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EXPECTATIONS OF WHEELCHAIR DEPENDENCE IN RECENTLY DIAGNOSED PATIENTS WITH MULTIPLE SCLEROSIS AND THEIR PARTNERS

Abstract

The aim of the present paper was to quantify expectations of wheelchair dependence in patients recently diagnosed with MS (n=101) and their partners (n=78). Expectations focused on the risk and seriousness of becoming wheelchair-dependent in two years, ten years or lifetime. Expectations were compared with natural history data, compared between patients and their partners, and related to clinical characteristics. Our results show that patients overestimated their 2-year and 10-year risks of wheelchair dependence, but underestimated their lifetime risks. A large number of patients were uncertain about their 2-year risk, even those with no or only minimal disability (EDSS < 3.0). One-third of the patients perceived the 10-year and lifetime risk to be 50%, which, as they explained in the interviews, reflected their uncertainty: they did not know what to expect – it might happen or it might not. Patients with more functional limitations had higher perceptions of risk, but lower perceptions of seriousness. Concordance in perceived risk and seriousness between patients and partners was moderate. The overestimation of the short-term risks and the substantial differences in expectations within couples warrant further research on the impact of expectations on their treatment decisions and psychological well-being.

Introduction

Patients with multiple sclerosis (MS) face enormous prognostic uncertainty as the disease is characterized by an unpredictable variation in symptoms, severity and progression.^[1] While aware of the serious consequences of their disease, patients cannot be informed about their personal prognosis. This uncertainty may disrupt their psychological well-being,^[2-4] and may particularly be a problem for newly diagnosed patients.^[5] MS affects young individuals between 20 and 40 years of age. At these ages, people are making decisions about relationships, family planning and career that have important implications for their lives. As these decisions cannot be based on clinical prognostic information, patients' expectations of disease progression will play a major role.

Expectations of the risk of future health states have frequently been studied in other medical areas, such as prenatal testing,^[6] presymptomatic genetic testing,^[7,8] surgery,^[9] and preventive medicine.^[10,11] However, studies on expectations of prognosis, or perception of *prognostic* risk, are still scarce. We know of one study that described perceived risk of major consequences of type 1 diabetes mellitus. This study showed that patients considerably overestimated their 20-year risks of end state renal disease, amputation and blindness.^[12] In MS, patients' expectations have only been studied with regard to the benefits of treatment. This study demonstrated that patients who had unrealistic expectations about the possible improvement in functional status showed poorer adherence to treatment.^[13]

The aim of our study was to quantify expectations of prognosis in patients recently diagnosed with MS and their partners. We focused on the risk of wheelchair dependence, as this is one of the major and most recognized consequences of the disease by patients. We intended to answer the following questions: (1) Do recently diagnosed MS patients expect to become wheelchair-dependent in the short, medium or long term? (2) How serious do they consider it is to become wheelchair-dependent? (3) Do expectations differ between patients and their partners? (4) Are expectations about the risk and seriousness of wheelchair dependence related to clinical characteristics?

Methods

Participants and procedures

Patients were recruited through the Departments of Neurology of the Erasmus MC (Rotterdam), three hospitals within the region of this academic hospital, and the VU Medical Center (Amsterdam) in the period of March 1999 – December 2002. Patients were eligible if they were diagnosed with definite or probable MS^[14] within two years before entry in the study, were between 18 and 55 years old, and

had given informed consent. Patients with serious comorbidity of other neurological or systemic diseases, or with insufficient understanding of the Dutch language were excluded. Of the 120 patients who met the criteria, 101 agreed to participate. Patients who declined participation mentioned the emotional burden (n=3) or a lack of interest (n=3). Nine others declined without additional comments and four never responded to our reminders. Ninety out of 101 patients had a partner, of whom 78 (87%) did participate. Two partners did not speak Dutch, six were not living with the patient and for that reason not invited by the patient, and four refused for unknown reasons.

Patients underwent a neurological examination, filled out questionnaires and were interviewed. The questionnaires were sent one week before the neurological examination and had to be completed before the interview, which was scheduled one week after the examination. Interviews were carried out by the first author at the patient's home. Partners received their questionnaires before the patient's interview and were asked to complete these in another room. This procedure was chosen to control separate completion of the questionnaires by patients and partners in order to assess their expectations independently. The study protocol was approved by the medical ethical committees of the participating hospitals.

