

Health-Related Quality of Life in Multiple System Atrophy using EQ-5D-5L: A Large Cross-Sectional Study in China

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Abstract

Background Multiple system atrophy (MSA) is a rare neurodegenerative disease, featuring autonomic failure plus parkinsonism and/or cerebellar ataxia. These symptoms impact the health-related quality of life (HRQoL) of MSA.

Objective We aimed to evaluate the HRQoL of MSA with a preference-based instrument, the five-level EuroQol five-dimensions questionnaire (EQ-5D-5L), for the first time.

Methods EQ-5D-5L was used to evaluate the HRQoL. The result of HRQoL was displayed as health utility index and visual analog scale (EQ-VAS) score. Specific scales were used to measure the disease severity, cognition, frontal lobe function, anxiety, depression, fatigue, and sleep disorders. The forward logistic model was used to explore the determinants of HRQoL in MSA.

Results A total of 205 patients with cerebellar variant (MSA-C, 53.9%) and 175 patients with parkinsonian variant (MSA-P, 46.1%) patients were included in the study. The mean scores of the health utility index and EQ-VAS were 0.558 and 59.5, respectively. Mobility was reported by the largest proportion (92.1%) of MSA patients, followed by usual activities (88.7%), self-care (81.3%), anxiety/depression (72.1%), and pain/discomfort (53.9%). The determinants of the lower health utility index in MSA were female sex, greater total Unified Multiple System Atrophy Rating Scale (UMSARS) scores, fatigue, and Parkinson's disease-related sleep problems (PD-SP). Lower EQ-VAS score was associated with greater total UMSARS scores, fatigue, PD-SP, and anxiety symptom. MSA-P patients reported more frequent problems in pain/discomfort than MSA-C patients, while MSA-C patients reported more problems in mobility than MSA-P patients.

Conclusion Patients with MSA had poor HRQoL evaluated by EQ-5D-5L. The most frequent affected problem is mobility in the Chinese MSA population. Besides the severity of MSA, fatigue, PD-SP and anxiety were determinants for poor HRQoL. Our research provides important information to improve the health status of patients with MSA.

Introduction

Multiple system atrophy (MSA) is a rare, rapidly progressive neurodegenerative disease featured with autonomic failure, parkinsonism, and/or cerebellar ataxia. To date, the etiology of MSA is still uncertain and no cure for MSA is available. MSA can be subclassified into a cerebellar variant (MSA-C) and a parkinsonian variant (MSA-P) according to the predominant motor symptom. [1] Except for experiencing severe disability in a short duration, many patients undergo various non-motor symptoms including urinary incontinence, dysarthria, depression, anxiety, sleep disorders, and so on. In addition, non-motor symptoms such as depression, anxiety, and autonomic deficit have a negative impact on the patients' lives and lead to a heavy disease burden. [2-5]

Patients with MSA were reported to have a poorer health-related quality of life (HRQoL) than healthy controls and patients with Parkinson's disease (PD). [5, 6] Compared to the common scale such as Parkinson's disease Questionnaire-39 item version (PDQ-39) which was used to measure the quality of life (QoL) of PD patients, the common symptoms of autonomic and cerebellar in MSA were not adequately reflected by PDQ-39. [7] The five-level EuroQol five-dimensions questionnaire (EQ-5D-5L), a preference-based instrument developed by EuroQol groups, is one of the most frequently used tools to evaluate HRQoL worldwide. [8] It can be used to estimate quality-adjusted life-years (QALYs), helping public health specialists and epidemiologists to plan interventions to improve patient's health status. The EQ-5D-5L was developed from the 3-level version of EQ-5D (EQ-5D-3L) and was proved to have some advantages over EQ-5D-3L. [9] However, there were only a few studies focused on the HRQoL of MSA by using EQ-5D-3L. [3-5, 10] These studies found that the most frequently affected dimensions in MSA were mobility, self-care, and usual activities. [3-5, 10] Besides the severity of the disease [4-6, 11], some studies found that depression was the determinant of HRQoL of MSA. [3, 5] No study on QoL of MSA evaluated by using EQ-5D-5L was available. Moreover, there was no study focused on the impact of non-motor symptoms measuring by comprehensive assessment scales on the QoL evaluated by using EQ-5D-5L in MSA.

