Addressing Health-Related Quality of Life Among Children With Multiple Sclerosis

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CE INFORMATION

ACTIVITY AVAILABLE ONLINE: To access the article and evaluation online, go to https://www.highmarksce.com/mscare.

TARGET AUDIENCE: The target audience for this activity is physicians, advanced practice clinicians, nursing professionals, mental health professionals, social workers, and other health care providers involved in the management of patients with multiple sclerosis (MS).

LEARNING OBJECTIVES:

1. Describe the three theories discussed and characterize their overlap with usual care in order to implement changes to improve health-related quality of life in children with MS.
2. Describe how recommendations derived from these theories may improve the health-related quality of life of children with MS and their parents by strengthening self-concept, hope, and knowledge.

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ABSTRACT

BACKGROUND: Children with the chronic disease multiple sclerosis (MS) report lower health-related quality of life (HRQOL) compared with children who experience transient illness. The relationship between an MS diagnosis and the HRQOL of affected children is mediated by parental HRQOL. Interventions to improve the HRQOL of children with MS should, therefore, include parents of affected children.

METHODS: We performed a configurative review for improvements in the HRQOL of children facing diseases similar to MS and their parents. We used the generated concepts to form theories. Next, we performed qualitative interviews with clinicians who care for children with MS to characterize overlap between the proposed theories and usual care. Finally, we generated recommendations for improving the HRQOL of children with MS and their parents.

RESULTS: We theorize that the HRQOL of children with MS and their parents may be improved by strengthening self-concept, hope, and knowledge. Qualitative interviews with 7 clinicians who care for children with MS revealed no common psychosocial care protocol. The interviews did, however, reveal sources of psychosocial care that overlap with the proposed theories and barriers to optimizing such care.

CONCLUSIONS: Grounded in theory and clinically oriented practice, recommendations to improve the HRQOL of children with MS and their parents are to implement standardized screening, pool provider counseling strategies, create computer applications with psychosocial interventions, promote age-appropriate education resources, and secure positions for MS specialists.

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Children with multiple sclerosis (MS) report lower health-related quality of life (HRQOL) compared with children who experience monophasic acquired demyelinating syndromes.[1] Whereas HRQOL among adults with MS is associated with physical impairments, children with MS with low HRQOL mostly do not have functional physical impairments. Instead, parental HRQOL mediates the relationship between the diagnosis of MS and the children's HRQOL, suggesting that interventions to improve the HRQOL of children with MS should include their parents.[1] To date, research has focused primarily on diagnostic and treatment issues. No psychosocial care guidelines exist for improving the HRQOL of children with MS, and no studies have evaluated psychosocial interventions for children with MS and their parents.

When interviewed, children with MS report psychosocial challenges, including a more negative self-image after diagnosis,[2,3] the loss of their hoped-for future, and fear of the unknown.[4] Talking about the MS diagnosis with peers may aid the transition toward acceptance and psychosocial development; however, children have also reported strained peer relationships due to peer misinformation about pediatric MS, such as the need for a wheelchair.[3,4] Children with MS report similar challenges when discussing their illness and associated needs with teachers.[2]

Parents of children with MS also face psychosocial challenges. Parents can be anxious about their child's ability to continue participating in day-to-day life[5] and worry that the MS diagnosis is a death sentence.[6] They can also be frustrated when they are unable to address skepticism from family and others about their child's MS diagnosis due to their own poor understanding of the disease[7] and when they are unprepared for MS sequelae.[4,7] Parents describe using information searching as a coping mechanism; however, some parents despair when they uncover negative stories that create further uncertainty about how the disease will manifest in their lives.[5]

This study aimed to (1) generate theories to optimize the HRQOL of children with MS and their parents based on a review of concepts attributed to improvements in HRQOL among families facing similar diseases and (2) characterize overlap between proposed theories and the current psychosocial care delivered to children with MS and their families through qualitative interviews with care providers.

METHODS

Configurative Review

We performed a configurative review, which is a systematic review that tries to interpret and understand the world by arranging concepts to develop theories.[8] We identified concepts cited for improvements in HRQOL of children and parents facing diseases with stable physical statuses and unpredictable heterogeneous outcomes. Concepts were configured to generate theories (**APPENDIX S1**, available online at IJMSC.org).

