Journal of the Neurological Sciences 341 (2014) 51–54

Contents lists available at ScienceDirect

Journal of the Neurological Sciences

journal homepage: www.elsevier.com/locate/jns

Hand dexterity and direct disease related cost in multiple sclerosis

Marcus W. Koch [a,b,c,*], T. Jock Murray [d], John Fisk [d], Jamie Greenfield [a,b], Virender Bhan [d], Philip Jacobs [e], Murray Brown [f], Luanne M. Metz [a,b]

[a] Department of Clinical Neurosciences, University of Calgary, Calgary, Alberta, Canada
[b] Hotchkiss Brain Institute, University of Calgary, Calgary, Alberta, Canada
[c] Department of Community Health Sciences, University of Calgary, Calgary, Alberta, Canada
[d] Department of Medicine, Dalhousie University, Halifax, Nova Scotia, Canada
[e] Department of Medicine, University of Alberta, Edmonton, Alberta, Canada
[f] Department of Community Health and Epidemiology, Dalhousie University, Halifax, Nova Scotia, Canada

ARTICLE INFO

Article history:
Received 17 January 2014
Received in revised form 7 March 2014
Accepted 25 March 2014
Available online 2 April 2014

Keywords:
Multiple sclerosis
Outcome measures
Hand dexterity
Nine hole peg test
Health economics
Disease related cost

ABSTRACT

Methods: The nine hole peg test (9HPT) is an emerging outcome measure in clinical trials in multiple sclerosis (MS). In this study we investigated how performance on the 9HPT at baseline is related to annualized direct MS related cost.
Methods: We enrolled patients with a definite diagnosis of MS from two Canadian MS centers. 9HPT and demographic information were recorded at baseline, and patients prospectively recorded all MS related costs for 6 months. Costs were compared among five groups according to the baseline 9HPT, and we built a multiple linear regression model including cost (dependent variable) and 9HPT at baseline, age, disease duration, sex and disease course (independent predictor variables).
Results: We analyzed data from 298 patients. Cost significantly increased with increasing 9HPT scores ($p < 0.0001$), with the costs for health care providers, changes to the home or car and long-term care dominating in the most disabled patient groups. The 9HPT score was a significant predictor of cost in the regression model ($p = 0.006$).
Conclusion: Performance on the 9HPT is closely related to cost. Our data add another aspect of patient relevance to using the 9HPT as an outcome measure in clinical trials.

1. Introduction

Multiple sclerosis (MS) is a chronic disease of the central nervous system that usually causes some degree of disability during its course. MS affects all areas of neurological function, from mobility and dexterity to cognition and mood. Because of the wide variety of functional impairments that MS can cause, and the fact that the degree and speed of progression of functional impairment differ from patient to patient, the measurement of disability in MS is an ongoing challenge, and continues to be an active area of research.

Disability in MS is usually measured with the Expanded Disability Status Scale (EDSS) [1], which was developed as a composite scale that measures a variety of important areas of function, such as cerebellar, sensory, motor and cerebral function. The EDSS is a very useful long-term outcome measure, for example in natural history studies of MS, but its use as an outcome measure in short term studies such as clinical trials has been criticized for its emphasis on ambulation and for its poor inter-rater reliability [2,3]. Moreover, the fact that the EDSS is a categorical scale rather than a continuous measure makes it difficult to compare change from different baseline levels. A study using original trial data from several large trials has questioned its validity as an outcome measure for unremitting disability [4]. Thus, especially for trials in progressive forms of MS there is a wish for alternative outcome measures [5,6].

The Multiple Sclerosis Functional Composite (MSFC) is an alternative to the EDSS that aims to be a comprehensive measure of disability. The MSFC consists of three continuous measures that examine ambulation, hand dexterity and cognitive function, each of which is weighted equally and combined into an overall summary score. While improving on the EDSS with regard to intra-rater and inter-rater reliability, a disadvantage of the MSFC is the difficulty for clinicians to conceptualize the clinical meaning of its summary score which has no intuitive clinical correlate. Hand dexterity in the MSFC is measured with the nine hole peg test (9HPT) which records the time it takes a subject to insert and then remove nine pegs from a board. While the 9HPT is a component of the MSFC, it is also a useful outcome in its own right. The advantages

* Corresponding author at: Department of Clinical Neurosciences, Room 178 HMRB, 3330 Hospital Drive NW, T2N 4N1, Calgary, Alberta, Canada. Tel.: +1 403 944 2509.
E-mail address: mwkoch@ucalgary.ca (M.W. Koch).

http://dx.doi.org/10.1016/j.jns.2014.03.047
0022-510X/

of the 9HPT are its quantitative nature, the fact that it is quick and easy to perform, that it has very high inter- and intra-rater reliability [7] and that it is easy for a clinician to interpret the score. In addition, changes in the 9HPT have been found to be related to patient rated daily life disability, which suggests that the 9HPT is a patient-relevant outcome [8]. Another advantage is the fact that the 9HPT can be used in patients who are no longer ambulatory but yet would benefit from therapy that slowed their further deterioration. This group of severely disabled patients is currently excluded from most clinical trials, even though they could benefit from new treatments that target the progressive phase of MS [6].

It is well established that MS is a disease with a large socio-economic burden [9,10], but little is known about how individual disability measures such as the 9HPT are related to MS related costs. Direct disease related costs are a consequence of MS for patients and their families and the relevance of an outcome measure such as the 9HPT can in part be judged by its relationship with cost. In this study we investigate how 9HPT scores are related to the prospectively collected annualized direct MS related costs in a cohort drawn from two Canadian MS clinics.

