Humanistic and Economic Burden of Multiple System Atrophy: A Systematic Review

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INTRODUCTION

- Multiple system atrophy (MSA) is a rare neurodegenerative disorder characterized by a combination of parkinsonism, autonomic dysfunction, and cerebellar ataxia with MSA-P (parkinsonian type), and MSA-C (cerebellar type) as subtypes
- Prevalence of MSA ranges from 3 to 5 cases per 100,000 individuals
- Currently, there is no cure for MSA, and treatment options are largely supportive and symptomatic

OBJECTIVE

- To evaluate the overall humanistic and economic burden associated with MSA and identify any existing gaps in the current evidence

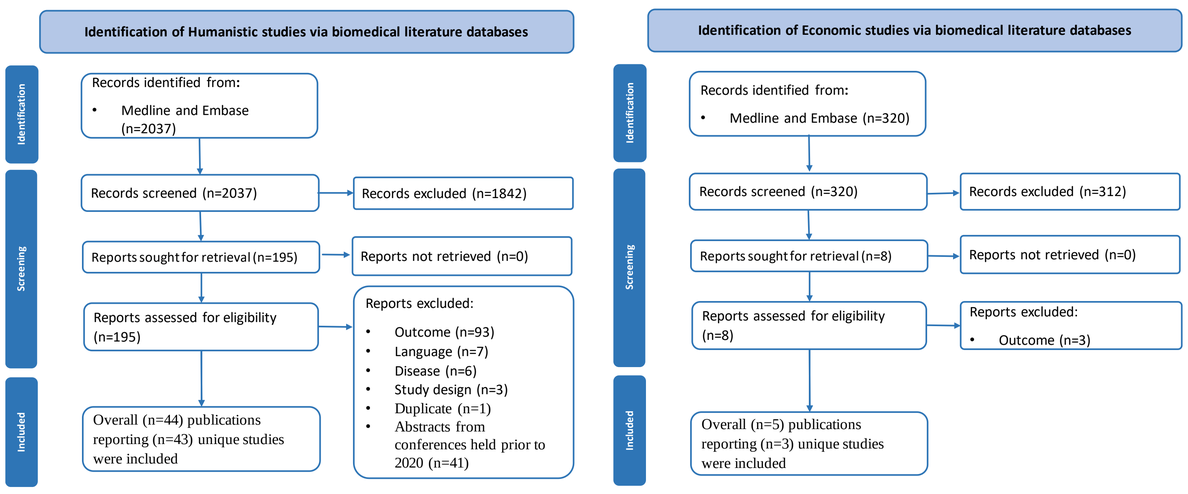
METHODS

- **Key eligibility criteria:** Studies published in English between 01/2013-06/2023 reporting the humanistic and economic burden of MSA
- **Literature databases searched:** Embase® and MEDLINE
- **Study screening process:** Studies were assessed for inclusion/exclusion by two independent reviewers based on a pre-specified criteria. Any disagreement was resolved by a third reviewer
- **Data extraction:** Extracted from the relevant studies in a pre-designed template

RESULTS

- Figure 1 presents detailed PRISMA diagrams depicting finalisation of 46 unique studies (humanistic, n=43; economic, n=3) out of a total of 49 included publications

Figure 1: PRISMA diagrams for economic and humanistic burden reviews



- Majority of the humanistic burden studies were either prospective or cross-sectional in nature (20 each; Figure 2) and were majorly conducted in China (n=17) and Europe (n=11) (Figure 3)