Literature Search Strategy

Because the theories generated from this study are intended to inform the development of interventions for children with MS and their parents, we included studies that used the PedsQL measurement tools to detect changes in HRQOL.[1] PedsQL questionnaires are validated for use among children with chronic diseases and their parents and are used widely.[9,10] The literature search was conducted using the index of PedsQL publications made available via the instrument's author on the PedsQL website (http://www.pedsql.org). This index is believed to be comprehensive because to use the

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PedsQL tools, all publications must report to the author for inclusion in the index.

A single author (J.O'M.) performed a 2-stage literature screening process. She applied an open code to each article at each screening stage to capture the primary reason for inclusion or exclusion. This search strategy aligns with the methods of configurative reviews, which are meant to identify sufficient cases to explore patterns and are not meant to be exhaustive.[8]

We excluded articles during the first stage if (1) the primary objective was methodological (eg, validation of the PedsQL); (2) they involved evaluation of interventions that would preclude attribution of changes in HRQOL to improved management of psychosocial challenges; (3) the patient populations were palliative and faced psychological complexities associated with imminent death; (4) they did not evaluate interventions, mediators, buffers, or moderators; or (5) they evaluated diseases in which pain was a predominant symptom as pain is an uncommon symptom among children with MS and negatively impacts HRQOL.[11] Cross-disease comparisons of HRQOL were limited to intrastudy rather than interstudy comparisons to control for external factors that might contribute to differences in HRQOL across populations.

The second round of screening appraised whether each article provided evidence for a psychosocial intervention or mediator/moderator/buffer. We recorded all concepts cited as being associated with the HRQOL of children or their parents, the strength of evidence provided by the study, and the applicability to the present study. We excluded articles that did not provide evidence of a psychosocial intervention on HRQOL.

Concept Categorization

We recorded all concepts cited in the included articles associated with improved HRQOL of children or parents. Axial coding was performed to identify relationships among the concepts. The information about the concepts provided in the included articles informed the configuration of 3 theoretical models (Appendix S1).

Qualitative Interviews

One of us (J.O'M.) then performed qualitative interviews to characterize how the theories generated from the configurative review align with current clinical practice. Interviews and accompanying analyses were guided by the principles of grounded theory to allow for exploration and in-depth understanding of clinicians' perspectives.[12] The University of Toronto Research Ethics Board approved this study.

Sampling and Recruitment

To capture standards of psychosocial care across different avenues of care, we invited 3 groups of health care providers to participate in qualitative interviews: pediatric MS specialists, adult MS specialists who treat children with MS, and pediatric non-MS specialists who treat children with MS. Drs Banwell and Marrie are principal investigators of the Canadian Pediatric Demyelinating Disease Network and identified the providers who were invited to undergo interviews.[13]

Data Collection

Interviews were directed by a topic guide (**APPENDIX S2**), conducted in person or via telephone, recorded, and transcribed verbatim by one of us (J.O'M.). Data collection continued until categories were fully developed and saturation was reached. One of us (J.O'M.) presented a verbal summary with visual adjuncts of our previous empirical findings[1] and an overview of the proposed theories so that clinicians could discuss how the theories generated from the configurative review aligned with their usual clinical care. Participants did not have access to the empirical findings or theories before the interviews.

Data Analysis

Interviews were analyzed inductively using qualitative data analysis software (NVivo; QSR International).[14] Initial codes were developed from line-by-line coding that examined experiences, actions, and assumptions.[12] Open and focused coding was used to develop categories and understand their relationships. We adhered to standards for performing and reporting qualitative research.[15]

Reflexivity

The interviewer (J.O'M.) maintained a reflexive standpoint throughout the study to account for her potential to influence the research process and co-construct data with interviewees.[16] A reflexive journal was used to record her beliefs, actions, and observations that might have influenced data generation and analysis. She considered how her positionality and professional knowledge may have shaped her interaction with the interviewees.

RESULTS

Configurative Review

Sixty-seven of the 1677 articles listed on the PedsQL index on March 4, 2019, cleared the first round of screening, and 36 articles cleared the second round. Characteristics of the included articles are summarized in **TABLE S1**.

The concepts identified as being associated with improved HRQOL of children or parents were configured into self-concept, hope, and disease-specific knowledge theories.

