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Original Article

Multiple System Atrophy Caregivers' Experience: A Mixed Methods Study

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Abstract: ***Background:*** Multiple system atrophy (MSA) is a rare Parkinson-plus syndrome with rapid progression and a high symptom burden. The experience of caregivers of people with MSA has not been closely examined. We therefore sought to document the impact of MSA on caregivers using a mixed methods approach. ***Methods:*** Patients and caregivers were recruited from a movement disorders program in Edmonton, Canada. Participants completed the following survey instruments based on their own or their loved one's symptoms: 36-Item Short-Form Health Survey (SF-36), Multiple System Atrophy health-related Quality of Life scale (MSA-QoL), and Hospital Anxiety and Depression Scale (HADS). Caregivers also completed the Zarit Burden Interview and HADS based on their own experience. Qualitative data were obtained through semi-structured interviews. ***Results:*** Nine people with MSA (PwMSA) (age range: 48–78 years) and 11 caregivers (49–76 years) participated. All completed surveys; 7 PwMSA and 10 caregivers were interviewed. Eight PwMSA had the parkinsonian type of MSA (MSA-P) and one a mixed type. Caregivers had on average mild-moderate caregiver burden and mild anxiety. Caregiver burden and anxiety were correlated. Qualitative subthemes under the caregiving theme included keeping the patient safe, caregivers' own health, and communication symptoms cause frustration. The rapid progression of illness was bewildering to caregivers and increased their workload. Public home care services were invaluable to caregivers' maintaining their loved ones at home. Caregivers were inventive in finding sources of hope and quality of life for their loved ones. ***Conclusion:*** Publicly funded home care was essential for caregivers of PwMSA in this study. Caregiver support is needed to provide this unrecognized workforce with information and resources to face this challenging condition.

RÉSUMÉ : Expérience de proches aidants s'occupant de patients atteints d'atrophie multi-systématisée : une étude basée sur diverses méthodes de recherche. ***Contexte :*** L'atrophie multi-systématisée (AMS) constitue un syndrome parkinsonien atypique peu fréquent dont la progression est rapide et dont les symptômes imposent un lourd fardeau. À cet égard, l'expérience de proches aidants s'occupant de patients atteints d'AMS n'a pas été étudiée de près. Nous avons donc cherché à documenter l'impact de l'AMS sur les proches aidants en recourant à une approche basée sur diverses méthodes de recherche (*mixed methods approach*). ***Méthodes :*** Les patients et leurs proches aidants ont été recrutés à partir d'un programme de prise en charge des troubles du mouvement organisé dans la ville d'Edmonton (Canada). Les participants ont rempli les instruments d'enquête suivants en fonction de leurs propres symptômes ou de ceux des personnes dont ils prenaient soin : SF-36 (*36-item short form*), MSA-QoL (*multiple system atrophy quality of life)* et HADS (*hospital anxiety and depression scale*). Les proches aidants ont également rempli les instruments ZBI (*Zarit burden interview*) et HADS sur la base de leur propre expérience. Enfin, mentionnons que des données qualitatives ont été obtenues par l'entremise d'entrevues semi-structurées. ***Résultats :*** Au total, nous avons pu compter sur la participation de neuf patients atteints d'AMS (tranche d'âge : 48 à 78 ans) et de onze proches aidants âgés de 49 à 76 ans. Ces vingt individus ont tous complété les instruments d'enquête cités ci-dessus ; sept patients et dix proches aidants ont été par ailleurs interviewés. Huit patients ont donné à voir le type parkinsonien de l'AMS alors qu'un seul était atteint d'un type mixte. Les proches aidants ont fait part d'un fardeau en moyenne léger à modéré et d'une légère anxiété. À ce sujet, on a établi qu'il y avait une corrélation entre leur fardeau et leur anxiété. Parmi les sous-thèmes abordés par les proches aidants, soulignons les suivants : assurer la sécurité des patients ; veiller à leur propre santé ; et faire face à la frustration causée par des symptômes liés à la communication. La progression rapide de la maladie s'est avérée déconcertante pour ces proches aidants et a eu pour effet d'augmenter leur charge de travail. Aux yeux des proches aidants, les services publics de soins à domicile ont aussi représenté un apport inestimable en vue de maintenir leurs proches malades à domicile. Ces proches aidants ont été par ailleurs inventifs pour trouver des sources d'espoir et de qualité de vie pour leurs proches malades. ***Conclusion :*** Dans cette étude, on a pu observer que les soins à domicile financés par des fonds publics étaient essentiels pour les proches aidants de personnes atteintes d'AMS. Du soutien procuré à ces proches aidants (des ressources et de l'information) est donc nécessaire afin que cette main-d'œuvre méconnue puisse faire face à cette condition médicale difficile.

Keywords: Multiple system atrophy (MSA); Caregivers; Carers; Caregiver experience; Caregiver burden; Quality of life

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Introduction

Multiple system atrophy (MSA) is a rare parkinsonian syndrome (prevalence 1–9/100,000[1]) characterized by parkinsonism, cerebellar symptoms, and autonomic symptoms in varying combinations.[2] It is rapidly progressive, with a mean duration from clinical diagnosis to death of 9 years.[3] Due to the lack of specific treatments and the rapid progression of MSA, maintaining an acceptable quality of life throughout the course of the illness is of high importance to patients and clinicians, and palliative care approaches are beneficial.[4] Informal caregivers, usually the patient's spouse, play a central role in the care of people with MSA (PwMSA). Caregivers are an essential and often under-recognized health care resource worldwide.[5] In Canada, one in four adults provide care to a family member with a long-term health condition or disability.[6] In the USA, as much as 18.2% of the population provides care to an adult relative.[7] Understanding the needs and concerns of caregivers is essential to providing effective information, resources, and services to support them in the care of their loved ones.

