The Bournemouth questionnaire as an outcome measure in the rehabilitation of a person suffering with mechanical neck and arm pain and concurrent Charcot-Marie-Tooth disease: a case report

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Objective: The objective of this paper was to present a report in which the Bournemouth Questionnaire was used as an outcome measure in the treatment and rehabilitation of an individual previously diagnosed with Charcot-Marie-Tooth disease and concurrently suffering with mechanical neck and arm pain.

Design Architecture: This was a retrospective case report.

Sample profile: An individual over 18 years of age previously diagnosed with Charcot-Marie-Tooth disease and now suffering with mechanical neck and arm pain. The individual is free from other illnesses and the individual is free from contra-indications to chiropractic care and rehabilitation.

Outcome measures: The outcome measures of this case report were based on the Bournemouth Neck Questionnaire.

Method: The course of treatment involved diversified chiropractic manipulative therapy of involved joints, soft tissue therapy and specific rehabilitive postural exercises. Outcome measures were compared to pretreatment baselines. The Bournemouth Neck Questionnaire was completed at weeks 0, and 6. The length of the study was 6 weeks. Re-evaluation of treatment occurred at similar intervals.

Results: The Bournemouth patient raw scores preintervention were 6, 3, 2, 7, 2, 4, 2 with a total score of 26/70. Post intervention raw scores were 4, 2, 0, 7, 1, 2, 1 with a total score of 17/70. The percentage change score was 34.6. Objectif: L'objectif du présent article est de présenter un rapport dans lequel le questionnaire Bournemouth a été utilisé comme indicateur des résultats dans le traitement et le rétablissement d'une personne chez qui on avait précédemment diagnostiqué la maladie de Charcot-Marie-Tooth et qui souffrait alors de motricité du cou et de douleurs aux bras.

Architecture d'études : Il s'agissait d'un exposé rétrospectif de cas.

Profil de l'échantillon: Une personne âgée de plus de 18 ans chez qui on avait précédemment diagnostiqué la maladie de Charcot Marie-Tooth et qui souffrait maintenant de motricité du cou et de douleurs aux bras. La personne ne souffre pas d'aucune autre maladie et il ne lui est pas interdit de recevoir des soins d'un chiropraticien pour se rétablir.

Indicateurs des résultats : Les indicateurs des résultats de ce rapport sur certains cas ont été fondés sur les réponses au questionnaire Bournemouth Neck.

Méthode: La série de traitements a consisté en diverses manipulations chiropratiques des articulations impliquées, la thérapie des tissus mous et des exercices spécifiques de rétablissement de la posture. Les indicateurs de résultats ont été comparés aux niveaux de base du prétraitement. Le questionnaire Bournemouth Neck a été rempli aux semaines 0 et 6. La réévaluation du traitement s'est produite à des intervalles similaires. L'étude a duré six semaines. La réévaluation du traitement s'est faite au même rythme.

Résultats : Les résultats bruts du patient de Bournemouth avant l'intervention étaient de 6, 3, 2, 7, 2,

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Conclusion: The change in raw scores was not clinically important. The percentage change score is considered indicative of clinically important change. The following two questions for further study are presented. Can the questions relating to depression and mood in the Bournemouth Questionnaire be used as a clinical indicator of adherence to home exercise programs? Can the Bournemouth Questionnaire be used as an appropriate outcome measure of non-specific musculoskeletal pain in individuals suffering from Charcot-Marie-Tooth disease? (JCCA 2006; 50(3):190–194)

KEY WORDS: neckpain, Bournemouth, treatment, outcome measure.

Introduction

Charcot-Marie-Tooth (CMT) disease is the most common inherited peripheral neuropathy affecting 1 in 2500.1,2 The disease has been reclassified as a Hereditary Motor Sensory Neuropathy (HMSN) and there are seven main subtypes with further classification among these.³ By far the most common types are HMSN 1 and HMSN 2 which account for over 70% of cases.^{2,3} These disorders are characterized by slowly progressive neuropathic processes involving the distal muscles with a highly variable severity and onset.^{3,4} The two forms are clinically difficult to differentiate with initial presentation often being foot pain secondary to foot deformity.^{3,5} Lower extremity weakness, 'clumsiness' and foot drop or steppage gait are also common.³ Both categories of the disease result in muscle atrophy, weakness and deconditioning visible upon examination.^{1,3} The two categories are differentiated by electro diagnosis and have specific nerve conduction velocity (NCV) findings.³ HMSN 1 has markedly decreased NCVs whereas HMSN 2 has only mildly to moderately reduced NCVs.3 These and other electro diagnostic findings appear to indicate that HMSN 1 is the result of chronic progressive axonal degeneration and 4, 2 pour un résultat final de 26/70. Les résultats bruts après l'intervention était de 4, 2, 0, 7, 1, 2, 1 pour un total de 17/70. L'écart en pourcentage était de 34,6.