Self-concept

Of the 36 included studies, 8 described interventions that facilitated the child's or parent's formation of a new self-schema that was not jeopardized by the child's disease (**TABLE 1**). Self-schema—beliefs about one's self that inform one's overall self-image—relate to various aspects of the current and future self, including personality, skills, abilities, occupation, hobbies, and physical characteristics.[17] Each person forms self-schema based on their environment and external cues. Self-concept is the summation of the self-image and the ideal self.[17] Discordance between the self-image and the ideal self can cause low self-esteem and emotional dysfunction, observed among children with MS and their parents.[1] A person may experience reduced HRQOL when they are unable to fulfill their current self-schema or are unsure of their ability to fulfill

TABLE 1. Concepts in the Included Articles Associated With Improved HRQOL for Children or Parents and Their Associated Theory

Concept	Articles, No.	Theories
Disease-specific knowledge	14	Disease-specific knowledge
(Perceived) social support (emotional and instrumental)	7	Disease-specific knowledge
Peer support or acceptance	2	Disease-specific knowledge
Conflict resolution	2	Disease-specific knowledge
Self-esteem enhancement	3	Self-concept
Self-concept	2	Self-concept
Family rituals	1	Self-concept
Family cohesion	2	Self-concept
Problem-solving	5	Hope
Self-efficacy	3	Hope
Perceived vulnerability	2	Hope
Hope	1	Hope
Goal setting	4	Hope
Locus of control	1	Hope
Coping skills	3	Hope
Motivational interviews	3	Hope
Empowerment	2	Hope
Positive self-talk	2	Hope
Communication (role playing, parent-child, parent-child-HCP)	7	NA
Discussing HRQOL	1	NA
Resilience	2	NA
Parental distress and stress management	4	NA
Worry	1	NA

HCP, health care provider; HRQOL, health-related quality of life; NA, not directly included in theories configured for this review.

their future self-schema, as may be the case for children with MS and their parents.

We theorize that the HRQOL of children with MS and their parents may be improved by ensuring they form self-schemas that are not threatened by MS (**FIGURE S1**). We propose that this can be achieved by changing the environment, changing the external cues, and reframing ideal-self expectations. We further theorize that parents and children enable formation or reframing of self-schema in one another through environmental changes and external cues.

Hope

The Snyder hope theory is grounded in the observation that humans are constantly thinking about how to reach their goals and that their emotions reflect that perceived capacity; a high-hope person has enduring positive emotions with a sense of affective zest, whereas a low-hope person has negative emotions with a sense of affective lethargy.[18] Thus, hope is determined by whether there are workable routes (ie, pathways) to desired goals and the person's willingness to use the identified routes to reach desired goals (ie, agency).[18] Concepts associated with the Snyder hope theory were cited by 14 of the studies as related to improved HRQOL among children and parents (Table 1).

We theorize that children with MS and their parents may report improved HRQOL if they can achieve agency and establish pathways to achieve their goals even when the goals are unrelated to the child's MS (**FIGURE S2**).

Disease-Specific Knowledge

The Mishel uncertainty in illness theory provides a framework to understand how disease-specific knowledge may improve HRQOL by reducing uncertainty. Uncertainty in the context of illness occurs when those affected are unable to understand how the disease will impact their lives.[11] The HRQOL may improve when affected individuals are able to detect patterns in the disease course and clearly understand how it may impact their lives.

Disease-specific knowledge may also improve HRQOL through strengthened social connections by providing families with the vocabulary to respond to questions or skepticism and communicate their experiences, fears, and needs.[5] For families choosing not to disclose the child's MS diagnosis, disease-specific knowledge may allow them to communicate what is likely central to their lives without violating their child's privacy. Disease-specific knowledge was associated with improved HRQOL in 14 of the included articles.

We theorize that disease-specific knowledge may improve HRQOL among children with MS and their parents by reducing perceived uncertainty (**FIGURE S3**) and by providing families with the knowledge necessary to communicate their experiences to others.

Qualitative Interviews

Of 14 providers invited to undergo qualitative interviews, 13 (93%) agreed to participate and 1 did not respond. Saturation was reached on completion of interviews with 7 participants. Characteristics of the 7 participants are presented in **TABLE 2**. No differences were observed between in-person and telephone interviews. Interviews adhered to the topic guide, with flexibility for development of themes.

Ascertainment of HRQOL

Participating clinicians reported that they detect low HRQOL through red flags, conversations with families, responses to questions about mood during clinical assessments, and families reaching out to them. Red flags cited by providers included nonadherence to treatments, depressive symptoms, comments about substantial frustration with the limitations of their illness, moodiness, irritability, and absence from school. One clinician who treats adults uses a questionnaire to screen for depression among people with MS. None of the participants used measurement tools to ascertain HRQOL as part of usual care of their patients or their patients' parents.