The experiences of caregivers of PwMSA have rarely been studied; however, MSA caregivers may face unique challenges due to the rapid progression of MSA[4] and the prevalence of MSA symptoms affecting communication.[8] In a study of caregivers of people with parkinsonism, the diagnosis of a Parkinson-plus syndrome such as MSA was associated with increased caregiver burden.[9] In an interview study that examined quality of life in MSA and progressive supranuclear palsy (PSP) – another rare Parkinson-plus syndrome – caregivers experienced challenges in maintaining a social life outside the home and worried about the safety of the person receiving care if they left them on their own.[8] However, the caregivers' relationships with their spouses (the person with MSA or PSP) remained positive and meaningful despite severe dysarthria making communication challenging.[8]

This study used mixed methods (quantitative surveys and qualitative semi-structured interviews) to investigate the experiences of caregivers of people with MSA. We hypothesized that there would be associations between disease severity and caregiver burden, patient quality of life and caregiver burden, patient anxiety and depression and caregiver anxiety and depression, and caregiver anxiety and depression and caregiver burden. Since most caregiver survey instruments characterize caregiving in terms of burden and strain, we also sought to explore the experience of caregiving through qualitative interviews and analysis to more fully characterize the caregiving experience and to allow for triangulation[10] with the quantitative data.

Methods

Ethics approval was granted by the University of Alberta Health Research Ethics Board – Health Panel (Pro00090253).

Research Participants

Participants were recruited from the University of Alberta Parkinson and Movement Disorders Program and the Parkinson Association of Alberta. Inclusion criteria for people with MSA were an established diagnosis of MSA using the Second Consensus Statement on the Diagnosis of MSA[11], able to provide consent, able to complete an oral interview, and fluency in spoken English. Exclusion criteria for people with MSA were unable to provide consent or assent, or unable to complete an oral interview. Inclusion criteria for caregivers were person identified by the person with MSA as their caregiver, able to provide informed consent, able to complete an oral interview, and fluency in spoken English. Exclusion criterion for caregivers was unable to provide informed consent.

Study Design

This study was structured using a convergent parallel mixed methods design.[12] Data from quantitative surveys and qualitative interviews were collected concurrently, then analyzed separately.[12] Concurrent collection of quantitative and qualitative data was chosen to facilitate the timely participation of people with MSA, who face increasing communication difficulties as the illness progresses. Data collection took place between July 2019 and March 2020.

Study Measures

Baseline Characteristics

Baseline demographic and medical data collected for PwMSA included age, sex, MSA type (MSA-P, parkinsonian type, or MSA-C, cerebellar type), duration of MSA, geographic location (urban vs. rural), and household income. Baseline data for caregivers included age, sex, relation to the participant with MSA, geographic location, household income, duration of MSA, and duration of caregiving role.

Quantitative Instruments

All participants with MSA completed the following instruments: the 36-Item Short-Form Health Survey (SF-36, a measure of health-related quality of life, or HRQoL[13]), MSA-QoL (a measure of HRQoL specific to MSA[14]), and the Hospital Anxiety and Depression Scale (HADS, a screening tool for anxiety and depression[15]). MSA participants could have assistance to complete the surveys if needed (due to impaired manual dexterity) but were instructed that answers should reflect their own responses.

Caregivers completed the Zarit Burden Interview (ZBI, a measure of caregiver burden[16]) modified to include two questions regarding physical and sexual aggression[16], SF-36, MSA-QoL, and HADS based on their assessment of their dyad participant with MSA, and HADS based on their own symptoms.

Movement disorders neurologists completed the Unified MSA Rating Scale (UMSARS, a measure of disease severity[17]) and Hoehn and Yahr stage (a measure of disease severity[17]) and provided information on MSA type (MSA-P or MSA-C).

Statistical Analysis

Statistical tests were carried out in R (version 4.0.4). The Pearson correlation test was used to measure correlation between variables. To limit the number of statistical comparisons made, data from the SF-36, which consists of eight scales, were transformed before analysis into two component scores, the physical component score (PCS) and mental component score (MCS),[18] using Canadian population norms.[19]

Qualitative Interviews

Interviews were conducted by BL, using a semi-structured interview guide (Supplemental data 1) developed by BL and JMM. Interviews with PwMSA included questions regarding participants' experience of MSA, the disclosure of MSA by the participant's neurologist, symptoms, and supports and services. Caregiver participants were asked the same questions as well as additional questions about their experience of caregiving. The interview structure was flexible and allowed for follow-up and probing questions. Review of the first two interviews allowed refinement of the interview process. Interviews took place in person or by phone, lasted approximately 25–40 minutes, and were audiotaped and transcribed by a professional research transcriptionist. Each participant was interviewed individually.

Coding of interviews was conducted using NVivo 12 qualitative coding software by QSR International. Coding occurred continuously as interviews were completed. Earlier interviews were open-coded to identify potential themes. Codes were refined during coding of subsequent interviews, including development of definitions and examples for codes. Qualitative validity and reliability were ensured by: 1) reading and re-reading interview transcripts to ensure full understanding; 2) ensuring a clear definition of each code agreed upon by BL and JM; 3) meetings to discuss coding and analysis of coding; 4) using consensus to resolve disagreements in coding; and 5) member checking of codes, after all coding was complete, in January and February 2021. Member checking involved obtaining feedback from participants regarding identified themes and interpretations and is a tool to increase the validity of qualitative results.[20] Qualitative research guidelines (COREQ[21] and Yardley's criteria[22]) were applied to the study methods to improve the validity and reporting of the qualitative portion of the study.

