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'Real life' remote dystonia assessment:
Feasibility, accuracy and practice implications

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Word count: 754

Running Title: Remote dystonia assessment

Key words: Dystonia; Telemedicine; Reproducibility of Results

Telemedicine, defined as the use of synchronous two-way communication media to provide medical care, has seen a surge in uptake during the COVID-19 pandemic. Its future role in movement disorder clinical and research practice however remains to be defined. Boasting high levels of satisfaction, alongside the potential to increase accessibility and reduce costs, many favour increasing its adoption[1]. However, remote visits are not the same as in-person assessments. They may produce lower diagnostic certainty[2], are limited by technical factors, and curb some aspects of 'traditional' care, including the neurological examination.

Structured video ratings form a cornerstone of dystonia clinical and research practice, allowing documentation of phenomenology for later review, blinded rating or independent assessment. Traditionally, recordings are performed in-person using high-resolution equipment in a clinical setting.

It remains unknown whether standardised dystonia assessments conducted remotely are valid substitutes for in-person ratings. Here, we sought to determine, under 'real-life' conditions, the feasibility and accuracy of remote dystonia severity assessments using the Burke-Fahn-Marsden dystonia rating scale(BFM), compared to traditional in-person assessments[3].

The motor sub-score of the BFM was administered in the standard fashion[3] to clinically stable patients with dystonia under two conditions:

a) During an in-person assessment, and

b) During a virtual routine follow-up clinical consultation carried out via Zoom Video Communications Inc.(Zoom)

Interactions were recorded using a high-resolution camera or the secure video recordings function of Zoom, respectively. Patients where therapy changed between both conditions, or if videos were performed >3months apart were excluded. Two trained assessors (KS, EM), blinded to each others' ratings, independently scored all videos. Rating of each video category was separated by 1.5 months to avoid memory bias.

Zoom calls were performed using devices available to participants. They were instructed to be in an appropriate environment for examination. In-line with 'real-life' conditions, a dedicated technical support team was not available. Rather, troubleshooting was carried our by the physician. Informed consent for video recordings was obtained from all patients.

Between October and December 2020, 36 patients underwent in-person BFM assessments at our institution. Of these, 6 were unable to participate due to lack of internet access or availability of a compatible device. A further 5 declined to participate. A total of 25 patients(13 women, mean age 53+/-18 years, 19 generalised dystonia, 6 segmental dystonia) underwent both assessments. Clinical details are provided in supplementary Table 1. In 7 Zoom videos, at least one body part was un-rateable (supplementary table 2). All un-rateable items were excluded from the final analysis.

Dystonia severity scores were generally lower for Zoom ratings, particularly for eyes, mouth and legs(Table 1). Technical issues were encountered in many patients(supplementary table 3).

To our knowledge, this is the first study examining the validity of 'real-life' remote dystonia assessments conducted in patients' own home environments. Previous similar studies required patients to travel to locals hospitals(where specialist teleconferencing equipment was available)[4], undergo telemedicine training and meet specific device/internet access criteria[5], or have restricted assessments to easily visualised body parts e.g. the neck[5]. Consequently, they lacked generalizability to day-to-day practice settings.

Our data suggests that remote BFM assessments may be feasible, with ICCs showing excellent intra-rater reliability for total BFM scores and moderate-excellent agreement for individual items of the BFM (Table 1). However, the tendency to under-score dystonia severity (particularly of the face and legs) on virtual encounters needs further consideration, as it may limit the validity of remote video assessments as substitutes for in-person ratings. This is especially important for research studies, where under-scoring of video ratings might significantly bias results. Poor visualisation due to technical factors, limited camera resolution, and absence of assistants(especially for severely disabled patients) are factors which could be addressed in order to improve agreement between both modalities(supplementary table 3).

Our study has some limitations. First, dystonia severity may fluctuate over time. Differences in scoring may therefore reflect changes in disease rather than issues with video assessments per-se. Second, we restricted our assessment only to the BFM scale, which is purely visual and does not require in-person rater intervention. It is therefore not generalizable to other assessments. Third, our sample size was small and our findings would therefore need to be replicated in larger cohorts.