Therefore, in the current study, we aimed to fill the gap to evaluate the HRQoL of MSA using EQ-5D-5L. Additionally, we used specific scales to measure the severity of non-motor symptoms including cognition, frontal lobe dysfunction, depression, anxiety, sleep disorders, and fatigue. We aimed to explore the determinants of HRQoL in MSA and the differences between MSA-C and MSA-P.

Method

Patients and Clinical features evaluation

Consecutive patients were recruited from the Department of Neurology, West China Hospital, Sichuan University from March 2018 to July 2021. All the patients reached the diagnostic criteria of probable MSA through comprehensive medical history reviewing and physical examination. [1] The exclusion criteria were: 1) magnetic resonance imaging scan, spinal cerebellar ataxia genetic tests (SCA1, 2, 3, 6, 7), or blood test indicating a diagnosis of other neurological diseases; 2) did not finish the whole interview because of dysarthria, weakness, or other reasons. This study was approved by the Ethics Committee of West China Hospital of Sichuan University and informed consent forms were signed by all subjects.

The clinical data were collected by experienced neurologists during a face-to-face interview. The demographic characteristics including age, sex, education, disease duration, and age of onset were collected. The disease severity was measured by the Unified Multiple System Atrophy Rating Scale (UMSARS). [12] The total UMSARS score was the sum of parts I and II. The orthostatic hypotension (OH) was defined as the drop of systolic blood pressure ≥ 30 mm Hg and/or diastolic blood pressure ≥ 15 mm Hg. The global cognitive function was evaluated using the Montreal Cognitive Assessment (MoCA). [13] The cognitive impairment was defined as MoCA < 19 for individuals with no more than 6 years of education, MoCA < 22 for individuals with 7 to 12 years of education, and MoCA < 24 for individuals with more than 12 years of education. [14] Frontal lobe dysfunction was defined as the frontal assessment battery (FAB) score < 16 . [15] The depressive and anxiety emotion were respectively defined as the Hamilton Depression Rating Scale (HAMD, 24 items) score ≥ 8 [16] and the Hamilton Anxiety Rating Scale (HAMA) score ≥ 6 . [17] Fatigue was defined as the mean score of Fatigue Severity Scale (FSS) ≥ 4 . [18] Parkinson's disease-related sleep problems (PD-SP) were defined as the Parkinson's Disease Sleep Scale 2nd version (PDSS-2) score ≥ 18 . [19] Excessive daytime sleepiness (EDS) was defined as the Epworth Sleepiness Scale (ESS) score ≥ 10 . [20] Rapid eye movement sleep behavioral disorder (RBD) was defined as the Rapid Eye Movement Sleep Behavior Disorder Screening Questionnaire (RBDSQ) score ≥ 5 . [21]

Assessment Of Hrqol

The HRQoL was evaluated using EQ-5D-5L. This scale was developed by the EuroQol Group and was used in evaluating healthy populations and patients with different diseases. [8] The EQ-5D-5L was designed for self-completion. It contains a questionnaire with five dimensions and a vertical visual analogue scale (EQ-VAS). The five dimensions are Mobility, Self-Care, Usual Activities, Pain/Discomfort, and Anxiety/Depression. Each dimension has five levels ranging from "no problems", "slight-moderate-severe" problems, to "unable to do". Descriptive levels of each dimension were dichotomized to "no problems"(level one) and "problems" (level two to five). The descriptive scores of the five dimensions were converted to health utility values according to the EQ-5D-5L value set for China. [22] The health utility values are anchored at 0 (death) and 1 (perfect health) and can be used to assess QALYs. The results of EQ-VAS were self-rated scores marked from 0 to 100, reflecting the patient's opinion on their overall health on the day of the interview.