Results

Baseline Characteristics

Nine PwMSA and 11 caregivers enrolled in the study. All participated in the survey portion of the study. Seven PwMSA and 10 caregivers were interviewed; one PwMSA and one caregiver were lost to follow-up after survey completion, and one PwMSA was not able to participate in the interview due to severe dysarthria. During the recruitment process, an additional four eligible patients or their families were approached to participate but declined or did not return consent forms. All participants were recruited from the University of Alberta Parkinson and Movement Disorders Program; the study was also advertised through the Parkinson Association of Alberta, but it did not yield additional participants. Demographic information of participants is presented in Tables 1 and 2. Three men and five women with MSA participated. The age range was 48–78 years (mean = 67). The majority of PwMSA lived in a major urban center. Six PwMSA provided household income data, with four having household income of $100,000 Canadian dollars or more. Eight participants had MSA-P and one a mixed type of MSA. The majority (55.6%) of PwMSA were Hoehn and Yahr stage 3 (range = 2.5–5). The duration of diagnosis was 2–24 months with an average duration of 11.4 months. Among caregivers, six women and four men participated with age range 49–76 years (mean = 63). The average duration of caregiving was 50 months with a range of 8 months to 8 years. Eight were spouse caregivers, one was a sibling, and one was an adult child. A broader range of household income was reported by caregiver participants, although five reported incomes of $100,000 Canadian dollars or more.

Table 1: Characteristics of participants with MSA

Variable	People with MSA	
Total sample size, *n*	*n* = 9*	
Age, mean ± SD	*n* = 8	67.4 ± 9.4
Range		48–78
Sex, *n* (%)		
Female		5 (62.5)
Male		3 (37.5)
Geographic location, *n* (%)		
Within major center (Edmonton, Alberta)		5 (62.5)
Within 50 km of major center		2 (25)
>200 km to major center		1 (12.5)
Household income, *n* (%)	*n* = 6**	
$40,000–$59,999		1 (16.7)
$60,000–$99,999		1 (16.7)
$100,000–$149,999		2 (33.3)
$150,000–$250,000		2 (33.3)
MSA subtype, *n* (%)	*n* = 9	
MSA-P		8 (88.9)
MSA-C		–
Mixed		1 (11.1)
Hoehn and Yahr, *n* (%)	*n* = 9	
Stage II.5		1 (11.1)
Stage III		5 (55.6)
Stage IV		1 (11.1)
Stage V		2 (22.2)
UMSARS Part IV, *n* (%)	*n* = 9	
3		2 (22.2)
4		6 (66.7)
5		1 (11.1)
Duration of MSA diagnosis	*n* = 8*	
Months, mean ± SD		11.4 ± 9.3
Range		2–24

*Demographic information (except for MSA subtype, Hoehn and Yahr stage, and UMSARS) was not obtained for one PwMSA/caregiver pair who did not participate in the interview portion of the study.**Income data not available for participants who declined or did not have information on household income.

Quantitative Results

Surveys were completed by 9 PwMSA and 11 caregiver participants. The mean HADS anxiety score (HADS-A) for caregivers was 8.64 ± 3.38, and the mean HADS depression score (HADS-D) for caregivers was 5.18 ± 4.00 (Table 3). HADS scores below 8 indicate no depression or anxiety, and scores between 8 and 10 indicate possible cases of anxiety or depression.[23] The mean modified ZBI score was 36.73 ± 16.38 (range = 6–66), consistent with mild to moderate caregiving burden (Table 3).

There was no correlation between PwMSA quality of life scales or subscales (SF-36 and MSA-QoL) and caregiver ZBI (Table 4).

Table 2: Characteristics of caregivers

Variable	Caregivers
Total sample size, *n*	*n* = 10*
Age, mean ± SD	62.6 ± 8.9
Range	49–76
Sex, *n* (%)	
Female	6 (60)
Male	4 (40)
Relationship of caregiver to PwMSA, *n* (%)	
Spouse	8 (80)
Sibling	1 (10)
Child	1 (10)
Geographic location, *n* (%)	
Within major center (Edmonton, Alberta)	5 (50)
Within 50 km of major center	4 (40)
>200 km to major center	1 (10)
Household income, *n* (%)	*n* = 9**
$40,000–$59,999	2 (22.2)
$60,000–$99,999	2 (22.2)
$100,000–$149,999	2 (22.2)
$150,000–$250,000	3 (33.3)
Duration of MSA diagnosis	
Months, mean ± SD	16.3 ± 17.4
Range	2–60
Duration of caregiving	*n* = 9***
Months, mean ± SD	50 ± 35.2
Range	8 months–8 years

*Demographic information was not obtained for one PwMSA/caregiver pair who did not participate in the interview portion of the study.**Income data not available for participants who declined or did not have information on household income.***Omitted data from one participant who had been caring for spouse since unrelated illness predating MSA.

Table 3: HADS scores and modified Zarit Burden Interview scores of caregivers

	n	Mean ± SD	Range
HADS-A	11	8.64 ± 3.38	5–15
HADS-D	11	5.18 ± 4.00	0–11
Modified ZBI score	11	36.73 ± 16.38	6–66

HADS: Hospital Anxiety and Depression Scale, HADS-A: HADS anxiety subscale, HADS-D: HADS depression subscale, Modified ZBI: modified Zarit Burden Interview (this modified, 24-item scale has a maximum score of 96).

The planned statistical comparisons between disease severity and caregiver burden were not possible due to the homogeneity of MSA severity as measured by UMSARS Part IV and Hoehn and Yahr stage.

PwMSA's self-rated HADS-D scores were correlated with caregivers' self-rated HADS-D scores (r = 0.73, p = 0.04) (Table 5). Caregiver-rated HADS-D for the PwMSA was also correlated with caregiver self-rated HADS-D score (r = 0.72, p = 0.013) (Table 5). There was no correlation between PwMSA participant and caregiver participant HADS-A (Table 5). However, HADS-A for caregiver participants did correlate with ZBI (r = 0.62, p = 0.041) (Table 6).