Figure 2: Study design- Humanistic burden

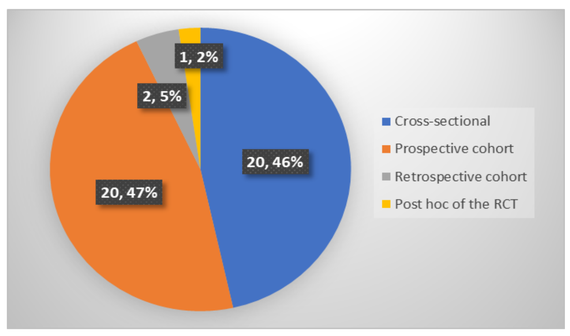
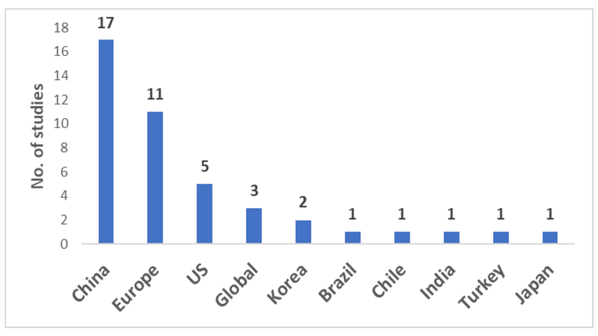


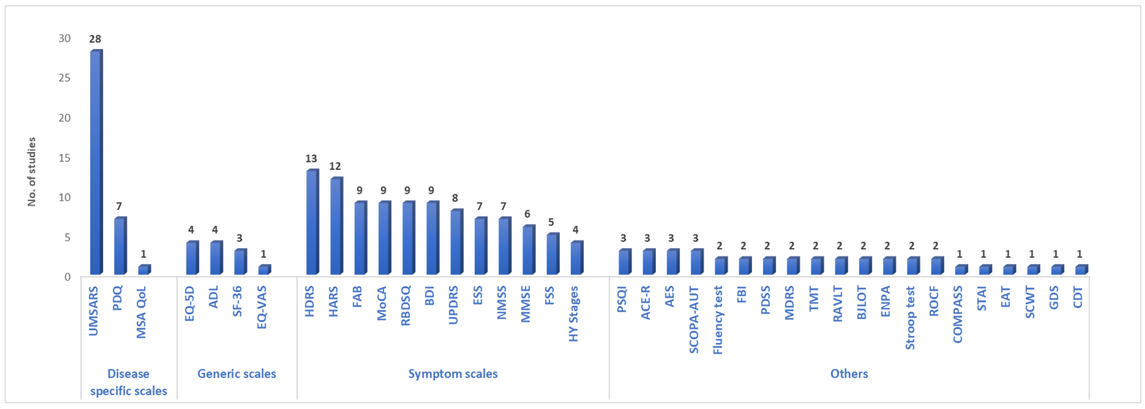
Figure 3: Study country- Humanistic burden



- In contrast, economic burden studies were retrospective (n=2) and cross-sectional (n=1); conducted in Canada, Germany, and the US (one each)
- Sample size across studies ranged from 7 to 483 and 47 to 3619 for humanistic and economic burden, respectively

- Out of a total of 39 different generic and MSA-specific scales used across studies to measure HRQoL, Unified Multiple System Atrophy Rating Scale (UMSARS) (n=28), PD specific health status questionnaire (PDQ) (n=7) and Multiple System Atrophy Quality of Life Questionnaire (MSA-QoL) (n=1) were disease-specific scales. Symptom scales that were referred to in ≤3 studies were grouped as 'Others' (Figure 4)

