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Letter to the Editor: New Observation

Virtual Multidisciplinary ALS Clinic Care During the COVID-19 Pandemic: A Canadian Cohort

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Given the heterogeneity and multiplicity of symptomatology and impairments in ALS, international clinical guidelines have recommended specialized multidisciplinary clinic care.[1] Outlined benefits of care within such multidisciplinary ALS Clinics include increased survival, reduced hospitalization, added utilization of therapeutics, and enhanced quality of life (QOL).[1] The COVID-19 pandemic presented significant challenges to the provision of ALS care and brought forward added potential impact of utilizing virtual modalities in the ALS Clinic setting.[2] The patient and caregiver perspective, including within this study's clinical context, has noted that VC may alleviate barriers to care, though there can be impediments to use, and in-person visits may be more preferable in certain circumstances.[3,4] However, there is very limited, and no Canadian, data pertaining to clinical outcomes associated with VC in ALS, including for patient-reported outcome measures (PROMs) such as QOL.[5]

The primary objective of this investigation was to measure patient-reported, health-related QOL among patients receiving virtual multidisciplinary ALS Clinic care over a 6-month period. Secondary aims included the quantification of ALS clinical progression, caregiver burden, and travel/time savings.

Participants with a diagnosis of ALS who received VC between March-August 2020 through the multidisciplinary Kingston ALS Clinic were eligible. We set up the virtual ALS Clinic throughout clinic rooms and multidisciplinary team members rotated through the rooms, while a given patient/caregiver(s) connected (via their own technology) and remained virtually in the same clinic room throughout. VC encounters were completed via video on regulated, secure platforms or audio (telephone), based on patient/caregiver preference. If there were irremediable technical challenges on video platforms, the visit was converted to audio.

This is a single-arm, prospective descriptive cohort study. The primary outcome of interest is patient-reported, health-related QOL, as measured through the EQ-5D-5L instrument.[6] Secondary measures included clinical progression via the ALS Functional Rating Scale-Revised (ALSFRS-R) score, the Clinical Frailty Scale (CFS); caregiver burden, via the Zarit Burden Interview (ZBI) score; and travel distance/time savings, calculated utilizing the fastest driving route available for each patient. Study instruments were administered separately from clinic visits in a follow-up phone call via a research assistant. This study was approved by Queen's University's Research Ethics Board (File# 6029650).

Baseline and 6-month follow-up outcome data were compared across EQ-5D-5L, EQ-VAS, ALFSRS-R (total and subscores), CFS, and ZBI scores. Data from the EQ-5D-5L questionnaire were analyzed utilizing the exploratory data analysis method described by Devlin et al. and a summary of changes via the Paretian Classification of Health Change.[6] Paired t-tests were used to compare baseline and 6-month follow-up for quantitative outcomes measures (EQ-VAS & ZBI). Missing data were addressed using case-wise analysis. All descriptive statistics and analytical statistics were completed within SPSS version 26.0, IBM with significance set to < 0.05.

In total, 11 participants were included in the study (2 females, 9 males; average age 65.9; demographic information outlined in Supplementary Table 1). Patient visit and travel information are outlined in Supplementary Table 2. The average roundtrip travel distance (patient's home to hospital) was 167.8 km (range: 26.2–372, SD: 108.8), leading to an average travel time per clinic visit of 1 hour 54 minutes (range: 36-222, SD: 58.3).

The ALSFRS-R (total and subscale scores) data are presented in Table 1. The mean (SD) ALSFRS-R total score for participants decreased from 33.5 (3.2) at baseline to 29.8 (13.1) at 6 months, showing a decline of 0.62 points per month. The mean (SD) CFS significantly increased from 5.0 (1.48) at baseline to 5.8 (1.14) at 6 months. The mean (SD) of ZBI for caregivers showed a statistically non-significant trend down ($P = .334$) from 24.67 (17.61) at baseline to 20.83 (SD = 16.68) at 6 months. The proportion of caregivers with high burden, defined as a ZBI score of 17 or more, also demonstrated a statistically non-significant ($P = .400$) trend of reduction from 55.5% at baseline to 33.3% at 6-month follow-up.