Table 4: Relationships between PwMSA quality of life scale scores and ZBI

PwMSA quality of life scale	n	Correlation with caregiver modified ZBI score (correlation coefficient, r)	p-Value
SF-36 PCS (PwMSA-rated)	9	−0.25	0.51
SF-36 MCS (PwMSA-rated)	9	−0.22	0.57
SF-36 PCS (caregiver-rated)	10	0.0016	1
SF-36 MCS (caregiver-rated)	10	−0.44	0.21
MSA-QoL motor subscale (PwMSA-rated)	9	0.58	0.1
MSA-QoL non-motor subscale (PwMSA-rated)	9	0.069	0.86
MSA-QoL social/emotional (PwMSA-rated)	9	0.16	0.67
MSA-QoL VAS (PwMSA-rated)	9	−0.32	0.41
MSA-QoL motor subscale (caregiver-rated)	11	0.48	0.13
MSA-QoL non-motor subscale (caregiver-rated)	11	0.39	0.24
MSA-QoL social/emotional (caregiver-rated)	11	0.44	0.17
MSA-QoL VAS (caregiver-rated)	11	−0.17	0.62

ZBI: Zarit Burden Interview, SF-36: 36-Item Short-Form Health Survey, PCS: physical component scale, MCS: mental component scale, MSA-QoL: Multiple System Atrophy health-related Quality of Life scale, VAS: visual analog scale.

Table 5: Relationships between PwMSA anxiety and depression symptoms and caregiver anxiety and depression symptoms

Comparison	n	Correlation coefficient (r)	p-Value
PwMSA HADS-A (PwMSA-rated) and caregiver HADS-A	8	0.04	0.92
PwMSA HADS-D (PwMSA-rated) and caregiver HADS-D	8	0.73	0.04*
PwMSA HADS-A (caregiver-rated) and caregiver HADS-A	11	0.26	0.44
PwMSA HADS-D (caregiver-rated) and caregiver HADS-D	11	0.72	0.013*

HADS: Hospital Anxiety and Depression Scale, HADS-A: HADS anxiety subscale, HADS-D: HADS depression subscale.
*Statistically significant result.

Table 6: Relationship between caregiver HADS scores and ZBI

HADS scale	n	Correlation with caregiver ZBI score (correlation coefficient, r)	p-Value
Caregiver HADS-A	11	0.62	0.041*
Caregiver HADS-D	11	0.52	0.099

HADS: Hospital Anxiety and Depression Scale, HADS-A: HADS anxiety subscale, HADS-D: HADS depression subscale.
*Statistically significant result.

Table 7: Types of tasks performed by informal caregivers

Types of tasks performed by caregivers	Examples
Activities of daily living	Assist with dressing, assist in the washroom, transfer PwMSA to/from chair or bed
Instrumental activities of daily living	Housekeeping, yard work, shopping, and finances
Transportation	Drive to/from appointments
Emotional and social support	Caregivers were often among the few people who took time to understand PwMSA with communication difficulties
Manager	Interview and hire self-managed care employees, keep a schedule of medical appointments and home care, advocate for home care enhancements, source and purchase medical equipment
Advocate	Advocate for referrals to health care services, advocate for additional care for patients in long-term care
Information manager	Keep track of patient's health information, research MSA, and communicate patient's health information to health professionals

Categories adapted from a framework of caregiving activities proposed in Health Quality Council of Alberta, 2019.[24]

Qualitative Analysis

Ten family caregivers and seven PwMSA were interviewed. Of the caregivers, eight were the PwMSA's spouse, one was the PwMSA's adult child, and one was the PwMSA's sibling (Table 2). The following five major themes were identified from qualitative analysis of the interviews: information, meaning of diagnosis, symptoms affect identity, supports and services, and caregiver experience. The theme of caregiver experience encompasses all aspects of caregivers' experience caring for the person with MSA and is reported on here. After completion of the qualitative analysis, we conducted a member check where participants provided feedback on the qualitative results, focusing on the caregiver experience theme. Four caregiver participants provided feedback. There was agreement with the major qualitative themes and the caregiver experience subthemes. Participants provided additional details within specific subthemes, which were incorporated in the results.

The Caregiver Role

Caregivers described the various activities and roles involved in providing care for the PwMSA. Predictably, the caregivers of patients with more symptoms performed more personal care tasks and found caregiving more demanding. Three caregivers were providing care to patients in long-term care facilities, and though their caregiver role continued, they had fewer and different responsibilities than the family caregivers of PwMSA who lived at home. Table 7 lists the types of care that caregiver participants described providing.

Caregiver Experience

The remaining subthemes of caregiver experience are listed with sample quotes in Table 8 and discussed below.

Keeping the Patient Safe

The physical safety of the person with MSA was the primary concern of caregivers in this study. To varying degrees, mobility issues prevented PwMSA from completing activities of daily living such as eating, dressing, and going to the washroom by themselves, often making it unsafe for them to remain at home alone. Falls were also a concern. Speech symptoms led one caregiver to worry that the PwMSA would not be able to call and communicate with emergency services if he had an emergency when alone. Patient safety concerns sometimes interfered with caregivers' own activities, as caregivers often worried about leaving the PwMSA alone to go to work, run errands, or participate in hobbies. In many cases, home care services were essential for the caregiver to feel comfortable leaving the house. Most participants found it easy to access home care, which is publicly funded in Canada. However, many expressed difficulties with home care including limits on hours and not having home care services available at night.