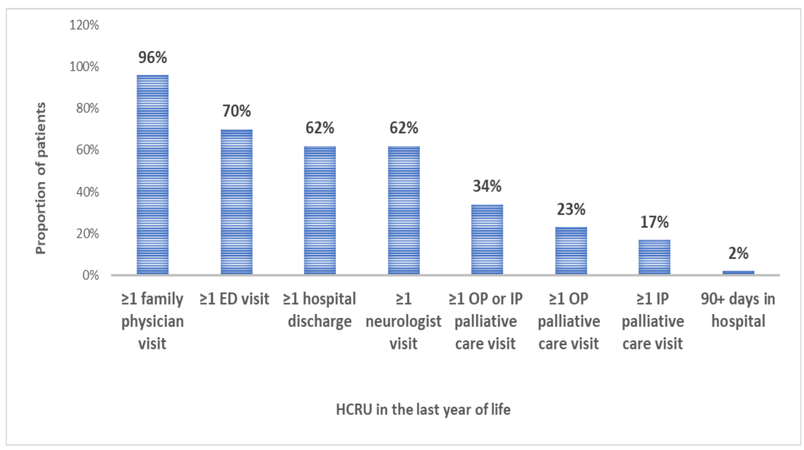
Figure 4: Type of HRQoL scales



Abbreviations: ACE-R, Addenbrooke's Cognitive Examination; ADL, Activities of daily living; AES, Apathy Evaluation Scale; BDI, Beck Depression Inventory; BJLOT, Benton Judgment of Line Orientation Test; CDT, Clock Drawing Test; COMPASS, Composite Autonomic Symptoms Scale; EAT, Emotion Attribution Task; ENPA, Esame Neuropsicologico per l'Afasia; EQ-5D, EuroQol-5 Dimension; EQ-VAS, EuroQol-visual analogue scales; ESS, Epworth sleepiness scale; FAB, Frontal assessment battery; FBI, Frontal Behavioral Inventory; FSS, Fatigue Severity Scale; GDS, Geriatric Depression Scale; HARS, Hamilton Anxiety Rating Scale; HDRS, Hamilton Depression Rating Scale; HY Stages, Hoehn and Yahr Scale; MDRS, Montgomery-Asberg Depression Rating Scale; MMSE, Mini-Mental State Examination; MoCA, Montreal Cognitive Assessment; MSA QoL, Multiple System Atrophy Quality of Life Questionnaire; NMSS, Non-Motor Symptoms Scale; PDQ, PD specific health status questionnaire; PDSS, Parkinson's Disease Sleep Scale; PSQI, Pittsburgh Sleep Quality Index; RAVLT, Rey Auditory Verbal Learning Test; RBDSQ, Rapid Eye Movement Sleep Behavior Disorder Screening Questionnaire; ROCF, Rey-Osterrieth complex figure test; SCOPA-AUT, Scales for Outcomes in Parkinson's Disease - Autonomic Dysfunction; SCWT, Stroop Color and Word Test; SF-36, 36-Item Short Form Survey; STAI, State-Trait Anxiety Inventory; TMT, Trail Making Test; UMSARS, Unified Multiple System Atrophy Rating Scale; UPDRS, Unified Parkinson's Disease Rating Scale

- Studies indicated that in both PD and MSA groups, significant impairments were observed in Frontal assessment battery (FAB), Addenbrooke's Cognitive Examination (ACE-R) total, memory, verbal fluency, speaking, and PDQ scores compared to the control group ($p < 0.05$). Furthermore, in the MSA group, all tests, except for memory, exhibited greater impairment compared to the PD group
- The studies suggest that MSA-P (parkinsonian subtype) individuals experience more severe motor impairment, hyposmia (reduced sense of smell), depression, anxiety, cognitive impairment, and lower HRQoL compared to those with MSA-C (cerebellar subtype)
- One study indicated that non-motor symptoms are more prevalent and severe in MSA-P patients, particularly in terms of mood/apathy and gastrointestinal symptoms
- An EQ-5D-5L index utility value of 0.558 has been reported in a study. It was observed that female patients and patients experiencing frontal lobe dysfunction, cognitive impairment, depression, anxiety, excessive daytime sleepiness, PD-SP, and fatigue had lower EQ-5D-5L index values

Figure 5: HCRU in the last year of life



- As per the resource utilisation reported in a study, most participants, 96% and 70.2%, had >1 family physician and emergency department visits, respectively (Figure 5)
- Additionally, 62% of individuals had been hospitalised multiple times, and 25.5% spent more than 30 days in the hospital

CONCLUSION

- These findings highlight the considerable humanistic and economic challenges faced by individuals living with MSA
- Further research is needed to better quantify the economic burden associated with MSA

FUNDING

None