In total, 10 participants completed both the baseline and 6 months for the EQ-5D-5L (Table 2). The mean (SD) EQ-VAS

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Table 1: Baseline and 6-month follow-up outcome assessment

Outcome Measure		Baseline	6-Month	*P*-Value
ALSFRS-R	Mean (SD)			
Total Score:	[0–48]	33.5 (3.2)*	29.8 (13.1)*	
Subscale:	Bulbar [0–12]	8.7 (3.1)	9.0 (3.4)*	
	Motor [–24]	13.9 (6.9)	12.0 (8.3)*	
	Respiratory [0–12]	9.9 (3.2)*	8.8 (3.2)*	
EQ-5D-5L-visual analogue scale (EQ-VAS)	Mean (SD) [0–100]	51.3 (22.3)*	57.4 (21.6)*	0.591
Clinical Frailty Scale	Mean (SD) [1–9]	5.00 (1.5)	5.80 (1.1)*	0.042
Zarit Burden	Mean (SD) [0–88]	24.7 (17.6)**	20.8 (16.7)***	0.334
Interview (ZBI) Score	ZBI Score (> = 17)	5 (55.5%)	2 (33.3%)	0.400

ALSFRS-R = amyotrophic lateral sclerosis functional rating scale-revised; ZBI = zarit burden interview.
*$n = 10$ (1 missing).
**$n = 9$ (2 missing).
***$n = 6$ (5 missing).

score showed a statistically non-significant trend ($P = .591$) toward improvement from 51.3 (22.3)–57.4 (21.6) at baseline and 6 months, respectively. On the EQ-5D-5L questionnaire dimensions, there was a decrease in the degree of problems related to Anxiety/Depression and Self-Care, an increase for Pain/Discomfort and Usual Activities, and no change for Mobility. Utilizing the Paretian Classification of Health Change, there was an improvement of QOL in 2 (20%), decline in 3 (30%), no change in 1 (10%), and mixed change in 4 (40%).

Over a 6-month interval featuring virtual multidisciplinary ALS care, there was expected clinical progression yet significant improvement in several dimensions of patient-reported QOL, stability in overall QOL, trends toward improvement in patient-perceived health and reduction in caregiver burden, as well as considerable travel distance/time savings. These findings are in keeping with the previously cited benefits of specialized multidisciplinary in-person ALS Clinic care, including improved QOL,[1] however also offer added support to the feasibility and potential benefit of adopting VC strategies.

The specific potential advantages of incorporating VC into multidisciplinary ALS Clinic care are detailed in Supplementary Table 3. We identified significant travel distance/time reduction, without accounting for additional time elements (parking/transferring/check-in/clinic navigation/inclement road conditions-winter months). Moreover, patients/caregiver(s) often need to coordinate accessible transportation, and travel frequently necessitates caregiver absence from work/other responsibilities, identified challenges that would be alleviated with VC.[3,4]

There was some variability across other QOL sub-domains with improvement in several (Anxiety/Depression and Self-Care), while others showed stability or some deterioration. This could be a reflection of individual patient characteristics, method of adjustment to the disease, prognosis, family/social supports, and modifications to accommodate a progressive disease.[2] Related, in the non-VC context, as ALS progresses, the EQ-5D-5L instrument has demonstrated worsening on the more physical functional associated dimensions, but not in anxiety/depression.[7] Patient-perceived health showed a trend toward improvement, despite disease progression. Given the cited beneficial impact of multidisciplinary ALS Clinic care on QOL,[1,7] our results speak further to the potential of the virtual medium to facilitate this recommended care model.

Similarly, our findings also noted a trend toward reduction in caregiver burden despite clinical progression. This may reflect the degree of support offered through the multidisciplinary ALS Clinic, including care provision, but also education around disease status and progression as well as linking patients and caregivers with relevant community services.

A primary strength of our study is that it is the first assessment of implementation of virtual multidisciplinary ALS care in Canada, and we demonstrate benefits noted with the traditionally in-person model.[1] Limitations of this study include the relatively small number of followed patients within our cohort, from a condition with low incidence/prevalence, the absence of a control group, follow-up does not extend for the full disease duration, and the potential impact of the early stages of the COVID-19 pandemic. Of note, other investigation has suggested that pandemic-related public health measures/restrictions did not significantly impact QOL, but was associated with a significantly increased caregiver burden in an ALS cohort, using the same instruments as our study.[8]

Future research should focus on individual patient and disease characteristics that would determine a more optimal match for VC delivery in ALS, including hybrid models (in-person and virtual delivery) for further protocol optimization. Finally, ongoing work to address systemic and local barriers experienced by patients, caregivers, and ALS care teams in connecting via virtual modalities is essential.

In conclusion, our study featured the successful application of synchronous virtual multidisciplinary ALS Clinic care in Canada. The use of VC in this context demonstrated an improvement in several domains of patient-reported QOL, a trend toward reduced caregiver burden, and considerable reduction in travel distance/time over an interval with expected clinical progression. Given the demonstrated feasibility and efficacy of VC delivery within this and recent cohorts of patients, further investigation and work toward the implementation of virtual strategies within ALS care is warranted.