Statistical analysis

First, the Kruskal-Wallis test was performed as the normality test. Mann-Whitney test was conducted to compare the health utility values and EQ-VAS scores of patients in different subgroups regarding sex, subtype, frontal lobe dysfunction, cognitive impairment, depressive emotion, anxiety emotion, EDS, RBD, PD-SP, and fatigue. T-test or Chi-square tests were conducted to compare demographic characteristics between MSA-C and MSA-P for continuous and categorical variables. Spearman's correlation analysis was conducted to explore relationships between EQ-5D-5L scores and clinical symptoms. Forward stepwise multiple logistic regression analysis was used to explore the potential determinants of HRQoL in MSA and two subtypes. Patients were divided into two groups according to the median of the health utility value and EQ-VAS score. The health utility value or EQ-VAS score was used as the dependent variable. The demographic and clinical variables were included in the model as independent variables. A level of $P < 0.05$ was set for a variable entered into the model and $P > 0.10$ for removal from the model. Statistically significant was considered as $P < 0.05$. All analyses were performed using SPSS 22.0 (SPSS, Inc., Chicago, IL, USA).

Results

Clinical characteristics of MSA and two subtypes

Finally, 205 MSA-C (53.9%) and 175 MSA-P (46.1%) were included in the study. The clinical characteristics of patients are displayed in Table 1. The average age and disease duration were 60.3 years old and 2.6 years respectively. There were 212 male patients (55.8%) and 168 females (44.2%). Patients with MSA-P had older age and age of onset and higher scores of PDSS-2 and ESS than patients with MSA-C. Patients with MSA-C had a higher proportion of OH and higher scores of RBDSQ than patients with MSA-P. There were no significant differences in other clinical features between the two subtypes.

Table 1
Demographic and clinical characteristics of patients with MSA and subtypes.

	Total (n=380)	MSA-C (n=205)	MSA-P (n=175)	P value
Sex (male)	212 (55.8%)	117 (57.1%)	95 (54.3%)	0.586
Age (years)	60.3 (9.3)	58.6 (9.2)	62.3 (9.0)	<0.001*
Age of onset (years)	57.7 (9.1)	56.1 (9.1)	59.5 (8.9)	<0.001*
Disease duration (years)	2.6 (1.5)	2.6 (1.4)	2.7 (1.6)	0.295
Education (years)	9.6 (3.7)	9.6 (3.6)	9.6 (3.8)	0.920
UMSARS-☒	16.6 (6.7)	16.8 (6.7)	16.4 (6.7)	0.536
UMSARS-☒	18.4 (6.9)	18.0 (6.7)	18.8 (7.2)	0.294
UMSARS-☒	2.2 (1.0)	2.2 (1.0)	2.2 (1.1)	0.981
UMSARS-total	35.1 (12.7)	34.9 (12.4)	35.2 (13.1)	0.806
FAB	14.4 (2.9)	14.5 (2.8)	14.3 (2.9)	0.613
MoCA	21.9 (5.0)	21.8 (4.8)	22.0 (5.3)	0.755
FSS	37.9 (19.3)	37.0 (19.8)	38.9 (18.7)	0.332
PDSS-2	11.6 (7.5)	10.4 (6.9)	13.0 (8.0)	0.001*
ESS	5.7 (4.9)	5.1 (4.6)	6.5 (5.1)	0.004*
RBDSQ	5.5 (3.6)	6.0 (3.7)	5.0 (3.4)	0.007*
HAMD	12.5 (8.1)	12.4 (8.0)	12.7 (8.3)	0.737
HAMA	10.2 (7.0)	9.9 (6.7)	10.6 (7.4)	0.354
OH	155 (40.8%)	94 (45.9%)	61 (34.9%)	0.030*
Continuous variables are displayed as mean scores and standard deviation. Dichotomous variables are displayed as numbers and percentages. T-test or Chi-square test was performed comparing characteristics between MSA-C and MSA-P. *Significant difference.				
Abbreviations: MSA: multiple system atrophy. MSA-P: MSA with predominately parkinsonism. MSA-C: MSA with predominately cerebellar ataxia. UMSARS: the Unified Multiple System Atrophy Rating Scale. FAB: the frontal assessment battery. MoCA: the Montreal Cognitive Assessment. FSS: the Fatigue Severity Scale. PDSS-2: the Parkinson's Disease Sleep Scale 2nd version. ESS: the Epworth Sleepiness Scale. RBDSQ: Rapid Eye Movement Sleep Behavior Disorder Screening Questionnaire. HAMD: the Hamilton Depression Rating Scale. HAMA: the Hamilton Anxiety Rating Scale. OH: orthostatic hypotension.				

