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A Targeted Literature Review of Health State Utility Values in Neuromyelitis Optica Spectrum Disorder

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Introduction and objectives

- Neuromyelitis optica spectrum disorder (NMOSD) is a rare autoimmune disease that effects mainly the optic nerves and spinal cord.[1]
- In 2005, NMOSD characterized as new disease entity distinct from multiple sclerosis. Global mortality rates range from 3.3 to 32% in untreated, relapsed or respiratory NMOSD patients.[2,3]

Due to its relapsing and debilitating nature, NMOSD has a negative impact on patient's quality of life (QoL) and impose significant burden on patients QoL.[4]

- Understanding health utility values (HUVs) while living with this severe disease is important for health technology evaluations and was the focus of this review.

Methodology

- A literature search was conducted in Embase® and MEDLINE® via Ovid by using combination of Emtree/ MeSH terms and keywords to identify English language articles published from database inception to 8th June 2022.
- All the identified studies were screened based on the title/abstracts and followed by full-texts screening against the eligibility criteria (**Table 1**) and data extraction by one reviewer.
- The quality assessment of included studies was evaluated using Cochrane risk of bias tool and Newcastle-Ottawa Scale (NOS) for randomized controlled trials (RCTs) and observational studies, respectively. NOS score ranges from 0-9; score 7-9, 6-5, ≤4 graded as high, moderate and low quality, respectively.
- A trial was considered as high quality if it has low risk of bias in all domains

Results

❖Study Selection:

- After removing duplicates, 819 records were obtained through databases and additional searches.
- A total of 8 studies (9 publications) were included, of which 3 were multicounty studies wherein other studies were conducted each in USA, UK, Germany, Thailand and South Korea (**Figure 1**).
- Out of 8 studies, 4 were cross-sectional studies and 3 randomized controlled trials and remaining 1 prospective longitudinal observational study.

❖Quality Assessment:

- Out of 8 studies, 7 studies marked as high-quality [4 observational (NOS score: 7-8) and 3 RCTs]. Remaining one cross-sectional study marked as low quality (NOS score: 4) due to insufficient information in conference abstract about ascertainment of exposure, non-responder and statistical analysis.

❖Reported Utility Values for NMOSD:

- All studies derived the HUVs using EQ-5D and mean HUVs at baseline was ranged from 0.41 (Thailand) to 0.74 (USA) and median utility value was 0.82 in South Korea.[5,6,7]
- In UK, the mean HUVs at baseline was 0.54 and reported stable HUVs during the follow-up as 0.56, 0.56 and 0.59 at 6, 9 and 12 months, respectively.[8]
- Three studies reported a significant reductions in HUVs with increase in disease severity measured using Expanded Disability Status Scale (EDSS) score[5,8,9](**Supplementary Table 1**).

Conclusions

The NMOSD patients has worse HUVs compared to healthy controls.

HUVs were reduced significantly with increase (severe disease) in EDSS score and being relapsed.

Even though few studies reported HUVs data, these were of high quality.

HUVs from this review will aid in conducting economic-evaluations for decision-making and identifying subgroups having worse HUVs in NMOSD patients.

References:
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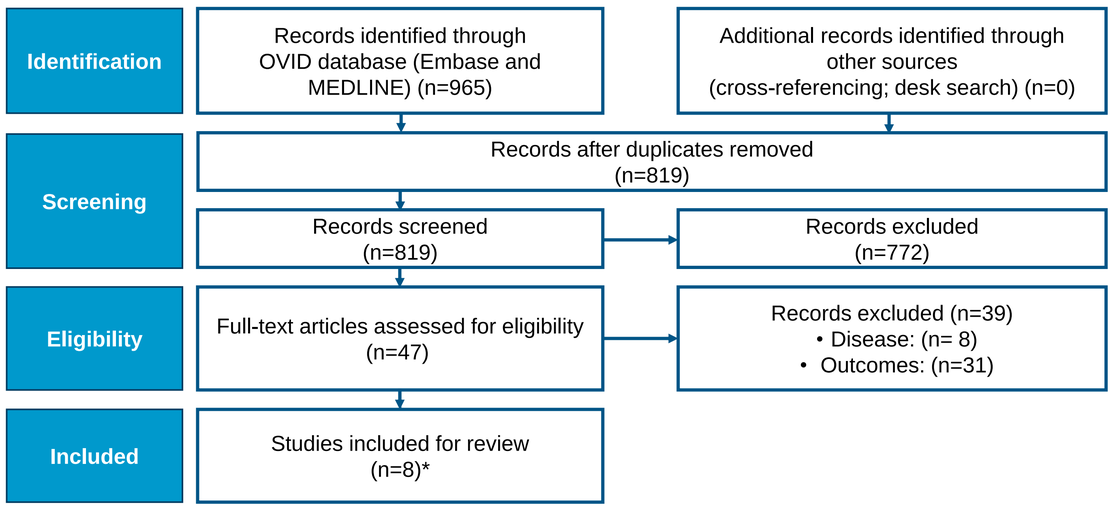
Results