Even being in a different part of the home from the PwMSA was a concern for some caregivers. One caregiver carried a pager so his spouse could contact him throughout the home or yard. Safety concerns occurred when the PwMSA traveled outside the home, as well. One PwMSA was unable to attend day programs because of insufficient staffing. Their caregiver was required to attend the day program with them to assist with personal care tasks, negating the benefit of caregiver respite. One caregiver expressed concern with differing ways that taxi drivers secured the patient and her wheelchair in their vehicles. Another expressed concern about slippery flooring at a vacation rental. Safety concerns limited both PwMSA's and, by extension, caregivers', abilities to get out into the world.

Safety concerns did not end when the PwMSA was admitted to a long-term care facility. PwMSA were still at risk of falls, resulting in ongoing concerns for their families.

Caregiver's Own Health

Caregivers acknowledged the potential negative impacts on their own health due to the caregiving role. Lack of sleep, high stress, and the possibility of injuring oneself while assisting the PwMSA were often cited as risks to the caregiver's own health. Poorer diet and cutting back on exercise outside the home were also mentioned. Seeing the PwMSA suffer due to their illness was frequently cited as a contributor to stress.

Interviewees recognized the need to maintain their own health to continue caring for their loved one. Most family caregivers of PwMSA were the patient's spouse, and therefore of similar age; their own age-related medical conditions became concerns. In one case, a PwMSA's spouse experienced his own serious health issues, leading to their adult child taking over as the primary caregiver. This led to legal complexities in terms of documentation of alternate decision-makers and access to the PwMSA's medical information. The primary caregiver often had not given detailed information to other family members, making it hard for a new person to assume the complex role of organizing care and advocating for the PwMSA.

Table 8: Caregiver experience subthemes

Keeping the patient safe	"The biggest thing is we were able to get people to, for the home care, the people here so you know someone's with her so she's safe."-C2, caregiver "You just hook this thing onto the button and I have this pager. Now this pager, I can be outside and it's good for about 100 feet. So I can be outside and [patient] can hit that button and a chime will go off and that'll give me a signal, come in, that she needs some help with something[.]"-C7 "And they don't have the buttons, help me I've fallen. So when she falls, and she's in a private room, which is good, so when she falls she's at the mercy of somebody coming in finding her."-C3, discussing the long-term care facility where her sister lives
Caregiver's own health	"I'm not going to ruin myself doing this, because if I ruin myself, I can't help him."-C8 "If I lose [my wife], I'm dead in the water."-P11, MSA patient "It's very, very, very stressful on you and you don't want to burn out because if you burn out then somebody has to take care of you."-C7
MSA is progressive	"The disease is not fixable so that's a frustrating part about being a caregiver is when you see that despite all your best efforts, the person you're caring for is not making any improvement and in fact the disease is making it more difficult. It's hard to not become frustrated with it."-C1 "As her disease progresses it's more demand on me so yeah it is difficult at times yes."-C7 "It was very difficult. And I think that was recognized by my family, by the home care people, by the nurses and the doctors, and the team once he moved into the hospital. I wasn't even the one that came up with the idea of him going into care. It really never dawned on me that he was actually at that point."-C10
Relationship change in marriage	"We had a very good relationship before, we still have a good relationship but it's not like it was before. So I'm a caregiver, I'm not the spouse and husband and friend and confidant. I have to be, it's totally different and she is very dependent on me for everything."-C1 "[Y]ou're just moving on in life and your role hasn't changed, you're still the spouse. You're just doing different things."-C2 "Sometimes I feel like I'm the maid. But at other times I think we've gotten closer[.]"-C9
Communication difficulties cause frustration	"I get kind of angry sometimes because I figure she's not pushing herself more, and her speech 'cause sometimes I can't hear her so I'm asking her what, what, louder and then it feels like she always feels I'm yelling at her and sometimes I am I guess[.]"-C4 "So if I find that I ask him two things at once, it's like he's trying to process each one. So for my frustration and his ease of talking, I think I just have to practice harder asking one thing[.]"-C9
Motivation for caregiving	"Well I'm a very caring person. I have a philosophy that I treat people the way that I want to be treated. So as a caregiver I provide how I would expect to be treated. So from that perspective it's been a lot of work for me. But every day I could go to bed and know that I'd provided my wife with the best care that I possibly could give her without any guilt about anything."-C1 "If I hurt my back she would help me, so what's the difference?"-C2 "Oh I love it. He's such a good guy. No, I like to do things for him."-C8 "[The MSA diagnosis] kinda gave our family a little bit more peace of mind, okay we've done everything we could, most everything that's been suggested, this is likely why things haven't worked out to a better scenario[.]"-C10
Needs of caregivers	
On-demand help	"I said what I really need is someone I can just phone up and say can you come? But I don't think that exists. But [self-managed care] just didn't, I mean I don't want to form another company and I don't want to worry about all that other stuff unless you have to."-C9 "I can walk in there [home care] any time, and they'll help me if there's something they can do."-C11
Feeling supported	"I don't need much help. His nephew that's a little older than him and his wife, they've offered to give us help whenever we need, you know, to call on them but so far I haven't had to ask anybody else."-C8 "And if I want to go out or something, one of them will either come and get him. Usually my daughter because she's got two other kids, and they love having him. My son will take him, so really I have some avenues out."-C11 "The gamut is there. If we want to talk to a social worker or spiritual person or a doctor or nurse, whatever, we've got access so it's been very good."-C1 "[Long-term care staff are] thankful that we're there as family. But when we leave, they thank us for coming. They tell me to have a good evening you know. And that's important for a caregiver to get that kind of little pat on the shoulder."-C10 "I wish I had someone that would share, especially visiting times."-C3
Someone to talk to	"[My husband] is a good listener. He was in management so he knows how to deal with people so if I'm having people problems he helps me with that."-C3, sister of P3 "[T]here's still times where I need that someone to talk to, help me out to say how I feel. I don't say how I feel. Sometimes I say it in a manner that, again I'm yelling or I'm getting angry at her, but reality is I need time for myself. I also need time to get away, self-care."-C4

Caregiver participants were assigned participant identifiers beginning with "C," while PwMSA were assigned identifiers beginning with "P."