Eq-5d-5l In Msa And Different Subgroups

The mean EQ-VAS score for MSA was 59.5 (SD: 17.81) and the mean health utility value was 0.558 (SD: 0.276). The health utility values and EQ-VAS scores of different subgroups are displayed in Table 2. Female patients and subjects with frontal lobe dysfunction, cognitive impairment, depression, anxiety, EDS, PD-SP, and fatigue had lower health utility values. Female patients and patients with OH, depression, anxiety, PD-SP, and fatigue had lower EQ-VAS scores. No significant differences in the health utility

values and EQ-VAS were observed between patients with MSA-C and MSA-P. The frequency of five levels in five dimensions of EQ-5D-5L in total MSA patients and two subtypes of patients are shown in Figure 1. For each dimension, mobility was reported by 92.1% of MSA patients, followed by usual activities (88.7%), self-care (81.3%), anxiety/depression (72.1%), and pain/discomfort (53.9%). The frequencies of reported problems of each dimension in MSA-C and MSA-P are displayed in Figure 2. The most common problem was mobility in both MSA-C and MSA-P patients, followed by usual activities, self-care, anxiety/depression, and pain/discomfort. The frequency of reported problems of pain/discomfort in patients with MSA-P was significantly higher than that in patients with MSA-C while the frequency of reported problems of mobility in patients with MSA-P was significantly lower than that in patients with MSA-C.

Table 2
Health utility index and EQ-VAS scores regarding the clinical features in MSA.

	Groups	Number	Health utility value	P value	EQ-VAS score	P value
Sex	Male	212	0.687 (0.485-0.792)	<0.001*	60 (50-70)	0.014*
	female	168	0.513 (0.281-0.734)		60 (50-70)	
Subtypes	MSA-C	205	0.606 (0.363-0.760)	0.498	60 (50-70)	0.148
	MSA-P	175	0.638 (0.363-0.792)		60 (50-70)	
OH	No	225	0.638 (0.400-0.783)	0.118	60 (50-70)	0.045*
	Yes	155	0.571 (0.347-0.778)		60 (50-70)	
FLD	No	164	0.691 (0.446-0.792)	<0.001*	60 (50-70)	0.051
	Yes	216	0.566 (0.290-0.734)		60 (50-70)	
CI	No	213	0.691 (0.444-0.792)	<0.001*	60 (50-70)	0.112
	Yes	167	0.513 (0.208-0.724)		60 (50-70)	
Depressive mood	No	130	0.749 (0.595-0.841)	<0.001*	70 (58-75-80)	<0.001*
	Yes	250	0.511 (0.259-0.723)		60 (50-70)	
Anxiety mood	No	109	0.748 (0.592-0.841)	<0.001*	70 (60-80)	<0.001*
	Yes	271	0.513 (0.296-0.734)		60 (50-70)	
EDS	No	300	0.638 (0.375-0.792)	0.021*	60 (50-70)	0.327
	Yes	80	0.513 (0.245-0.734)		60 (50-70)	
RBD	No	171	0.642 (0.438-0.783)	0.089	60 (50-70)	0.318
	Yes	209	0.513 (0.281-0.734)		60 (50-70)	
PD-SP	No	304	0.669 (0.441-0.792)	<0.001*	60 (50-70)	<0.001*
	Yes	76	0.431 (0.134-0.573)		60 (50-70)	
Fatigue	No	152	0.734 (0.521-0.841)	<0.001*	60 (50-80)	<0.001*
	Yes	228	0.513 (0.301-0.724)		60 (50-70)	

Health utility scores and EQ-VAS scores are displayed as the median and interquartile range (IQR). Higher health utility scores and EQ-VAS scores indicate a better health-related quality of life. Mann-Whitney test was used to compare the health utility scores and EQ-VAS scores between two levels of each subgroup. *Significant difference.

Abbreviations: MSA: multiple system atrophy. MSA-P: MSA with predominantly parkinsonism. MSA-C: MSA with predominantly cerebellar ataxia. EQ-VAS: visual analog scale. OH: orthostatic hypotension. EDS: Excessive daytime sleepiness. RBD: Rapid eye movement sleep behavioral disorder. PD-SP: Parkinson's disease-related sleep problems. FLD: frontal lobe dysfunction. CI: cognitive impairment.

