Patient Experience Journal

Volume 9 | Issue 1 Article 20

2022

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Recommended Citation

Reitzel SB, Magyari M, Skovgaard L, Kristiansen M. Patient involvement in the development of PROMs within the MS Field: A systematic review. *Patient Experience Journal*. 2022; 9(1):169-173. doi: 10.35680/2372-0247.1662.

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PXJ Patient Experience Journal
Volume 9, Issue 1 – 2022, pp. 169-173

Research

Patient involvement in the development of PROMs within the MS Field: A systematic review

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Abstract
This review reports on the development of patient-reported outcome measures (PROMs), published within the past ten years, for people with multiple sclerosis (PwMS). Moreover, this review evaluates the extent to which patient participation was integrated in the development of the PROMs. A systematic review was conducted, and four relevant articles were extracted, from which nine PROMs were identified. Patient involvement in the development phase was identified in three PROMs. The results emphasize the need for more patient involvement in the development of new MS-specific PROMs to ensure that the measures reflect the needs and priorities of PwMS.

Keywords
Systematic review, patient-reported outcome, patient involvement, Multiple Sclerosis

Introduction

Multiple Sclerosis (MS) is an autoimmune, neurodegenerative chronic disease of the central nervous system.[1] People with MS (PwMS) develop perivenular inflammatory lesions that cause demyelinating plaque. The symptomatology of MS results from a combination of the location and size of these lesions.[1] MS is a complex disease with a fluctuating symptom burden, and each patient has a unique disease course. Consequently, people with MS (PwMS) experience a range of symptoms that vary in severity, affect their quality of life and cause significant disability over time.[2]

The treatment of MS entails a combination of disease-modifying and symptomatic treatment options that are sometimes supplemented with complementary and alternative treatments and exercise and/or rehabilitation.[3]

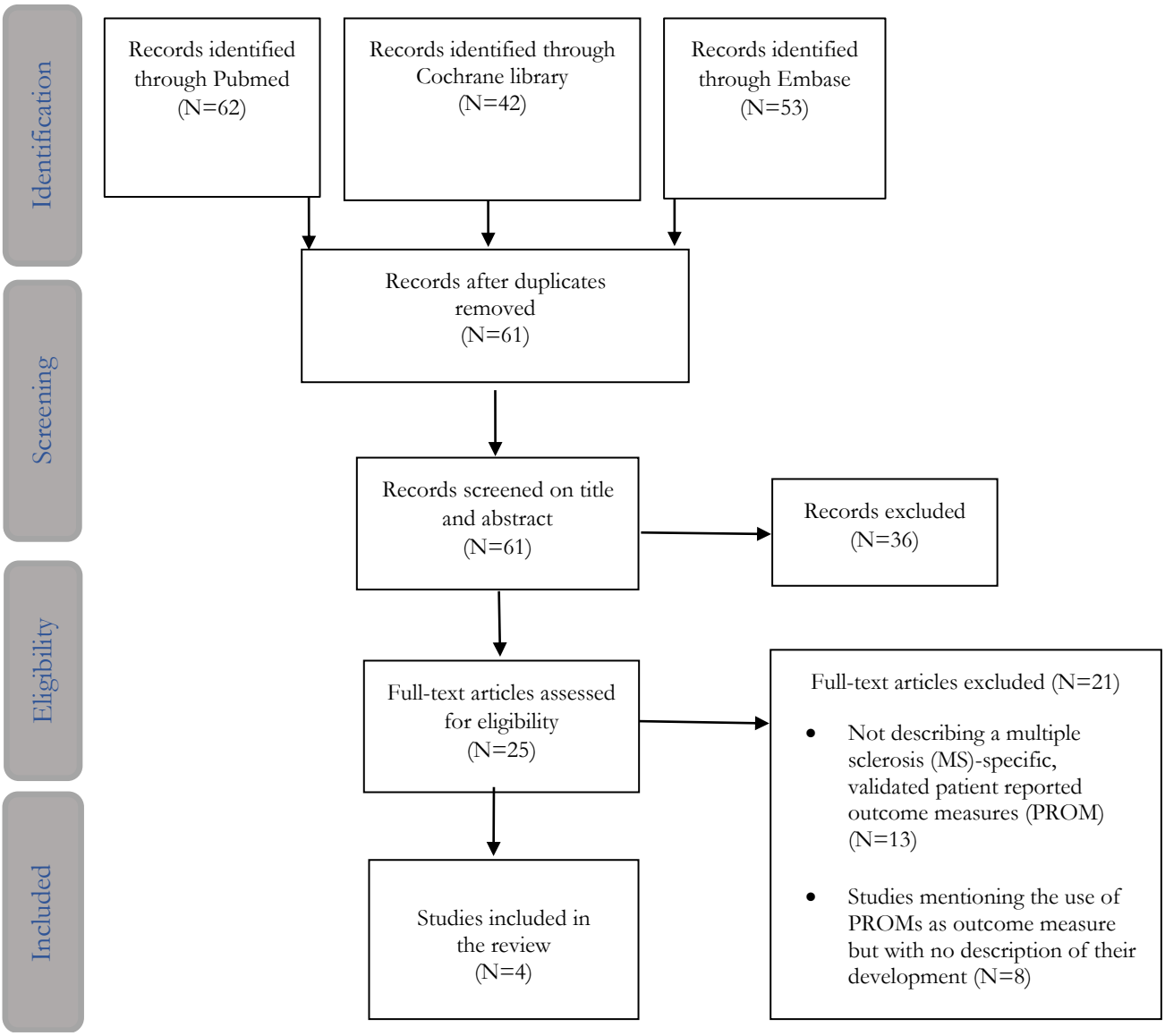
Patient-reported outcome measures (PROMs) measure patients' assessment of their state of health, symptoms, health-related quality of life and functional level.[4] The perspectives of patients obtained through PROMs are considered to be valuable for more comprehensive assessments of health status and the ability to adjust treatment and patient support accordingly.[5, 6] The complex disease manifestations of MS make PROMs even more pertinent and also render the development of relevant PROMs that can capture all aspects of the health status of PwMS more challenging.[7] Current evidence suggests that the views of patients, clinicians and researchers on relevant and valuable PROMs may differ substantially.[5, 6] To accurately reflect and assess patients' perceptions of their health status, their perspectives should be incorporated into the development process of PROMs.[6]