TABLE 2. Characteristics of the 7 Care Providers Who Participated in Qualitative Interviews

Characteristic	Values
Age at interview, median (range), y	52 (37–63)
Pediatric MS specialists, No.	
Neurologist	1
Pediatric non-MS specialists, No.	
Neurologist	2
Neurologist	1
Country of current medical practice, No.	
United States	3
Location of practice, No.	
Pediatric tertiary non-MS center	2
Adult tertiary MS center	2
Experience, median (range), No.	
Years since graduation from nursing program or medical school, median (range)	28 (11–43)
Children with MS seen since graduation from nursing or medical school,[b] median (range)	150 (20–185)
Adult-onset MS cases seen since graduation from nursing or medical school	50 (0 to >5000)
Qualitative interviews	
Duration, median (IQR), min	42 (36–50)

IQR, interquartile range; MS, multiple sclerosis.
[a]Three of 7 participants (43%) completed MS-specific training.
[b]One participant declined to respond to the number of children with MS to whom she has provided care.
[c]One participant declined to respond to the number of people with pediatric-onset MS to whom she has provided care.

Overlap Between Theories and Usual Care

Qualitative interviews revealed no common protocol for providing psychosocial care to children with MS and their parents but did identify 3 sources of psychosocial support that overlap with the proposed theories: provider counseling, standardized programs, and departmental referrals to social work, neuropsychology, and psychiatry.

Provider Counseling

Providers saw their own counsel as a form of psychosocial support delivered as part of routine clinical care or in response to parent outreach. They described standard pieces of information (herein, "talking points") that they conveyed to each child with MS and their parents. These talking points were formed by negative experiences that they observed among families facing pediatric MS. For instance, a pediatric neurologist observed family distress due to misinformation about the likelihood of wheelchair use that the family received from a referral center at the time of diagnosis. Thereafter, he made sure to identify and expel myths during his first encounter with children with MS. "I make a point in the first visit to tell them, and I'm cautious about how I say it, but I say that my goal is that, in 10 years, you're dancing at your wedding, you're having kids. My goal is to keep you as you are now in the future. I can't promise it, obviously, but my goal for you is to not be in a wheelchair" (provider 5).

Providers also described talking points that prepared families for what to expect during the disease course. "I usually tell them that there are very few emergencies in MS and so, oftentimes, things can wait until the morning without necessity of them going to the ER. I think what often relieves a lot of them, too, is the fact that they're not going to have to rush to the ER at midnight because their child is experiencing numbness" (provider 5).

Other talking points covered the distinction between progressive and relapsing disease; the disproportionate number of people with primary progressive MS on the internet whose experiences are unique to those with progressive disease; the low prevalence of physical deficits among children with MS; concrete analogies to explain MS pathobiology; and the importance of including the child when deciding to whom to disclose the child's diagnosis.

Talking points about the MS disease course tell affected children and their parents about how MS might affect the ability to fulfill their self-schema and achieve their goals; they align with the theories of self-concept and hope. Talking points that convey information about how families should respond to signs of MS align with the patterned information component of the knowledge theory. Talking points that explain MS pathobiology align with the communication component of the knowledge theory.

Standardized Programs

Of the interviewed clinicians, 2 offer standardized educational programs. The first offers a 45- to 60-minute session with a trained nurse who covers MS risk factors, diagnostic criteria, identifying relapses and what to do when they occur, prognosis, common symptom management, therapies, wellness, health maintenance, and resources available through the clinic. These sessions are tailored to the individuals with MS, but family members are welcome to attend. The other standardized program is a peer-to-peer match program for children and parents.

These standardized programs align with all the proposed theories. They provide families with an understanding of how the disease might affect their ability to fulfill their

self-schema and achieve their goals, thereby aligning with the self-concept and hope theories. They also align with the knowledge theory by providing families with information about possible manifestations of the disease and information to enhance communication.

Referrals

Referrals to social workers, clinical or health psychologists, and psychiatrists were consistent with the theories on self-concept and hope. Providers with specialized MS training may provide families with information about how the disease might affect their self-schema or pathways to achieve their goals, and specialists without MS-specific training may provide general reinforcement of self-schema and goal setting that may strengthen feelings of hope. External resources are also available, such as the National Multiple Sclerosis Society's MS Navigator program, which connects people with MS to mental health providers.[13]

Barriers and Recommendations

The qualitative interviews with the clinicians made it clear that providers offer a wealth of psychosocial resources to children with MS and their families that overlap with the proposed theories, but there are also barriers to optimizing these resources.

Limited Standardized Screening

Most participants used unsystematic and time-consuming strategies to detect low HRQOL. Psychosocial care that aligns with the proposed theories cannot be implemented if the need for such care is unknown.