Related factors of EQ-5D-5L scores in total MSA patients and patients with two subtypes

Spearman's analysis showed a moderate correlation between the EQ-VAS score and scores of total UMSARS, FSS, PDSS-2, HAMD, and HAMA in total MSA patients. The total UMSARS score was strongly correlated with health utility value. Scores for FAB, MoCA, FSS, PDSS-2, HAMD, and HAMA had weak to moderate correlations with health utility value in total MSA patients. (Table 3)

Table 3
Spearman correlation between EQ-VAS score, health utility index and clinical characteristics of MSA.

	EQ-VAS score (r)	P value	Health utility value (r)	P value
UMSARS-total	-0.379	<0.001*	-0.717	<0.001*
FAB	0.144	0.005*	0.246	<0.001*
MoCA	0.154	0.003*	0.289	<0.001*
FSS	-0.253	<0.001*	-0.380	<0.001*
PDSS-2	-0.304	<0.001*	-0.356	<0.001*
ESS	-0.029	0.579	-0.101	0.049*
RBDSQ	-0.075	0.142	-0.110	0.031*
HAMD	-0.380	<0.001*	-0.564	<0.001*
HAMA	-0.359	<0.001*	-0.479	<0.001*

The correlation coefficient (r) ≥ 0.8 is defined as a very strong correlation; 0.60–0.79, a strong correlation; 0.4–0.59, a moderate correlation; 0.20–0.39, a weak correlation; and $r \leq 0.19$, a negligible correlation. *Significant difference.

Abbreviations: MSA: multiple system atrophy. EQ-VAS: visual analog scale. UMSARS: the Unified Multiple System Atrophy Rating Scale. FAB: the frontal assessment battery. MoCA: the Montreal Cognitive Assessment. FSS: the Fatigue Severity Scale. PDSS-2: the Parkinson's Disease Sleep Scale 2nd version. ESS: the Epworth Sleepiness Scale. RBDSQ: Rapid Eye Movement Sleep Behavior Disorder Screening Questionnaire. HAMD: the Hamilton Depression Rating Scale. HAMA: the Hamilton Anxiety Rating Scale.

Multivariate logistic models showed the determinants of health utility value and EQ-VAS score in total MSA and two subtypes of patients. (Table 4) In the total MSA patients, female sex, fatigue, PD-SP, and greater total UMSARS score were the determinants for the lower health utility values. Fatigue, PD-SP, anxiety emotion, and greater total UMSARS score were the determinants for the lower EQ-VAS scores. There were some differences in the determinants for health utility value and EQ-VAS score between patients with MSA-C and MSA-P. For example, cognitive impairment was the determinant for lower health utility value in patients with MSA-P but not in patients with MSA-C. And frontal lobe dysfunction was the determinant for lower EQ-VAS score in patients with MSA-P but not in those with MSA-C. More details are displayed in Table 4.

Table 4
Multivariate logistic regression analysis of healthy utility index and EQ-VAS score for MSA and two subtypes.

MSA			MSA-C			MSA-P						
Health utility value	EQ-VAS score	Health utility value	EQ-VAS score	Health utility value	EQ-VAS score	OR (95% CI)	P value	OR (95% CI)	P value	OR (95% CI)	P value	
UMSARS-total	1.18 (1.14-1.22)	<0.001	1.04 (1.02-1.06)	<0.001	1.17 (1.12-1.23)	<0.001	1.04 (1.01-1.07)	0.003	1.20 (1.13-1.27)	<0.001	1.04 (1.01-1.07)	0.007
Fatigue	2.30 (1.28-4.09)	0.005	1.64 (1.02-2.65)	0.042	2.83 (1.13-5.96)	0.006				2.20 (1.01-4.77)	0.047	
PD-SP	2.78 (1.30-5.97)	0.009	1.86 (1.06-3.26)	0.031			2.91 (1.16-7.31)	0.023	4.42 (1.53-12.82)	0.006		
Anxiety mood			2.23 (1.29-3.88)	0.004						4.65 (1.76-12.31)	0.002	
Depressive mood						1.97 (1.02-3.80)	0.045					
CI								2.69 (1.12-6.46)	0.027			
FLD									2.33 (1.09-4.98)	0.030		
Sex	0.35 (0.20-0.62)	<0.001		0.26 (0.12-0.56)	0.001	0.49 (0.27-0.91)	0.023					

Variables that are statistically significant in the model are shown in the table.