Measurements

Expectations. We assessed patients' expectations with regard to the risk and seriousness of becoming wheelchair-dependent as a consequence of the disease.^[15] Wheelchair dependence was defined as the inability to walk beyond five meters. This definition equals a score of 7.0 on the Expanded Disability Status Scale (EDSS).^[16] The risk of wheelchair dependence was measured for the short (two years), medium (ten years) and long term (lifetime). Perception of risk was assessed using a 100mm visual analogue scale (VAS) which ends were anchored at 'Definitely not' (0%) and 'Definitely' (100%). The VAS is a widely-used instrument for the quantification of subjective phenomena including attitudes, pain, discomfort and perception of risk.^[17,18] Actual risks were derived from epidemiological data and estimated to be 5-10% for the 2-year risk, 20-25% for the 10-year risk and 70-80% for the lifetime risk.^[19] In a population of 800 patients the 10-year risk was calculated 24% (G.C. Ebers, MD, PhD, unpublished data, 2001), which confirmed our estimation. Further, patients were asked for each time period how serious they think it is to be wheelchair-dependent by that time. Answers were given on a VAS anchored at 'Not serious at all' (0) and 'The most serious thing I can imagine' (100). Partners completed the same questions. In the interview, patients were asked to explain their perception of the 10-year risk. These

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explanations were transcribed verbatim and independently evaluated by two psychologists (CJ and JdB). Similar explanations were grouped together in categories, and categories were labeled. Differences between the categorizations of the two psychologists were discussed with a third (JP), until a final categorization was reached by consensus. To determine differences in the use of specific explanations between groups, we created a variable for each category. This variable was coded 1 when an explanation of the patient was assigned to the category, and 0 if not. Patients could give multiple explanations. Interview data were available for 80 out of 101 patients.

Clinical data. The neurological examinations were assessed by physicians from the academic hospitals following a standardized research protocol. Functional limitations were rated on the EDSS,^[16] which ranges from 0.0 (no neurological symptoms) to 10.0 (death due to MS). Date of first symptoms was assessed during the neurological examination. Date of diagnosis and diagnostic certainty (probable or definite MS) were obtained from the medical records.

Statistical analysis

Differences in means of perceived risk and seriousness between patients and partners were analyzed using the paired t-test. To determine the degree of concordance in perception of risk and seriousness *within* pairs, we calculated the absolute differences between patient and partner scores. Perceptions of patients and partners were considered concordant when the absolute difference between the VAS scores was lower than 20, partially discordant when the difference was between 20 and 39, and fully discordant when the difference was 40 or higher. To determine the relationship with clinical characteristics, we calculated Spearman's rank correlations between perceived risk and seriousness, time since diagnosis, time since first symptoms and EDSS. The relationship of perceived risk and seriousness with EDSS was further investigated by comparing two groups and tested using the independent samples t-test. These groups were defined by the patient's EDSS score, combining no to minimal disability (EDSS < 3.0) and moderate to severe disability (EDSS ≥ 3.0).^[16] Frequencies of coded explanations of the perceived 10-year risk were compared between subgroups using Fisher's exact test.

Results

Description of subjects

Mean age of the patients was 37.5 years (SD 9.5) and 70% were women. Ninety percent was diagnosed with definite and 10% with probable MS. Mean time since

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diagnosis was 7.8 months (SD 6.5) and mean time since first symptoms 3.7 years (SD 4.6; median 2.1). EDSS scores ranged from 0.0 to 7.0. Median EDSS score was 2.5. Mean age of the partners was 39.3 years (SD 8.7) and 64% were men.

Perceived risk and seriousness of wheelchair dependence of patients

Mean perception of the 2-year risk of wheelchair dependence was 22.5% (SD 21.2), of the 10-year risk 38.7% (SD 25.5) and of the lifetime risk 54.0% (SD 24.6). These means were higher than the actual 2-year (actual risk 5-10%) and 10-year risks (actual 20-25%) and lower than the actual lifetime risk (actual 70-80%). The distributions of the perceived 2-year, 10-year and lifetime risk of wheelchair dependence are presented in Figure 1. Figure 1a shows that the majority of patients (82%) thought the risk to become wheelchair-dependent within two years was lower than 50% (VAS-scores 0-45). Twenty-eight percent thought it highly unlikely that they would need a wheelchair within two years (VAS 0-5), while 54% was less certain about this (VAS 6-45). Figure 1b shows that half of the patients (48%) thought the risk to become wheelchair-dependent within a period of ten years less than 50%. One-third of the patients perceived the 10-year risk to be about 50% (VAS 46-55). This group stands out as a major isolated peak. Similar results were found for the perceived lifetime risk (Figure 1c), albeit that a smaller group (28%) thought the lifetime risk to be lower than 50% (VAS 0-45).