Caregivers experienced tension between providing the best possible care and preserving their own health. Some discussed a need to find a balance between doing their best for the PwMSA and avoiding burnout or physical injury. It was common for caregivers to feel guilt over making their own health a priority.

Because their focus is on the PwMSA, caregivers were rarely proactive in seeking help to prevent caregiver burnout. Health professionals frequently initiated discussions about respite services, mental health services, or considering long-term care for the patient.

Many caregivers discussed lifting the PwMSA or breaking falls, even early in the course of the illness. Caregivers did not consistently receive information from health care professionals on how to do these tasks safety.

MSA is Progressive

The relentless and relatively fast progression of MSA took an emotional and psychological toll on caregivers. Caregivers reported that despite their considerable efforts to help their loved one, MSA caused an inexorable worsening of physical, cognitive, and at times emotional status for the PwMSA. Though the information was difficult to hear, understanding the disease reassured one caregiver and her family they had done everything they could despite their relative's worsening health. The progressive nature of MSA was often accommodated by the caregiver until outside observers, typically the health care team, mentioned the need for home care or long-term care.

Relationship Changes in Marriage

Most caregivers were the patient's spouse. The care needs of the PwMSA resulted in a gradual shift in the marital relationship. As the PwMSA became more dependent, there was a change from feeling like the husband or wife to feeling like "just the caregiver." Due to mobility symptoms, couples were not able to maintain the hobbies and activities they previously shared. Physical intimacy became difficult over time due to pain and mobility symptoms. In some cases, admission of the PwMSA to hospital or hospice as their illness progressed allowed caregivers to take a step back from their caregiving role and regain some of their role as the spouse. One caregiver noted that it was hard to find information on relationship changes due to illness and caregiving, and that such information would have been helpful.

Communication Difficulties Cause Frustration

Most stressful and frustrating of all were communication difficulties. While both caregivers and PwMSA were frustrated by PwMSA's inability to communicate and be understood, inability to communicate also impacted the relationship between the two. Caregivers especially experienced frustration if they felt the patient was not trying hard enough to speak clearly and be understood.

Caregivers and PwMSA were ingenious in finding solutions to communication difficulties. These included technological solutions such as voice amplifiers and communication boards as well as strategies such as asking simple yes/no questions. Despite considerable efforts on the part of caregivers, these solutions became less successful over time as dysarthria and motor symptoms worsened.

Motivation for Caregiving

In this study, 8 out of the 10 caregivers interviewed were the PwMSA's spouses. Helping care for their spouse was a natural and gradual evolution, rather than something they one day volunteered to do. Though participants were not directly asked about what motivated them to provide care to their loved one, the subject often came up. For example, one spouse expressed annoyance at the term "caregiver," as helping one's husband or wife when they are sick is an inherent part of the relationship. Others felt the term "caregiver" did capture their role. In general, the most cited benefit of caring for the PwMSA was being able to spend more time with them. Caregivers were also motivated by a desire to protect or shelter the PwMSA, and wanting to know they had done everything possible for them. Some participants felt no one else would step up to help the PwMSA. The caregivers of the two patients in the study with the least disease progression expressed that caregiving was not burdensome.

Needs of Caregivers

The interviews with caregivers uncovered three categories of needs that were distinct from the needs of patients. Often, these needs were unmet or incompletely met.

On-demand Help

Real-time medical help was highly valued by those caregivers who had access to it. For example, those who reported being able to call a home care nurse or palliative home care nurse and receive immediate advice or services were very appreciative. Caregivers differed in their perceptions of what constituted timely access to medical advice. For example, one caregiver received a return call from the movement disorders clinic in 48 hours and felt that was too long to wait, while others were satisfied with a similar response time. In addition to the caregivers, several PwMSA also commented on the importance and helpfulness of receiving help from health care professionals in a timely fashion.

The variability of MSA day-to-day was sometimes a challenge when it came to home care, as the public home care system did not allow for a flexible schedule. PwMSA experienced bad days when more help was needed, but not available. In both conventional and self-managed home care (a program where families receive funding to hire their own home care workers), personal care workers had a set schedule. Finding help at night was a particular challenge.

Feeling Supported

Caregivers who felt supported by family, friends, or members of their religious community had a more positive outlook on their situation. Even when other family members such as adult children or siblings did not have a large role in their relative's care, the primary caregiver highly valued their support. Other family members often served as respite caregivers, a sounding board, or backup if extra help was needed. One interviewee anticipated her children would play a greater role in their father's care as his MSA progressed.

Acknowledgment was also valuable to caregivers. Two caregivers mentioned that they appreciated acknowledgment of the work they were doing by long-term care staff and family members.

Someone to Talk to

Caregivers often mentioned the benefits of having people to talk to about their experience caring for their relative. Family members, friends, coworkers, and MSA support groups filled this role. Concerns about not wanting to share negative information and not being understood were sometimes present. In one case, the PwMSA and caregiver, a married couple, saw a counselor together. The caregiver found this helpful to interpret the relationship challenges related to the stress of caregiving.