Abbreviations: MSA: multiple system atrophy. EQ-VAS: visual analog scale. MSA-P: MSA with predominately parkinsonism. MSA-C: MSA with predominately cerebellar ataxia. OR: odds ratio. 95%CI: 95% confidence interval. UMSARS: the Unified Multiple System Atrophy Rating Scale. PD-SP: Parkinson's disease-related sleep problems. FLD: frontal lobe dysfunction. CI: cognitive impairment.

Discussion

Our research measured the HRQoL of MSA using EQ-5D-5L, providing important data on the QoL of Chinese MSA patients. We found that patients with MSA in a Chinese population had poor HRQoL. Mobility was the most commonly reported problem and pain/discomfort was the least common problem, which was consistent with the previous study using EQ-5D-3L. [4] Besides disease severity, sex, fatigue, and PD-SP were found to be the determinants for health utility value of MSA, and anxiety symptom, fatigue, and PD-SP were the determinants for EQ-VAS of MSA. Additionally, we found there were some differences in the determinants for HRQoL of MSA between MSA-C and MSA-P patients.

The mean health utility index (0.558) and the mean EQ-VAS score (59.5) of patients with MSA were lower than those of the Chinese healthy population (0.943, 82.9) and those of patients with coronary heart disease and stroke history (0.850, 61.0). [23, 24] The mean EQ-VAS score of Chinese patients with MSA (59.5) was higher than the results of the European study (44.5 and 36.9). [4, 5] This may be explained by the shorter disease duration and mild disease severity of patients enrolled in our study. Consistent with

the previous study, we also found that the total UMSARS score was the determinant of health utility value and VAS scores in the MSA and two subtypes. [4]

We found that mobility was the most commonly reported problem and pain/discomfort was the least common problem, which was consistent with previous studies. [4, 5] Furthermore, we found that a higher frequency of problem of mobility was reported in patients with MSA-C than patients with MSA-P, which was consistent with the previous study assessed by EQ-5D-3L. [4] However, we found there was no difference in the mean scores of motor examination and global disability scale of UMSARS between the two subtypes. Our finding suggested that UMSARS has limited ability to reflect mobility in patients with MSA-C. In addition, we found that patients with MSA-P reported more pain/discomfort than patients with MSA-C, which was consistent with the previous study assessed with EQ-5D-3L. [5] This may be due to the severer damage in the basal ganglia in MSA-P than in MSA-C. [25]

Depression was reported to be a determinant of the QoL of MSA.[3, 4, 26, 27] In our study, the depressive emotion was related to the EQ-VAS score of MSA-C. We also found that anxiety played an important role in the decreasing EQ-VAS score of MSA. [3] Attempts to treat these symptoms are important to improve the HRQoL of patients with MSA. Similar to the result of the previous study, we found that female sex was a negative determinant of health utility values of MSA. [4] Additionally, females were more likely to initially manifest motor symptoms and recurrent falls within three years. [28] More researches should be made to understand the gender difference in MSA.

Fatigue is common in several neurological diseases including PD and multiple sclerosis. [29] There were 29–82% of MSA patients who suffered from fatigue. [30], which was significantly higher than that of patients with PD and healthy control. [31] In the current study, we found that fatigue was negatively correlated with health utility value and EQ-VAS score of MSA, which was consistent with our previous study evaluating QoL of MSA patients using PDQ-39. [11] The pathological mechanisms of fatigue are still uncertain. A study found that fatigue in MSA was related to the decrease of the 5-hydroxytryptamine receptor in the raphe nuclei and brain stem. [31] We should pay more attention on fatigue when managing patients with MSA.

In our previous study, sleep-related disorders including PD-SP, RBD, and EDS were found to be associated with higher disease severity in MSA. [32] In the current study, PD-SP but not RBD and EDS was found to be a determinant of the health utility value and EQ-VAS score of MSA. Since PD-SP includes motor symptoms at night, PD symptoms at night, and disturbed sleep, the etiology of PD-SP in MSA is complex and is still under research. Management of PD-SP in MSA is necessary.