As was shown in Figure 1, one third of the patients perceived their 10-year or lifetime risk to be about 50% (VAS 46-55). When examining the interviews, these

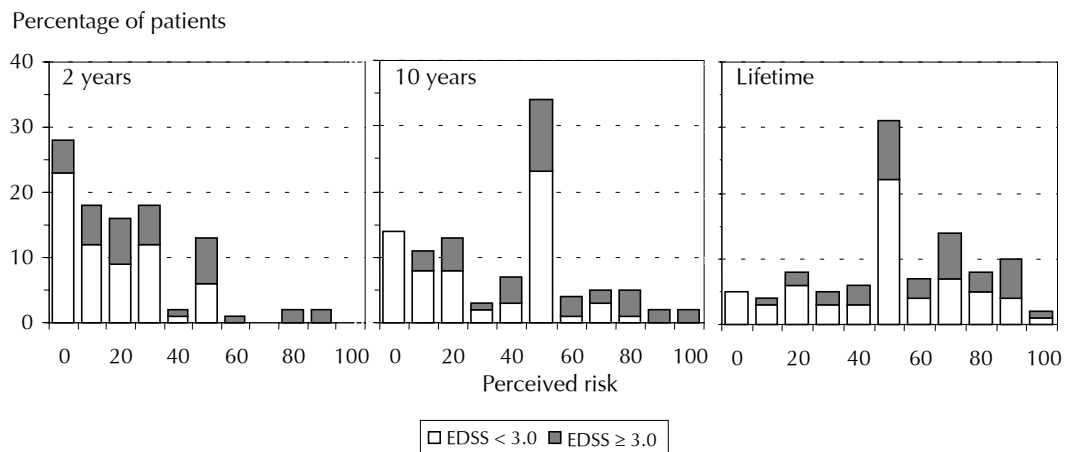


Figure 1 Perceived risk of wheelchair dependence of patients

Perceptions of risk (mm VAS) were recoded into 11 categories: 0-5 into 0, 6-15 into 10, and so on.

Table 1 Common explanations on perceived 10-year risk of 50%-responders in comparison with other patients

Explanation on perceived risk	Perceived 10-year risk		Relative frequency (95% CI)	p Value
	50% (n=30)	Other (n=50)		
"I don't know"	23 (77%)	10 (20%)	3.8 (2.1-6.9)	<0.001
"It might happen or not"	18 (60%)	1 (2%)	30.0 (4.2-213.4)	<0.001
Physical condition	10 (33%)	17 (34%)	1.0 (0.5-1.9)	0.95

Frequencies were compared between patients who gave a 50%-response and those who did not. A 50%-response was defined as a VAS-score between 46-55, whereas 'other' included patients with VAS-scores lower than 46 or higher than 55. Values indicate the number (%) of patients that mentioned the explanation in the interview. Multiple explanations were allowed. * Fisher's exact.

50%-responders significantly more often stressed their uncertainty. With regard to the 10-year risk (Table 1), explanations like "I don't know" (3.8 fold increase compared to others, $p < 0.001$) and "It might happen or might not happen" (30 fold increase compared to others, $p < 0.001$) were significantly more often used by 50%-responders. Peaks at 50% were found among patients with high and low EDSS and among partners (data not shown). Means of perceived risk were still higher than actual risks when 50%-responses (VAS 46-55) were excluded.

Mean perception of seriousness was slightly higher for the 2-year risk of wheelchair dependence (82.5, SD 19.7) than for the 10-year (74.5, SD 22.9) and lifetime risk (71.6, SD 24.0). These findings indicate that patients considered wheelchair dependence a serious consequence of their disease, particularly when it would happen within two years.

Differences between patients and partners

Mean perceptions of risk of partners did not significantly differ from that of patients ($p > 0.05$). Partners considered wheelchair dependence to be less serious than patients, but within-pair differences were significant only for the 2-year period (mean 73.6% versus 82.5%; $p=0.05$). Although differences in means were not significant, inspection of the data did show substantial differences within pairs at all time points: that is, patients had higher perceptions than partners in some couples, but lower in others. Analyses of absolute differences demonstrated that in approximately half of couples the differences in perceptions of risk and seriousness of patients and partners were less than 20 points on the VAS scales (Table 2). Fully discordant perceptions of risk (differences in VAS ≥ 40) were found in 14% of the couples and fully discordant perceptions of seriousness in 21%.