Other Qualitative Themes

Apart from the theme of caregiver experience, the other qualitative themes identified from interviews were information, meaning of diagnosis, symptoms affect identity, and supports and services. These themes were present in interviews with caregivers as well as PwMSA. Caregivers identified information as a major area where the health care system could support them. Both caregivers and PwMSA wanted more information early on about disease progression, while two caregivers acknowledged that too much information at diagnosis could be overwhelming. Symptom management was another area in which participants wanted more information. There were often discrepancies between PwMSA and their caregivers as to how much information each wanted, with no pattern as to whether the PwMSA or caregiver wanted more. The large majority of participants relied on the internet as well as health professionals for information on MSA.

Discussion

Our study used surveys and semi-structured interviews to characterize the population of MSA caregivers and understand many of the day-to-day challenges, concerns, and positive aspects that shaped their experiences. It also identified ways the health care system and clinicians can support MSA caregivers.

Caregiver burden has been a focus of the literature on caregiving[25] and was assessed in our study. Caregiver burden consists of objective caregiver burden (the tasks the caregiver is responsible for) and subjective caregiver burden (the emotional demands of caregiving).[25] In terms of objective burden, participants outlined the many activities required to support the person with MSA. These roles were myriad, including physical tasks (assisting with activities of daily living), administrative activities (such as scheduling appointments), emotional support, and advocating for the PwMSA within the health care system (Table 7). Caregiving activities performed by the MSA caregivers in our study were consistent with activities performed by caregivers in other conditions.[24] Since participants with MSA varied in their symptoms and the length in time since diagnosis, the objective burden of activities required to assist PwMSA varied from “not very much work” to a long list of tasks every day accounting for a significant amount of time. Subjective burden was assessed using a modified 24-item Zarit Burden Interview, and scores similarly ranged from mild burden to severe burden.

Through statistical analysis, we assessed whether caregiver burden was associated with other measured variables. Caregivers had mild anxiety on average, and caregiver anxiety correlated with caregiver burden. Caregivers of people with Parkinson’s disease (PD) have a similar association of higher anxiety with higher caregiver burden.[9,26,27] In PD caregivers, there is also an association between higher depression scores and higher caregiver burden that was not found among our caregiver participants.[9,26,27]

Patient quality of life is a factor that has been found to correlate with caregiver burden in studies in PD,[9,27] dementia,[28] and amyotrophic lateral sclerosis.[29,30] Our results did not find a link between the QoL of PwMSA and caregiver burden; however, our sample size was small and unlikely to find such an association. Disease severity and symptoms are other patient factors that have been associated with caregiver burden,[27,31] but quantitative measures of disease severity were too homogenous in our study to assess this.

Though caregivers in our study were on average not depressed, we found a correlation between patient and caregiver depression. This correlation has previously been reported in studies of caregivers of people with cancer[32] and patients in primary care clinics.[33] Whether the relationship between patient and caregiver depression symptoms is causal, and the direction of causation, cannot be determined by our results.

Qualitative results identified safety of the PwMSA as a primary concern of caregivers, similar to previous research in PD[34] and Parkinson-plus syndromes.[8] In our study, home care services were identified as essential to keeping PwMSA safe at home. Our participants had access to publicly funded home care based on their assessed needs without a time limit on the services. Caregivers were thankful for home care that allowed them to work outside the home, run errands, and participate in hobbies. However, despite access to publicly funded home care and health care navigation services, participants still encountered challenges with home care. These included limits to hours and the lack of flexibility of home care schedules to address day-to-day variations in care requirements. Similar difficulties were noted across the range of household incomes in our study. As a way to gain more control over their home care services, some PwMSA and families in our study opted into a publicly funded self-managed home care program.[35] In this model, patients are assessed and given a budget to work with. Hiring, training, and scheduling employees is then the responsibility of the patient or family.[35] While some participants preferred self-managed care for the added control it gave them over scheduling and hiring, others chose not participate in the program due to the management responsibilities required.

Expansion of publicly funded home care services would be helpful to PwMSA and their caregivers and would likely be helpful in other chronic health conditions. Home care is also beneficial from a health system perspective, as it is cost-effective compared to the alternatives of hospitalization or admission to a skilled nursing facility.[36] The limits of home care expansion that would still be cost-effective are not clear; however, in general, home care investment has not kept pace with the aging population.[36] Our study did not assess for a correlation between home care usage and caregiver burden or anxiety and depression symptoms. The effects of home care on caregiver burden and mood are inconsistent in the literature. One study from South Korea found no effect of home care usage on caregiver burden or satisfaction.[37] A study in Taiwan found home care buffered the effects of IADLs (instrumental activities of daily living), but not the effects of ADLs or care recipient cognitive impairment, on caregiver depression.[38] In both studies, underuse of home care services may have been a factor.[37,38]

Another important way the health care system supports MSA caregivers is by providing education on the illness as well as on caregiving skills. Caregivers in this study did not receive standardized education, and the information they received from health professionals varied. Standardized education programs for caregivers covering topics such as disease education, nursing skills, and psychosocial education show promise as an intervention for improving caregiver and patient outcomes.[39] A nurse-led education session for caregivers of people who had recently had a stroke was associated with reduced caregiver strain at time of discharge;[40] however, another study of in-hospital education for stroke caregivers found no significant difference in patient outcomes.[41] A hospital-based stroke education program that incorporated an in-home follow-up session resulted in improved caregiver and patient QoL and mood at 3 and 12 months as well as reduced health care costs.[42] In PD, a 9-week outpatient educational program for caregivers which incorporated psychoeducational sessions, when compared to an educational program without these sessions, showed improvements for both patients and caregivers in psychosocial adjustment and for caregivers in coping skills.[43] A similar 8-week program showed improvement in patient and caregiver mood and decreased psychosocial problems for caregivers, though there was a large variation in scores within intervention and control groups.[44]

Our caregiver participants recognized the importance of maintaining their own health to continue in their caregiving role. Caregiver health is particularly relevant when the care recipient still lives at home, and may influence admission to long-term care. In a survey of caregivers of patients with dementia in long-term care, 49% of family caregivers cited their own health as a factor in the decision to move their relative into long-term care.[45]