Recent studies found that a varying degree of cognitive impairment was common in MSA assessed by comprehensive neuropsychological testing. [33, 34] Our current study found that cognitive impairment was the determinant of health utility value in MSA-P but not in MSA-C. [35, 36] Kawai reported that patients with MSA-P had wider and severer cognitive impairment than patients with MSA-C. [35] Patients with MSA-P were reported to have lower MoCA scores than patients with MSA-C in a Chinese study. [37] Additionally, we found that frontal lobe dysfunction was the determinant of EQ-VAS score in MSA-P but not in MSA-C, which was similar to our previous finding that frontal lobe dysfunction was the determinant of HRQoL assessed by PDQ-39 in MSA. [26] Further study should pay attention to the cognitive function in patients with MSA.

This research had several limitations. First, this cross-sectional research explored the features and determinants of the EQ-5D-5L score in MSA. Further study should concentrate on the longitudinal change of the EQ-5D-5L scores of MSA. Second, the diagnosis of MSA was based on the clinical features but not the autopsy. Further study should build autopsy confirmed cohort to study the HRQoL of MSA.

Conclusion

The current research found that MSA patients had poor HRQoL and EQ-5D-5L was a validating tool to evaluate HRQoL in MSA. The determinants of lower health utility value were female sex, greater scores of total UMSARS, fatigue, and PD-SP while that of lower EQ-VAS scores were greater scores of total UMSARS, fatigue, PD-SP, and anxiety symptom. In addition, the determinants of EQ-5D-5L were different between patients with MSA-C and MSA-P. This research provided important information on the HRQoL and potential determinants of MSA, helping developing intervention strategies to improve their health status.

Declarations

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Author Contribution

Yi Xiao: Organization and execution of research project, design, and execution of statistical analysis, writing of the draft, review, and critique of the manuscript; Lingyu Zhang: Organization and execution of research project, design, and execution of statistical analysis, review, and critique of the manuscript; Qianqian Wei: Organization and execution of research project, design the statistical analysis; Tianmi Yang, Kuncheng Liu, Ruwei Ou, Yanbing Hou and Junyu Lin: Organization and Execution of research project; Huifang Shang: Conception, organization and execution of research project, review and critique of the manuscript.

Ethics approval and consent to participate

This study was approved by the institutional ethics committee of West China Hospital and informed consents were obtained from all the participants of the study.

Consent for publication

Not applicable.

Availability of data and materials

The data used in the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Figures

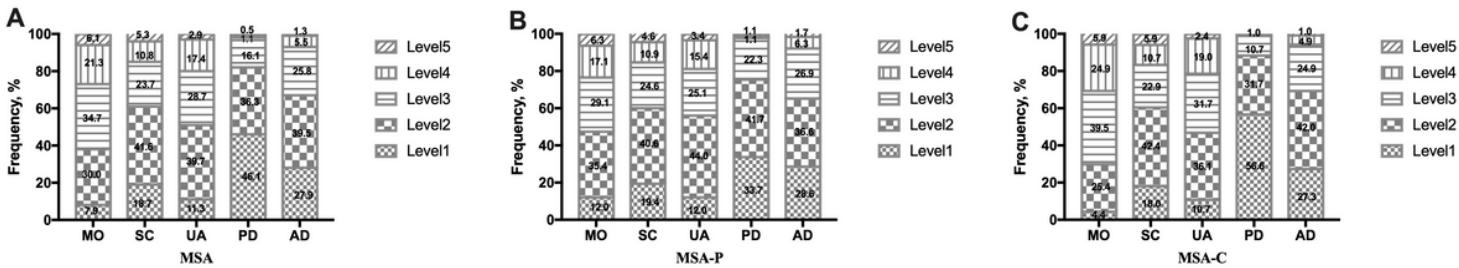


Figure 1

Distribution of 5 levels in each dimension of EQ-5D-5L. Figure A: EQ-5D-5L of total MSA; Figure B: EQ-5D-5L of patients with MSA-P; Figure C: EQ-5D-5L of patients with MSA-C. Level 1: no problems, level 2: mild problems, level 3: moderate problems, level 4: severe problems, level 5: extreme problems. Abbreviations: EQ-5D-5L: the five-level EuroQol-5 dimensions. MSA: multiple system atrophy. MSA-P: MSA with predominately parkinsonism. MSA-C: MSA with predominately cerebellar ataxia. MO: Mobility, SC: Self-care, UA: Usual activities, PD: Pain/discomfort, AD: Anxiety/depression.