Conclusion: L'écart dans les résultats bruts n'est pas important au plan clinique. Le pourcentage de l'écart des résultats est considéré comme une indication de changements importants au plan clinique. Nous présentons deux prochaines questions à inclure dans des études à effectuer: Est-ce que les questions reliées à la dépression et à l'humeur dans le questionnaire Bournemouth peuvent être utilisées comme un indicateur clinique de l'adhésion à des programmes d'exercices à la maison? Peut-on utiliser le questionnaire Bournemouth comme un indicateur des résultats de douleurs musculosquelettiques non spécifiques chez les personnes souffrant de la maladie de Charcot-Marie-Tooth? (JACC 2006; 50(3):190–194)

MOTS CLÉS: douleur au cou, Bournemouth, traitement, indicateur de résultats.

reinnervation with segmental demyelination.³ HMSN 2 has electro diagnostic findings more consistent with axonal loss involving the anterior horn cells of the spinal cord without evidence of segmental demyelination.^{3,5} The diseases are ultimately differentiated by genetics,⁵ which is beyond the scope of this paper.

There is no cure for the HMSN diseases at present,⁵ and treatments are aimed at functional restoration of gait via orthoses and surgery,³ treatment of neuropathic pain via medications,⁶ and rehabilitation exercises.^{1,5,6}

The purpose of this paper is to present a case in which the Bournemouth Questionnaire is used as an outcome measure in a patient with previously diagnosis HMSN 2 who is managed with conservative treatment and rehabilitation exercises for a concurrent problem of mechanical neck and arm pain.

Methods and Materials

Design architecture

The proposed study was a retrospective case report. The report was interventional, with the subject acting as his or her own control. There was comparison of pre and post outcome measures.

Sample profile/case report

The subject was an adult having been previously diagnosed with Charcot-Marie-Tooth disease. Since the treatment plan involves manipulation of involved and related joints, the subject did not have contra-indications to manipulations and was in otherwise good health. Some possible contra-indications to therapy and rehab included vascular complications, tumors, bone infections, traumatic injuries, psychological considerations, metabolic disorders, neurological complications, and unstable joints.⁷

The individual, a 30-year-old male, presented to a private chiropractic clinic with a complaint of neck, upper back, shoulder and upper arm pain of many years duration. The pain was described as 'muscle pain, like you feel after working out' and was constant. Worsening factors included stress at work and damp or cold days. Relieving factors included infrequent exercise and self manipulation. The patient reported to have been diagnosed with HMSN 2 since age 19 and that his father also suffered from the same affliction. Due to his condition, the patient had gait difficulties and foot deformity but had not received orthotic support. Secondary problems were reported as cataracts in both eyes and some hearing loss in his left ear. The patient was currently taking Robaxacet for pain and creatine supplements. He reported to neither smoke nor drink and did not exercise. He reported to be otherwise healthy.

Physical exam revealed noticeable wasting in the intrinsic hand muscles, and in the platarflexors of the legs. Pes planus with lateral foot flaring and excessive medial talar shift was noted. The 'medial talar shift' was subjectively noted during non-weight and weight bearing and during gait. Anterior head carriage and low left shoulder and hip were also noted. The patient also showed positive testing for faulty breathing patterns, faulty shoulder abduction, and upper cross syndrome.8 Range of motion for the cervical spine was pain free with mild reductions in left rotation and left lateral flexion. Active forward flexion for the thoracic and lumbar spine was self limited due to pain in the hamstrings. Extension end range was painful in the thoracic spine. Joint compression tests were negative throughout the spine. Cervical Doorbell Tests produced pain and tingling to the fingers for C5-7 levels on the right. Scalene group palpation produced pain referral into the chest, shoulder and arm bilaterally. The upper trapezius muscle referred cephalad on the right. Roos test was positive for numbness and pain into the right shoulder and arm. Joint restrictions were palpated at C4–7 left, C2 right and T2–5. Neurological testing was normal except for 4/5 motor strength for C8/T1 levels on the right.

The patient was diagnosed with Chronic Mechanical Neck and Arm pain secondary to postural strain and concurrent CMT type 2.9–12 Passive therapy included spinal manipulative therapy and soft tissue therapy and active therapy consisted of exercises aimed at postural correction and strengthening of proximal spinal and scapular stabilizing muscles such as longis coli and capitis, and middle and lower trapezius, and serratus anterior.8

Outcome measures

The outcome measure in this case study was the subjective questionnaire know as the Bournemouth Neck Questionnaire (BQ neck).¹³ The BQ neck is a subjective survey of seven core scales recorded on a eleven point numerical rating scale.14 It focuses on neck pain from a biopsychosocial model rather than from a medical model. 13,14 This biopsychosocial model holds that musculoskeletal pain is a complex and multicomponent entity. 13,14 As such, outcome measures in musculoskeletal cases should be focused on patient's perceptions of pain, quality of life and disability rather than just pain severity.¹⁴ This focus can be better obtained through a questionnaire like the BQ neck, which does address the psychological and social aspects of neck pain.¹⁴ The BQ neck has been shown to be a comprehensive, valid, reliable outcome measure with practical ease for repeated use in clinical settings. 13-16