Recommendation. We recommend standardized screening programs to detect low HRQOL among children with MS and their parents. Questionnaires are a straightforward way to identify clinically relevant concerns that may be missed during clinical examinations. Learning health systems are poised to enable families to record their HRQOL before clinical encounters and then join HRQOL and clinical data for clinicians to visualize and export findings to research registries. Patients have reported valuing learning health systems for the individualized information they provide.[19] Clinically meaningful cutoff values are not known for all HRQOL questionnaires, but individual questions can serve as clear beacons for clinician concern, including a response of "almost always" to statements on the PedsQL parent report such as "It is hard for me to tell doctors and nurses how I feel," "I worry about my child's future," "I worry about the side effects of my child's medication/medical treatments," and "I worry about how others will react to my child's condition."

Confidential questionnaires may also increase the chance of identifying concerns. Individuals may be more forthcoming about mental health concerns through written communication, and confidentiality offers children and parents the opportunity to report concerns that they may not feel comfortable raising in front of one another. Clinicians in this study sensed that parents were hesitant to raise concerns in front of their child for fear of making them worry or feel like a burden.

Resources Are Siloed

Three participants relayed experiences of misinformation communicated to children with MS and their parents by clinicians, often at referral centers, that are obvious sources of psychosocial distress for families.

"I've had experience with several patients who are told things at outside institutions that are sometimes very shocking in terms of what their life is going to be like. I mean I had kids who have been told, or at least tell me they've been told, that they were going to die at an early age, that they'll be in a wheelchair, that they could never get pregnant. It's pretty shocking" (provider 5).

Providers also noted that much of their counseling occurs as second nature, especially for those who care for thousands of patients, and would require further evaluation to understand.

"Some of these things we do without necessarily being conscious of why we do them. So, it's trying to ask somebody why you felt like this person needed that thing. So some of us will say, well 'I just felt it, knew, and I've seen it before,' but you really want to be able to decompile that so anybody on your team can actually identify that individual and do it and you're likely not to get there unless you actually have people who are consciously aware of what the issues are and then adopt some framework to work within it. And so, coming from different backgrounds, different members of the team probably frame these issues in a different way and adopt different interventions and some people probably don't have as conscious a framework. So it's probably worthwhile even as a starting point to help people understand what these frameworks are" (provider 3).

Recommendation. In the interviews, clinicians cited talking points as valuable components of psychosocial care; this overlaps with the proposed theories. We recommend that a set of talking points be made available so that all families are provided with accurate information to mitigate psychosocial distress. The International Pediatric Multiple Sclerosis Study Group is a global network of clinicians and researchers whose mission is to improve the care of children with MS by promoting clinical initiatives, education, and research[20] and would, therefore, be well-suited to compile and distribute these talking points.

The clinician interviews also highlighted a wealth of MS-specific clinical acumen that underlies psychosocial care and overlaps with the proposed theories, but participants noted that there are no frameworks for such counseling. We recommend study, production, and then distribution of psychosocial care frameworks for MS providers. Neurologists participating in postgraduate training in MS clinics would be well-suited to conduct qualitative studies including interviews with MS specialists to gain a better understanding of how they deliver care; these studies could be funded by national and international MS societies.

Limited Availability of Mental Health Professionals

Providers were confident in their ability to provide psychosocial support and issue referrals to mental health professionals; however, they also reported time constraints and the

limited availability of mental health professionals as barriers to optimizing these resources.

Recommendation. The articles included in the configurative review revealed that psychosocial intervention computer applications can have meaningful impacts on HRQOL.[21] Applications allow for broad access and do not require clinician time.

J.O'M.: It sounds as though a lot of the models that I'm proposing align with your beliefs and observations, but I'm not sure that they're being implemented.

Provider 4: No, I'm sure they're not. I'm sure they're not and they can't ever be. The clinicians don't have time so we have got to figure out some way of imparting it. Maybe an app. Maybe some sort of internet communication. It's time. It's time now.

We, therefore, recommend the use of computer applications with psychosocial interventions that align with the proposed theories. Importantly, any psychosocial intervention computer application available to people with MS should include a 24/7 crisis hotline number and a caveat that one should go to an emergency department when experiencing severe distress, psychopathology, and/or suicidal ideation.

Educational Resources

This study highlights the importance of disease-specific knowledge and the existing paucity of educational materials, particularly age-appropriate resources, for families facing pediatric-onset MS. With more than 1863 existing academic publications about pediatric-onset MS, and the data from longitudinal observational studies in the past 15 years, data to form the content of such resources are readily available.