2. Methods

2.1. Study participants and data collection

These data were obtained from a larger prospective cohort study designed to evaluate the economic and non-economic burden of MS. It was approved by the University of Calgary and the Capital District Health Authority Research Ethics Boards. Study participants were recruited at two Canadian MS clinics in Calgary, Alberta and Halifax, Nova Scotia between 1996 and 1998. This predated the widespread availability of MS disease modifying therapy, and costs for such treatments in the few patients that were using them at the time (n = 11) were excluded from the analysis. A subgroup of this cohort has been described in a previous publication [11]. All participants had definite MS according to the Poser Criteria [12]. Recruitment was stratified into four groups based on EDSS (EDSS 0–2.5, EDSS 3.0–5.5, EDSS 6.0–8.0 and EDSS 8.5–9.5). The aim was to recruit 50 patients for each group from each clinic but recruitment of more disabled patients proved challenging, especially in Nova Scotia which faced greater travel issues for disabled patients due to a smaller urban population to draw from. Informed consent was received from all participants or their proxies. Each patient performed the 9HPT at baseline in both hands. For statistical analyses the average value of both hands was used.

Patients recorded MS related direct costs in detailed cost diaries for 6 months. This period was chosen to limit the burden on patients, some of whom were moderately or severely disabled, and to maximize compliance. This prospective data collection included reported costs for the purchase of goods and services, as well as health care utilization data. In addition to the cost diaries, public sector costs associated with procedures and/or surgeries, major diagnostic tests, hospital admissions and extended care or respite admissions relevant to MS were collected by obtaining utilization data from government insurers.

Patients' diaries included information on all MS related costs and were summarized as 'medical costs' (visits to medical caregivers such as doctors, nurses, physiotherapists, and occupational therapists, surgical procedures, diagnostic tests, hospitalizations), 'medications' (all prescription medications), 'vitamins and supplements' (all vitamins, supplements, homeopathic or naturopathic remedies), 'transportation' (all private and public transportation, taxi costs, car rental or ambulance transportation), 'equipment and care products' (all necessary equipment such as canes, walkers, and wheelchairs, and care products such as disposable catheters), 'home care', 'long term and respite care', and 'changes to home or car' (all changes made to the home or car due to MS). Current retail prices (at the time of recording) were used for prescription costs. The costs for doctor's visits, diagnostic procedures and medical interventions were taken from provincial reimbursement schedules. Costs for nursing, home care and long-term care were collected as service type and units of service and assigned costs according to the regional fee schedules. The costs recorded by the patients were used for all other costs. All of these costs were annualized based on the time the diary was kept. Statistical significance was taken to be at the two-tailed 0.05 level. All statistical analyses were performed with the R statistical software package version 3.0.1 for Windows [13].

3. Results

While 319 patients were enrolled, cost diary information was not available in 21 patients so they were excluded from this analysis. Of the 298 participants included, 223 were women and 75 were men; 116 patients had a relapsing–remitting disease course, 88 had secondary progressive MS, 92 had primary progressive MS, and disease course was uncertain in 2 patients. The 9HPT scores ranged from 13.5 to 216.5 s. Two hundred seventy three (92%) of the 298 patients kept their cost diaries for the full 6 months, 6 patients kept their diaries for 5 months, 2 patients each kept their diaries for 4, 3 and 2 months, and 13 patients kept their diary for 1 month. The annualized cost of MS ranged from 0 to 136,655 Canadian Dollars (CAN$). Baseline 9HPT could be measured in both hands in 224 patients. By definition patients with very high disability as measured by the EDSS (EDSS > 8.0) had such limited hand function that they could not perform the task. Thus, the description of annual direct MS related costs included 298 patients but the multiple linear regression model included 224 patients because a 9HPT score was required. Participant characteristics are shown in Table 1.

Patients were arbitrarily divided into five groups by their baseline 9HPT performance: 'up to 20 s' (n = 44), 'more than 20 to 30 s' (n = 111), 'more than 30 to 40 s' (n = 46), 'more than 40 s' (n = 23), and 'no test performed' (n = 74). There was a significant difference in the annualized cost of MS between these groups (Kruskal–Wallis $p < 0.0001$). The distribution of MS related costs to different cost categories is shown in Table 2 and Fig. 1. The costs in all categories increase with increasing disability. The costs in the most disabled groups are dominated by costs associated with modifications made to the home or car, home care and long-term care. Long term care first becomes an important cost factor in the group of patients who could not perform the 9HPT.

We built a multiple linear regression model with annualized direct MS related costs as the dependent variable and 9HPT scores, age, disease duration, sex, and disease course as independent predictor variables.

Table 1
Characteristics of the patient cohort.

	Overall cohort who completed cost diaries	Patients included in the regression model (completed the 9HPT with both hands)
N	298	224
Sex: f/m (%)	223/75 (75%/25%)	170/54 (76%/24%)
Disease course: RRMS/SPMS/PPMS (%)	116/88/92 (39%/30%/41%)[a]	112/58/54 (50%/26%/24%)
Age: median (IQR)	46.5 (41–56)	46 (40–53)
Disease duration: median (IQR)	17 (9–23)[a]	14 (8–21)
9HPT [seconds]: median (IQR)		25 (21–32)

IQR: interquartile range.
[a] Unavailable in 2 patients.