl/zorgregistraties-eerste-lijn/fysiotherapeut. Accessed August 10, 2020.
8. Piscitelli D, Furmanek MP, Meroni R, De Caro W, Pellicciari L. Direct access in physical therapy: a systematic review. *Clin Ter*. 2018;169(5):e249-e260.
9. Groeneweg R, van Assen L, Kropman H, et al. Manual therapy compared with physical therapy in patients with non-specific neck pain: a randomized controlled trial. *Chiropr Man Therap*. 2017;25(1):12.
10. Kamper SJ, Grootjans SJM, Michaleff ZA, Maher CG, McAuley JH, Sterling M. Measuring pain intensity in patients

with neck pain: does it matter how you do it. *Pain Pract*. 2015;15(2):159-167.

11. Vernon H. The Neck Disability Index: state-of-the-art, 1991-2008. *J Manipulative Physiol Ther*. 2008;31(7):491-502.

12. Geri T, Piscitelli D, Meroni R, Bonetti F, Giovannico G, Traversi R, Testa M. Rasch analysis of the Neck Bournemouth Questionnaire to measure disability related to chronic neck pain. *J Rehabil Med*. 2015;47(9):836-843.

13. Cleland JA, Fritz JM, Childs JD. Psychometric properties of the Fear-Avoidance Beliefs Questionnaire and Tampa Scale of Kinesiophobia in patients with neck pain. *Am J Phys Med Rehabil*. 2008;87(2):109-117.

14. Bobos P, MacDermid JC, Walton DM, Gross A, Santaguida PL. Patient-reported outcome measures used for neck disorders: an overview of systematic reviews. *J Orthop Sports Phys Ther*. 2018;48(10):775-788.

15. Guyatt GH, Norman GR, Juniper EF, Griffith LE. A critical look at transition ratings. *J Clin Epidemiol*. 2002;55(9):900-908.

16. Kamper SJ, Maher CG, Herbert RD, Hancock MJ, Hush JM, Smeets RJ. How little pain and disability do patients with low back pain have to experience to feel that they have recovered? *Eur Spine J*. 2010;19(9):1495-1501.

17. Kamper SJ, Ostelo RWJG, Knol DL, Maher CG, de Vet HCW, Hancock MJ. Global Perceived Effect scales provided reliable assessments of health transition in people with musculoskeletal disorders, but ratings are strongly influenced by current status. *J Clin Epidemiol*. 2010;63(7):760-766.e1.