Remote video assessments have great appeal. However, further work is needed if they are to become valid substitutes for in-person ratings. Hidden costs, including equipment, set-up time, and the time required for the help of assistants should also be borne in mind. Future studies may wish to explore strategies for dealing with issues such as those identified in this cohort.

Author Roles:

1. Research project: A. Conception, B. Organization, C. Execution;
2. Statistical Analysis: A. Design, B. Execution, C. Review and Critique;
3. Manuscript Preparation: A. Writing of the first draft, B. Review and Critique.

EM: 1A, 1B, 1C, 2C, 3A, 3B

KGS: 1A, 1B, 1C, 2A, 2B, 2C, 3A

PL: 2C, 3B

TF: 1A, 1B, 2C, 3B

Disclosures:

Funding Sources and Conflict of Interest: No specific funding was received for this work. The authors declare that there are no conflicts of interest relevant to this work.

Financial Disclosures for the Previous 12 Months: EM is funded by the Edmond J. Safra Foundation. He also receives research support from the National Institute for Health Research University College London Hospitals Biomedical Research Centre. KGS reports no disclosures. PL has received honoraria and travel expenses from Medtronic and Boston Scientific we well as a grant from Boston Scientific. TF has received grants from National Institute of Health Research, Michael J Fox Foundation, John Black Charitable Foundation, Cure Parkinson's Trust, Innovate UK, Van Andel Research Institute and Defeat MSA. He has served on Advisory Boards for Peptron, Voyager Therapeutics, Handl therapeutics, Living Cell Technologies, Bial & Profile Pharma. He has received honoraria for talks sponsored by Bial, Profile Pharma, Boston Scientific.

Ethical Compliance Statement

All patients provided informed written (in-person ratings) and verbal (online ratings) consent for data collection, analysis and publication. We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines. The authors confirm that the approval of an institutional review board was not required for this work.

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Legends

Supplementary Table 1: Demographic and clinical characteristics of the studied cohort.

Supplementary table 2: Items removed from final analysis.

Supplementary table 3: commonly encountered issues during remote dystonia video assessments.

Table 1: Means and intra-rater reliability of the BFM scale for the clinic video and zoom video contexts

	Clinic video scores Mean (SD)	Zoom video scores Mean (SD)	ICC (95%CI)
Total BFM score	26.15 (21.52)	19.55 (18.48)	0.91 (0.71-0.96)
Eyes	0.48 (1.10)	0.18 (0.64)	0.56 (0.21-0.76)
Mouth	2.09 (2.34)	1.30 (2.24)	0.76 (0.540.87)
Speech and Swallowing	2.22 (3.59)	2.12 (3.51)	0.88 (0.79-0.93)
Neck	3.9 (2.69)	3.17 (2.40)	0.91 (0.79-0.96)
Right arm	3.8 (4.46)	2.74 (3.66)	0.87 (0.76-0.93)
Left arm	4.38 (4.65)	3.64 (4.43)	0.87 (0.75-0.93)
Trunk	3.16 (4.24)	3.87 (3.87)	0.74 (0.51-0.86)
Right leg	3.38 (4.68)	1.7 (3.95)	0.75 (0.47-0.88)
Left Leg	3.06 (4.63)	1.84(3.93)	0.82 (0.67-0.91)

Clinic video scores have been adjusted such that if an item of the BFM was unrateable via Zoom, it has also been removed from the clinic-video scores prior to analysis. Means consider average score across the two raters. BFM: Burke-Fahn-Marsden dystonia rating scale; ICC: Intraclass Correlation Coefficient; CI: Confidence Intervals Upper and Lower Bounds; SD: Standard Deviation.

An ICC of 0.5-0.75 indicate moderate agreement, 0.75-0.90 indicate good agreement and >0.90 indicate excellent agreement of scores.