The health effects of providing care for a family member are somewhat of a paradox. Despite higher levels of stress and depressive symptoms, in several studies, caregivers have had reduced mortality rates compared to similar non-caregivers.[46,47] These findings have not been consistent, and the influence of duration and intensity of caregiving on mortality are not well studied.[47] One possible explanation for caregivers' reduced mortality is the "healthy caregiver hypothesis," which states that healthy individuals are more likely to become caregivers and continue in that role.[47] A competing hypothesis is that helping others provides a psychological protective effect against the negative health effects of stress.[46,47] Caregiver participants in our study did report psychological benefits from caregiving, including satisfaction from knowing they were doing everything possible for the PwMSA. Still, they reported risks to their own health due to their role, including risk of injury. Caregiving can be physically demanding, and a survey of Canadian caregivers found that 8% of spouse caregivers and 6% of those caring for their parents have sustained injuries from their caregiving work.[48] Another survey of caregivers who helped their family member with activities of daily living such as dressing, bathing, and transfers found 94% experienced musculoskeletal discomfort in the preceding 4 weeks, most commonly low back pain.[49] Transfers were the most physically demanding task.[49] Even in early stages of MSA, our caregiver participants described helping patients stand up and catching them from near falls. More advanced PwMSA required help to transfer in all situations. Caregivers were not necessarily trained to safely perform these tasks. Physiotherapist or occupational therapist education for all caregivers on transfers and falls would be helpful.[42]

Two factors which make MSA a uniquely challenging illness are its rapid progression and the prominence of symptoms affecting communication.[1,4,8] The rapid progression of MSA was a stressor for caregivers. In addition to the emotional toll from watching the PwMSA's symptoms worsen, some participants expressed grief or frustration over the relentless progression despite their best efforts to help. Worries about future disease progression were common and have previously been found to affect caregiver QoL in Alzheimer's disease.[28] Caregiving activities escalated as illness severity increased, which likely impacted caregivers' health. Transition to heavy caregiving, defined as helping with activities of daily living (ADLs) has been associated with increased depression symptoms and poorer health behaviors compared with non-caregivers and those who transition to moderate caregiving, defined as helping only with IADLs.[50]

Dysarthria is a nearly universal symptom of MSA and was present in 100% of PwMSA in one study.[51] Speech often becomes unintelligible even to family members. Since cognitive concerns occur late in MSA,[3] this leaves the PwMSA unable to make their wishes known or be "heard," yet still have much to say. For caregiver participants, unintelligible speech was a source of loss and frustration. Inability to discern the PwMSA's wishes may have led caregivers to doubt their own effectiveness and reduced their satisfaction with caregiving, as feelings of competence are associated with satisfaction with the caregiver role.[52] Technologies such as voice augmentation and communication boards were trialed by several participants but were often useful for a brief window of time due to worsening dysarthria and motor dysfunction. Communication difficulties, as well as the dependence of the PwMSA on the caregiver, caused profound changes in some participants' marital relationships. This finding is in contrast to a previous qualitative study in MSA and PSP, in which the quality of participants' marital relationships was unchanged despite communication symptoms.[8] Some caregivers in our study described a gradual shift in roles from being a spouse to being a caregiver. Participants were often unprepared for this shift, and information or counseling on how to maintain a positive relationship through changing roles may be helpful.[53]

As reflected in participants' comments, caregiving was a multifaceted experience – neither all negative nor all positive. Caregivers identified advantages to caregiving, most importantly being able to spend more time with their loved one. Several supportive factors were identified. Support from family was valued, even when other family members were not directly involved in caring for the person with MSA. Satisfaction with social support networks has previously been linked to a positive view of caregiving,[54] and social isolation in caregivers has been linked to caregiver burden and depression.[55] Caregivers who do not feel socially supported may particularly benefit from interventions such as respite care, support groups, or psychotherapy.[39] Aside from social support, supportive factors identified by caregivers in this study included being able to access health services in a timely fashion when needed and having someone to talk to about their experiences.

This study has several strengths as well as several limitations. Its mixed methods design allowed a fuller picture of caregivers' experiences than either quantitative or qualitative methods alone. However, the convenience sampling recruitment strategy meant that several demographics were absent from this study. All participating PwMSA had the parkinsonian subtype of MSA (MSA-P), except for one patient who had a mixed type. No patient had the cerebellar subtype of MSA (MSA-C). This reflects underlying prevalence: MSA-P is more common than MSA-C, and in Western countries, MSA-P accounts for up to 80% of MSA.[3] MSA-C is most common in Asian ethnicities[1,3] and progression of disability is slower than in MSA-P;[56] therefore, the experience of MSA-C caregivers might differ in areas such as objective caregiver burden and cultural values related to caregiving. Immigrant or refugee patients and caregivers were not represented in the study; they often face additional barriers to accessing health care and may lack a social network to rely on.[57] Rural caregivers were underrepresented, and studies have found they have more financial barriers[58] and lower self-rated health status[59] than urban caregivers. Because recruitment took place at a specialized movement disorders clinic, we did not include PwMSA without access to a specialized movement disorders clinic, who likely have higher unmet needs.

Our results suggest several areas where the health care system and individual clinicians can assist MSA caregivers. These include providing more information on MSA, ensuring training on caregiving skills such as transfers and how to assist a person who has fallen, providing information and counseling for spouse caregivers experiencing illness-related changes in their marriage, and expanding publicly funded home care services. Caregivers are an essential part of the health care team, and more can be done to support them in their important role.

Supplementary Material. To view supplementary material for this article, please visit https://doi.org/10.1017/cjn.2021.252

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