Recommendation. Providers noted the importance of information, with a clinician stating, "Information is one of the things that people need to figure out their pathway" (provider 3). Information is a core component of the knowledge theory. We recommend that age-appropriate educational materials be compiled and made centrally available. Educational materials are priorities for national MS societies; this effort would, therefore, be likely to garner their support.

Limited MS Workforce

This study revealed that MS specialists are valuable sources of and gateways to psychosocial care that overlaps with the proposed theories. It is, therefore, imperative to address the projected shortage of MS specialists anticipated to occur in the next 2 decades.[22] A participant described the potential consequences of a shortage to the psychosocial health of children with MS who do not have access to pediatric MS clinics. "We have children and adults in the same clinic, which may be prone to increased emotional distress because they see adults in wheelchairs and using canes and elderly individuals because it's a general neurology clinic that deals with strokes and you know people with severe epilepsy wearing helmets, so it's an environment that can be potentially overwhelming for kids. Ideally there would be enough kids or space where one could have them be in a more pediatric-centered environment" (provider 3).

PRACTICE POINTS

The health-related quality of life of children with multiple sclerosis (MS) and their parents may be improved by encouraging them to not define themselves by the MS diagnosis, promoting goal setting and goal attainment, and enhancing their MS knowledge.

Psychosocial care for children with MS and their parents may be improved by implementing standardized screening for low health-related quality of life, ensuring the MS specialist workforce, and providing clinicians with talking points for children with MS and their parents, computer applications with psychosocial interventions, and age-appropriate educational resources. ■

Recommendation. We recommend implementation of strategies to encourage postgraduate medical residents, medical students, and health psychologists to pursue specialized training in MS (eg, fellowships) to facilitate ongoing delivery of psychosocial care that aligns with the proposed theories. Studies suggest that more neurology residents are interested in pursuing MS fellowships than there are funded positions available; increasing the number of funded MS fellowships may, therefore, help secure MS specialists.[22] In addition, securing funding for research grants for MS specialists may incentivize trainees to pursue an MS subspecialty.

DISCUSSION

This study generated 3 theories about interventions that may improve the HRQOL of children with MS and their parents. Qualitative interviews with clinicians characterized the existing psychosocial care that overlaps with the proposed theories and the barriers to the delivery of that care. The present findings allowed us to make 5 recommendations to optimize the HRQOL of children with MS and their parents that are grounded in theory and clinical practice.

The recommendations align with the conclusions of previous studies. Pooling provider talking points echoes previous calls to reassure families that MS during childhood is not fatal.[2,6,7] Computer applications address previous concerns about the inadequate availability of mental health services for youth with MS and their parents.[7] Standardized screening for low HRQOL is consistent with parents reporting that they would welcome screening and referrals for their own mental health from their

child's care providers.[23] Increased educational resources aligns with previous appeals for improved educational resources for families facing pediatric-onset MS.[6,7]

This study addressed the psychosocial needs of a specific subgroup: children with MS who face minimal physical impairments. Due to earlier diagnosis and the rapidly improving MS treatment landscape, more children live with MS without physical deficits and may benefit from this study. The present findings, however, are not necessarily generalizable to people who face persistent physical deficits, pain, or fatigue, which are known to affect HRQOL.[11] Strategies to maximize the HRQOL of people with MS who face such challenges warrant further investigation.

The configurative review benefited from systematic methods to limit bias and maximize the potential for reproducibility. The present findings, however, may have been influenced by publication bias from the disproportionate publication of studies with statistically significant results.[24] Publication bias may have led to an overestimate of the potential for the proposed theories to positively impact HRQOL. Future studies should account for this potential bias. We made a conscious effort to limit the terminology and concepts of the theories to those cited within the articles included in the configurative review to maximize reproducibility. For example, models of resilience overlap with the hope theory and concepts such as identity, liminality, and cognitive dissonance overlap with the self-concept theory; however, these concepts did not appear in the configurative review articles and, therefore, were not cited in the proposed models. Spiritual and religious beliefs were not cited in the included articles; however, they have been cited elsewhere by children with MS and their parents and may warrant further exploration.[7] To overcome the limitation of having a single reviewer, each stage of article adjudication and inclusion were fully documented.[8]

The recommendations generated from this review address psychosocial challenges previously raised by families facing pediatric-onset MS and substantiate the conclusions of previous studies. The resources needed to implement the recommendations and improve the HRQOL of children with MS and their parents are readily available. ■

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