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Table 2 Differences in perceived risk and seriousness of wheelchair dependence between patients and partners (n = 77 couples)

	Concordance (%)	Discordance (%)	
		Partial	Full
Difference in VAS scores:	0 - 19	20-39	≥ 40
Perceived absolute risk			
2-year	55	31	14
10-year	59	27	14
Lifetime	57	27	16
Perceived seriousness			
2-year	57	22	21
10-year	49	31	20
Lifetime	54	25	21

Correlation with clinical characteristics

Patients with more functional limitations perceived themselves to be at higher risk of wheelchair dependence (Figure 2). The relation between perceived risk and EDSS was stronger for the shorter periods than for the lifetime risk: the correlation of EDSS with perceived 2-year risk was 0.48 ($p < 0.001$), perceived 10-year risk 0.50 ($p < 0.001$) and perceived lifetime risk 0.26 ($p = 0.01$). Perceptions of risk were also significantly higher among partners of patients with more functional limitations compared to partners of patients with EDSS < 3.0 ($p < 0.04$; Figure 2). Perceived seriousness of wheelchair dependence was inversely related to EDSS only in patients: patients with *more* functional limitations evaluated wheelchair dependence to be less serious ($p < 0.01$). No relation between perceived seriousness and EDSS was seen in partners. Neither in patients nor partners, perceptions of risk and seriousness were related to sex, age, diagnostic certainty (definite versus probable MS), time since diagnosis and time since first symptoms (data not shown).

Discussion

Our study demonstrated that patients recently diagnosed with MS overestimated their 2-year and 10-year risks of wheelchair dependence, and underestimated their lifetime risks. A large number of patients were uncertain about their 2-year risk, even those with no or minimal disability. This uncertainty was even more predominant in medium- and long-term expectations. Patients with more functional limitations had higher perceptions of risk, but lower perceptions of

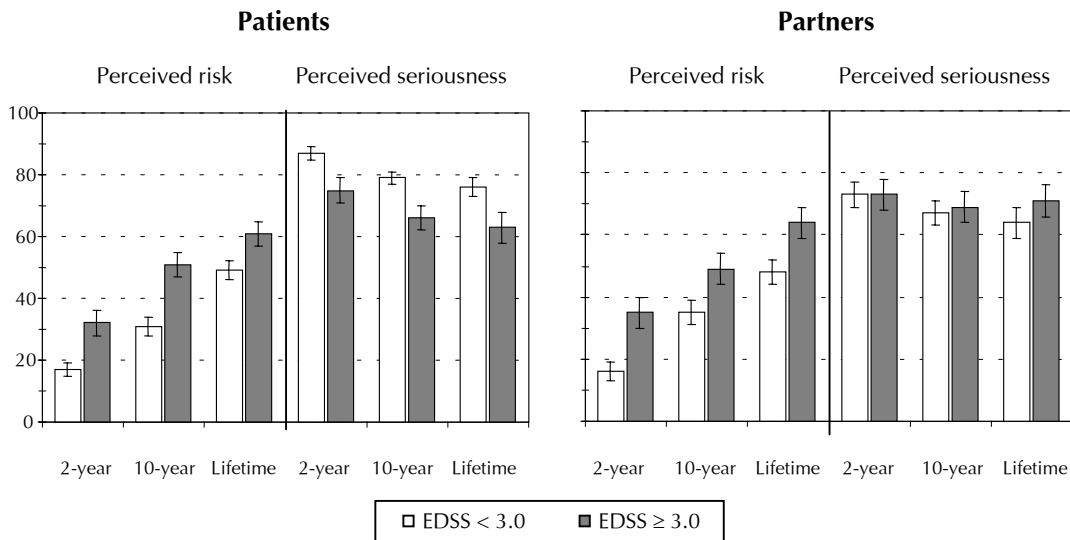


Figure 2 Means (SE) of perceived risk and seriousness of wheelchair dependence by EDSS

seriousness. Concordance in perceived risk and seriousness between patients and partners was moderate.

Before interpreting the data from a clinical perspective, three issues remain to be elucidated. First, we found that perceived risks were not related to clinical parameters such as time since diagnosis, time since onset of symptoms and diagnostic certainty (definite or probable MS). A point of consideration is that all patients were diagnosed within two years prior to inclusion. This means that there was limited variation in the time since diagnosis and time since first symptoms of our patients. Further, we intended to include patients with definite and probable MS to investigate the impact of diagnostic certainty, but could include only ten patients with probable MS. This small number implies that the findings of this study are primarily generalizable to patients recently diagnosed with definite MS.