This systematic review aims to present the state of the art of PROMs for PwMS by identifying articles published within the last ten years that report on the development of MS-specific PROMs. Furthermore, we aim to explore the extent to which explicit patient participation was included in the development phases of these validated MS-specific PROMs.

Methods

MS-specific PROMs and the degree of patient participation in the development of these measures were identified in a systematic literature search according to the PRISMA statement.[8] The search was conducted on September 3, 2020 in the MEDLINE database via the PubMed platform, in the Embase database and the Cochrane library. The search was limited to studies published during 2011–2020 in English, Danish, Swedish or Norwegian. Four separate searches were performed on each database, with a variety of MeSH (medical subject headings) terms (i.e., multiple sclerosis, patient-reported outcome, patient outcome assessment, patient participation, survey/questionnaire) and free search terms. Search terms were identified based on an initial search of

Figure 1. Review process for the identification of PROMs in multiple sclerosis



the literature, and search strategies were developed with advice from an information retrieval specialist with expertise in health research and systematic reviews. (Search strategies can be obtained by contacting the correspondent author.) Two reviewers selected the articles independently, with discussions afterwards to avoid discrepancies.

Studies were included if they described a PROM developed specifically for PwMS. The exclusion criteria were as follows:

- study population not PwMS
- study does not describe an MS-specific validated PROMs
- study mentions the use of PROMs as an outcome measure but without a description of the development of the PROM

Explicit evidence of patient participation was explored using information extracted from the selected articles.

Results

A total of 157 articles were identified and titles and abstracts of 61 articles were screened, while a full text review was undertaken for 25 articles, of which 21 were excluded. The flow diagram in Figure 1 shows the number

Table 1. PROMs in MS identified from the literature published within the past ten years

PROM instrument	Author, year	Domains	Evidence of patient participation
12-item MS Walking Scale (MSWS-12)	Lejbkowicz et al., 2012[9]	Walking abilities	Yes (Developed based on interviews with PwMS)
The Fatigue Severity Scale (FSS)	Lejbkowicz et al., 2012[9]	Fatigue	No
Multiple Sclerosis Quality of Life-54 (MSQoL-54)	Lejbkowicz et al., 2012[9]	Health-related quality of life	No
Functional Assessment of MS (FAMS)	Lejbkowicz et al., 2012[9]	Health-related quality of life	Yes (Item generation by PwMS)
The MS Quality-of-Life Inventory	Lejbkowicz et al., 2012[9]	Health-related quality of life	No
The Leeds Multiple Sclerosis Quality of Life (LMSQoL)	Lejbkowicz et al, 2012[9]	Health-related quality of life	Yes (Developed from focus groups with PwMS)
The DYMUS Questionnaire	Solaro et al., 2012[10]	Dysphagia	No
MS Quality of Life (MSQOL)	Taheri et al., 2015[11]	Health-related quality of life	No
Patient-Determined Disease Steps (PDDS)	Learmonth et al., 2013[12]	Disability	No

of articles identified, included or excluded and the reasons for exclusion at the full-text level.