- The mean HUVs were decreased from pre-relapse (0.656) to post-relapse state (0.595)[10]. Extrapolation of one relapse to multiple relapses predicted that each relapse led to decrease in HUVs from 0.7 to 0.3 (-43%) after 5 relapses.[11]
- In Germany, a cross-sectional study reported HUVs as 0.663 and 0.761 in NMOSD patients with AQP4 antibody–positive and double seronegative, respectively.[9]
- Patients with older age, longer disease duration (≥2 years), and higher EDSS score 8—9.5 were significantly associated with lower mean HUVs in NMOSD patients.[5]
- In addition to EQ-5D, 2 cross-sectional studies were also used VisQol and EQ-VAS scales and reported that mean HUVs at baseline was 0.79 and 0.65, respectively.[5, 8] More detailed results are presented in **Table 2**.

Table 1: Study eligibility criteria

PICOS	Inclusion criteria
Populations	Neuromyelitis optica spectrum disorder (NMOSD)
Interventions/ Comparators	No restriction
Outcomes	Health utility scores/index
Study designs	No restriction

Figure 1: PRISMA flow chart with identified studies



Note: * 8 studies (9 publications) included for review

Table 2: Health utility values reported in included studies

Study	Country	Scale	Utility values
Siritho, 2018[5]	Thailand	EQ-5D	• At baseline, mean (SD), (n=186): 0.41 (0.32)
	Thailand	EQ-VAS	• At baseline, mean (SD), (n=186): 0.65 (0.18)
Mealy, 2019[6]	USA	EQ-5D-5L	At baseline, mean (SD), • NMOSD (n=21) vs. US & world healthy control (NR): 0.738 (0.163) vs. 0.867 & 0.902
Kim, 2022[7]	South Korea	EQ-5D	• Median at baseline (n=57): 0.82
Hughes, 2022[8]	UK	EQ-5D-5L	• At baseline, Mean (95% central range (CR)) (n=111): 0.54 (0.49, 0.60)
	UK	VisQoL	• At baseline, Mean (95% CR) (n=111): 0.79 (0.74, 0.84)
Hummert, 2022[9]	Germany	EQ-5D-5L	• At baseline, mean (95% CI), (n=166): 0.693 (0.65 to 0.73) • AQP4 antibody-positive NMOSD, mean (95% CI) (n=141): 0.663 (0.61 to 0.72) • Double seronegative NMOSD, mean (95% CI) (n=25): 0.761 (0.67 to 0.85)
Berthele, 2020*[10]	Multicounty	EQ-5D-3L	Patients treated with eculizumab, and placebo groups were pooled; • Pre-relapse, mean (n=24): 0.656 • Post-relapse, mean (n=22): 0.595
Berthele, 2021*[11]	Multicounty	EQ-5D-3L	• Patients treated with eculizumab (n=7) and placebo (20) groups were pooled, who experienced a relapse and one relapse to multiple relapses lead to decrease in HUVs from 0.7 to 0.3
Levy, 2022[12] SAkuraSky trial	Multicounty	EQ-5D	At baseline, mean (SD) • Satralizumab vs. placebo: 0.70 (0.26) vs. 0.63 (0.29) At 24 weeks: • Satralizumab vs. placebo: 0.68 (0.29) vs. 0.73 (0.23)
Levy, 2022[12] SAkuraStar trial	Multicounty	EQ-5D	At baseline, mean (SD) • Satralizumab vs. placebo: 0.57 (0.31) vs. 0.63 (0.33) At 24 weeks: • Satralizumab vs. placebo: 0.64 (0.29) vs. 0.68 (0.31)

Keywords: EQ-5D-5L: EuroQoL Group 5 Dimension 5 Level Scale; EQ-5D-3L: European Quality of Life 5-Dimension questionnaire 3-Level; VisQoL: Vision and Quality of Life Index; EQ-VAS: EQ visual analog scale; SD: Standard Deviation; AQP4: Aquaporin-4; NMOSD: Neuromyelitis optica spectrum disorders; *Both are linked publications