Second, for studying differences between patients and partners, it is important that they completed their questionnaires separately. We arranged this by asking partners to fill in their questionnaires while the patients were interviewed. Nevertheless, we cannot completely rule out that partners had discussed and retained the patients' results prior to completion of their own questionnaires. If partner responses had been biased, differences in expectations between patients and partners were more likely underestimated rather than overestimated.

Third, up to one-third of the participants perceived their 10-year and lifetime risk about 50%. Our interview data confirmed that these 50%-responses primarily

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reflected patients' uncertainty about prognosis, rather than their belief that the risk was about 50%.^[20] The question that remains to be answered is *why* uncertainty elicits 50%-responses. First of all, participants who have absolutely no idea what to expect may perceive the risk of wheelchair dependence to be equal to the risk of not becoming wheelchair-dependent, as explained in the interview as 'it might happen or it might not'. Second, 50% is a neutral response that will be justifiable whatever happens.^[21] Patients who do not expect to become wheelchair-dependent may prefer this neutral answer to prevent future disappointment, whereas for those who do expect to become wheelchair-dependent, it is the most optimistic answer that is not yet unrealistic. Finally, the 50%-response may be an escape for whom thinking about consequences of MS is too threatening. From a clinical point of view, it may be relevant to distinguish between these underlying motivations, because they are informative on how patients deal with uncertain future consequences.

Patients with more functional limitations evaluated their risks of wheelchair dependence to be higher than did patients with fewer limitations, while at the same time, they considered wheelchair dependence as less serious. The first results suggest that patients extrapolate present functional limitations into expectations of future disease progression. This may be justified for expectations of short-term disease progression, but less so for the long term. The unpredictable and uncertain course of MS implies that this early period after diagnosis is not representative for the long-term disease course. Second, the fact that patients with higher disability evaluated wheelchair dependence to be less serious may be explained by the fact that differences in disability status alter the need and significance of a wheelchair, and hence change the criteria to evaluate its seriousness.^[22,23] Patients who are limited in walking may value the need of a wheelchair as an increase of mobility, whereas others who are fully ambulant view wheelchair dependence as a major step backwards. This shift in criteria is characteristic for accommodative coping processes, which enable patients to maintain positive outlooks in situations of increasing functional limitations.^[24,25]

Comparison of mean perceptions of risk and seriousness revealed no significant differences between patients and partners. However, we found considerable discordance in perceptions within couples, which implies that patients had higher perceptions than their partners in some couples, but lower in others. Previous studies have also demonstrated poor agreement between spouses in the judgment of quality of life of patients with metastatic cancer^[26] and in the assessment of behavioral risk factors of healthy workers.^[27] Further research is needed to investigate the consequences of discordant expectations for the

relationship between patients and partners focusing on their communication, mutual support, future plans and well-being.

Natural history data showed that the 'actual' 2-year risk of EDSS 7.0 after diagnosis is approximately 5-10%.^[19] In our study, this was overestimated by patients (mean VAS score 23%) and their partners (mean 25%). They also overestimated the 10-year risk ('actual' risk 20-25%), but underestimated the lifetime risk ('actual' risk 70-80%) as compared with natural history data.^[19] These biases in perceptions were found in patients with low EDSS (< 3.0) and high EDSS (\geq 3.0). These findings raise the question whether and how their perceptions of prognostic risks can be improved in a clinical setting. For this purpose, it is important to know what factors influence perception of risk. The present study showed that perception of risk was related to disability status, suggesting that patients' expectations were determined by their experience of symptoms. This means that for patients it is important to learn the difference between common symptoms and MS-related symptoms, which may not be easy in the early phase. Another factor that will determine patients' expectations is the information they receive about MS. Particularly in uncertain and unpredictable diseases as MS, patients may rely on the information that is provided by neurologists and other health professionals. A point of consideration is that information is also increasingly obtained from the Internet. Notwithstanding that this is a valuable source of facts and figures about the disease, patients may not be able to judge what certain information means for their particular situation. To help in this interpretation, it is important that neurologists discuss expectations of future disease progression, treatment benefits and other MS-related issues in the clinical consultation with patients and partners.

To our knowledge, this is the first paper on expectations of prognosis in patients with MS and their partners. Overall, patients overestimated their short-term risks of wheelchair dependence and underestimated the long-term risk, even those with low EDSS. Further research is needed to understand the impact of uncertainty and unfavorable perceptions of prognostic risk on psychological well-being, and the consequences of discordant expectations for the relationship between patients and their partners.

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