A total of four articles that represent literature published during the last ten years were included. Nine MS-specific validated PROMs were identified in these articles. The identified articles were scrutinized for explicit evidence of patient involvement in the development of the PROMs, and patient contributions to the specific PROM instruments were evident in three of the nine identified PROMs. A summary of the MS-specific validated PROMs and the evidence of contributions by PwMS to their development is presented in Table 1.

Discussion

In this review, MS-specific PROMs validated for patient populations with MS were explored with the aim of updating the state of the art of PROMs, according to developments during the past decade and gauging the extent of explicit patient participation in the development phases.

We identified four articles published within the last ten years that comment on the development of MS-specific validated PROMs.[9–12] The low number of articles indicates that the development of new MS-specific PROMs might have been negligibly small, suggesting that the field of MS is dominated by older PROMs. Of the nine PROMs found in the identified articles, only two (MSWS-12 and PDDS) were developed within the last ten years. This emphasizes a development gap and stresses the need for new MS-specific disease outcomes including PROMs that are validated, feasible in clinical encounters and representative of new insights into disease development. Within the past years, the MS treatment paradigm has changed rapidly from a focus mainly on the effectiveness of disease modifying therapies measured by the risk of relapses and Expanded Disability Status Scale (EDSS) score development to acknowledging that cognition, activities of daily living, fatigue and, more widely, rehabilitation are important for many PwMS.[13] Furthermore, there is increasing acknowledgement of the fact that for PwMS individualized symptomatic treatment options are strongly linked to quality of life.[2, 13, 14]

While we identified nine PROMs from the literature published within the last ten years, Khurana et al.[4] found 82 PROMs used in MS to assess symptom burden, function, quality of life, caregiver burden, treatment satisfaction and other attributes in their 2017 review of literature published between 1996 and 2015. Khurana et al. identified the eight PROMs most commonly used in clinical trials, namely, MSIS-29, LMSQoL, MSWS-12, FAMS, HAQUAMS, MUSIQoL, PRIMUS and MSQoL. Similar to the PROMS identified in this review, the majority of these had all been developed and validated prior to 2010. Based on Khurana et al.'s review, it appears

that the PROMs most commonly used within the field of MS are developed prior to 2010 and with limited patient involvement. The present review affirms that the MS-specific PROMs have not evolved much since then, and it is highly likely that the eight PROMs identified by Khurana et al. are still most commonly used within the field of MS. While the development of PROMs has not progressed, treatments for MS have advanced considerably over recent years. MS-specific PROMs should reflect the developments in MS care to be valuable to both patients and clinicians, which emphasizes the need for the development of newer MS-specific PROMs.

In recent years, patient involvement in the development of healthcare services has gained momentum as a key driver to ensure person-centered, acceptable and accessible treatments.[15, 16] PROMs constitute instruments that provide an opportunity for patients to contribute information regarding a range of relevant dimensions that affect the disease burden, quality of life, treatment trajectories and patient support. From a patient perspective, PROMs is an important tool for enhanced patient involvement. However, the PROMs do not necessarily reflect the patients' priorities and perspectives and may not successfully generate person-centered outcomes if they are not comprehensive and responsive to what patients experience with the disease.[6, 14]

In this review, we found evidence of patient involvement in the development of three of the nine MS-specific PROMs. Trujols et al. argued that the implementation of person-centered treatment entails applying outcome measures that reflect patients' priorities and perspectives.[6] Hence, patient participation in the development of PROMs is crucial to understand which domains to incorporate into PROMs to adequately capture and support the needs of PwMS in clinical encounters.[14] A study by Westergaard et al.[14] concluded that PwMS prefer PROMs to encompass a broad range of measures regarding neurological symptoms, cognitive impairments, mental health and well-being, self-care activities, and social challenges, and highlighted that the perspectives of patients do not always match the ones of the health professionals.[14]

Limitations

Many of the PROMs identified in this review were developed prior to 2010, and we are therefore unable to provide a thorough description of the development of each of these measurements or the potential extent of patient participation in their development processes. The determination of the exact extent of patient participation in the development of the identified PROMs was limited by the varying levels of information provided by authors.

Conclusion

This review identified nine different MS-specific validated PROM instruments. Explicit evidence of patients' contributions to the development of the PROMs were found in three instruments and only two identified PROMs were developed after 2010. This emphasizes the importance of patient involvement in the development of new MS-specific PROMs to ensure that they reflect and meet the needs and priorities of PwMS, as their perspectives on the most relevant domains may be divergent from the healthcare professionals.

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