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Table 2: Baseline and 6-month follow-up EQ-5D-5L questionnaire data

	Dimensions (of health)									
	Mobility		Self-care		Usual activities		Pain / discomfort		Anxiety / depression	
Levels (of response)	Base	6mo	Base	6mo	Base	6mo	Base	6mo	Base	6mo
1 (no problems)	2 (20%)	2 (20%)	3 (30%)	4 (40.0)	1 (10.0)	0	3 (30.0)	2 (20.0)	2 (20.0)	4 (40.0)
2 (some problems)	2 (20%)	2 (20%)	3 (30%)	0	1 (10.0)	4 (40.0)	4 (40.0)	4 (40.0)	4 (40.0)	3 (30.0)
3 (moderate problems)	2 (20%)	1 (10%)	1 (10%)	2 (20.0)	3 (30.0)	1 (10.0)	1 (10.0)	4 (40.0)	2 (20.0)	2 (20.0)
4 (severe problems)	1 (10%)	0	1 (10%)	0	2 (20.0)	1 (10.0)	2 (20.0)	0	2 (20.0)	1 (10.0)
5 (unable/extreme[a])	3 (30%)	5 (50%)	2 (20%)	4 (40.0)	3 (30.0)	4 (40.0)	0	0	0	0
Total[b]	10 (100%)	10 (100%)	10 (100%)	10 (100%)	10 (100%)	10 (100%)	10 (100%)	10 (100%)	10 (100%)	10 (100%)
Number Reporting Problems[c]	8 (80%)	8 (80%)	7 (70%)	6 (60%)	9 (90%)	10 (100%)	7 (70%)	8 (80%)	8 (80%)	6 (60%)
Changes in Numbers Reporting Problems	0		−1		+1		+1		−2	
% Change in Numbers Reporting Problems	0%		−14.2%		+11.1%		+14.2%		−25.0%	
Rank of Dimensions in Terms of % Changes	4		2=		3		2=		1	
Paretian Classification of Health Change										
No Change	Improve		Worsen		Mixed Change		Total			
1 (10%)	2 (20%)		3 (30%)		4 (40%)		10 (100%)			

[a]Extreme response descriptor used for Pain/Discomfort and Anxiety/Depression dimensions.
[b]Results are for those who responded to both the baseline and the 6-month follow-up questionnaire. As follow-up data not available, Patient 7 baseline data not included: Mobility – 3, Self-Care – 3, Usual Activities – 4, Pain / Discomfort – 3, Anxiety / Depression – 2,
[c]"Problems" = levels 2 + 3 + 4 + 5.

Supplementary material. The supplementary material for this article can be found at https://doi.org/10.1017/cjn.2023.302.

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References

1. Shoesmith C, Abrahao A, Benstead T, et al. Canadian best practice recommendations for the management of amyotrophic lateral sclerosis. Can Med Assoc J. 2020;192:E1453–E1468.
2. De Marchi F, Sarnelli MF, Serioli M, et al. Telehealth approach for amyotrophic lateral sclerosis patients: the experience during COVID-19 pandemic. Acta Neurol Scand. 2021;143:489–96.
3. Chan S, Karacam M, Ritsma B, et al. Patient perceptions on the virtual amyotrophic lateral sclerosis clinic during COVID-19. Can J Neurol Sci. 2022;49(4):618–22.
4. Chan-Nguyen S, Ritsma B, Nguyen L, et al. Virtual care access and health equity during the COVID-19 pandemic, a qualitative study of patients with chronic diseases from Canada. Digit Heal. 2022;8: 205520762210744.
5. Andrews JA, Berry JD, Baloh RH, et al. Amyotrophic lateral sclerosis care and research in the United States during the COVID-19 pandemic: challenges and opportunities. Muscle Nerve. 2020;62:182–6.
6. Devlin NJ, Parkin D, Browne J. Patient-reported outcome measures in the NHS: new methods for analysing and reporting EQ-5D data. Health Econ. 2010;19:886–905.
7. Wei Q-Q, Hou Y, Chen Y, et al. Health-related quality of life in amyotrophic lateral sclerosis using EQ-5D-5L. Health Qual Life Outcomes. 2021;19:181.
8. Giusiano S, Peotta L, Iazzolino B, et al. Amyotrophic lateral sclerosis caregiver burden and patients' quality of life during COVID-19 pandemic. Amyotroph Lateral Scler Front Degener. 2022;23:146–8.