Protocol

Prior to treatment the subject completed the BQ neck questionnaire in order to establish baseline measurements. A chiropractor in private practice in Mississauga Ontario treated the subject for a six week period. The patient was seen for 15 visits over a period of six weeks. Passive treatment included manipulative therapy of the cervical and thoracic spine, ischemic compression of trigger points and cross frictional massage of the pectoralis major and minor, the scalenus group and the sub-occipital group of muscles. Active care included supine chin

Table 1	Raw Bournemouth	Neck	Questionn	iaire Scores
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	Score 1	Score 2	Score 3	Score 4	Score 5	Score 6	Score 7	Total Score
Pre-Test	6	3	2	7	2	4	2	26/70
Post-Test	4	2	0	7	1	2	1	17/70

Table 2 Percentage Change Score of Bournemouth Neck Questionnaire

Pre- Treatment Raw Score	Post-Treatment Raw Score	Percentage Change Score
26/70	17/70	34.6

tuck, wall angel, and push up plus, (first done standing, and then prone from the knees) exercises. The patient was instructed to do two sets per day with repetitions to tolerance or until exercise form was compromised.⁸ The rehabilitive exercises were given as a home program. Reevaluation of the treatments occurred at the end of 6 weeks. The BQ neck questionnaire was filled out by the patient at re-evaluation.

Data analysis strategy

The raw score and percentage change scores were calculated for the pre and post BQ neck questionnaires.

Results

The pre intervention raw patient's scores from the BQ neck were 6, 3, 2, 7, 2, 4, 2 with a total score of 26/70. The raw patient scores post intervention were 4, 2, 0, 7, 1, 2, 1 with a total score of 17/70. (See Table 1). The percentage change score was 34.6 and is shown in Table 2.

Discussion

There is some disagreement in the literature as to what scores on the BQ neck are indicative of important clinical change as a result of intervention. Bolton¹⁴ states that individual scores must change by three points, overall scores by 13 points and percentage change by 36% for the results of the BQ neck to be important. Hurst and Bolton¹⁶ have concluded that in neck pain raw change scores of 13 and percentage scores over 33% should be considered important. It is further stated that where there is a high variability in baseline values, the relationship between percentage change and clinical improvement is more consistent than that between raw score and clinical change.¹⁶ In this study we do have highly variable baseline scores. So while the raw score of nine is not consid-

ered important, the percentage change score of 34.6% would be considered indicative of clinically important change.

Although the BQ neck percentage change scores may be indicative of clinical change, the lack of a control in this report means that one can not conclude what was responsible for the clinical change. Interestingly, it was found upon the re-evaluation of the patient, that although the previously positive thoracic outlet tests were now negative, the faulty movement patterns were still occurring. The patient was questioned and asked to perform the prescribed exercises during the course of the treatment period, but, during the re-evaluation the patient confessed to 'not doing the exercises as often as he should.' It is therefore unknown to what extent the BQ neck scores would have changed had the patient been compliant with his active care.

Also of interest is the lack of change in the score relating to the psychological aspects of pain. This is the only score to show no change in the pre and post questionnaires. There is a well established connection between active therapy and depression and mood. 17 Recall that the patient admitted to not following the prescribed active therapy component to his care. Strength of a biopsychosocial questionnaire is that this information is not lost to the clinician. While undoubtedly the psychological aspects of pain are complex, perhaps this finding on the BQ neck could be a prompt to the clinician to investigate patient adherence to unsupervised active care.

There are numerous other limitations with a retrospective case study. Some of these include: selection bias, misclassification bias, blinding bias, and again because of a lack of a control, there is nothing to account for regression to the mean, natural history, or chance.¹⁸

Lastly, there is the issue of the outcome measure. Is the

BQ neck an appropriate measure in this case? The BQ neck is intended for use with nonspecific neck pain. Defention in their daily activities due to neuropathic pain. It is difficult to know the exact mechanism of pain in CMT patients, but it is thought that it is multifactoral. In addition to neuropathic pain CMT patients are often deconditioned 19,20 and therefore, the musculoskeletal system is also a likely pain generator. As such, could the BQ neck be considered an appropriate outcome measure for the nonspecific pain due to the musculoskeletal system in patients with CMT? It is therefore proposed that future investigations compare the BQ with a neuropathic pain measurement tool such as the Neuropathic Pain Scale, in patients with CMT.

Conclusion

This retrospective case study describes the treatment of a patient with CMT 2 and mechanical neck and arm pain. The patient was treated with spinal manipulation, soft tissue therapy, and postural re-education and cervical and scapular stability exercises. The outcome measure was the BQ neck taken at pre and post treatment intervals. Raw scores were not indicitive for clinically important change, whereas the percentage change score was indicative of clinically inportant change. Problems in patient compliance with active care were noted. It is proposed that the psychologically focused questions of the BQ neck could be used as indicators to investigate active care compliance, and furthermore, that the neuropathic and musculoskeletal sources of pain in CMT patients be examined in future studies by comparing the BQ neck with specific outcome measure questionnaires for neuropathic pain.

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