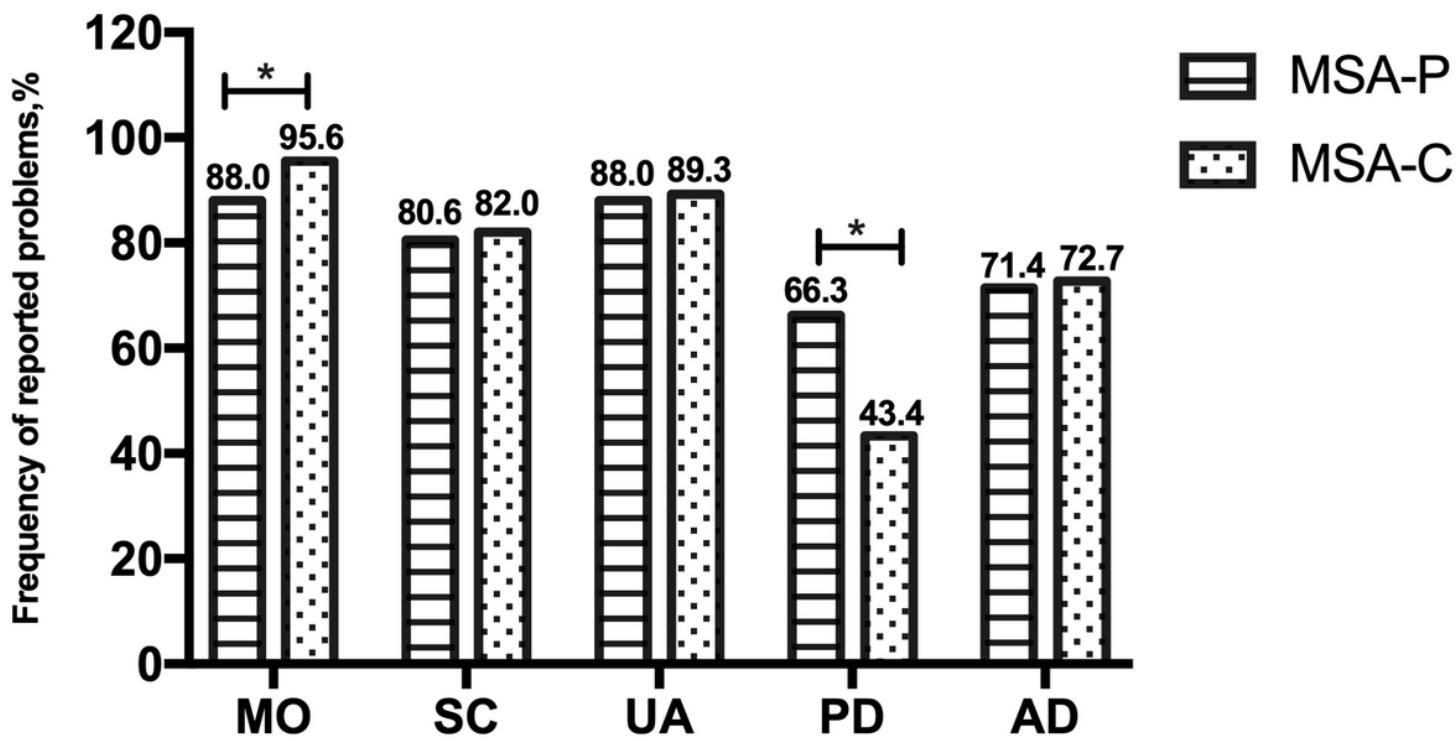


Figure 2

Frequency of reported problems for each dimension of EQ-5D-5L between MSA-P and MSA-C patients. *Significant difference. Abbreviations: EQ-5D-5L: the five-level EuroQol-5 dimensions. MSA: multiple system atrophy. MSA-P: MSA with predominately parkinsonism. MSA-C: MSA with predominately cerebellar ataxia. MO: Mobility, SC: Self-care, UA: Usual activities, PD: Pain/discomfort, AD: Anxiety/depression.