



# Tele-Yoga for the Management of Cervical Dystonia: A Safety and Feasibility Trial

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**Background:** Cervical dystonia impacts quality of life and activities of daily living. Botulinum toxin injections, the standard treatment, are not effective for all and often include bouts of recurring symptoms between injections. There is a need for supplementary treatments such as yoga, which has been shown to be beneficial for individuals with chronic neck pain and movement disorders. However, individuals with cervical dystonia experience barriers impeding access to in-person yoga. Thus, alternative delivery methods that can optimize access while maintaining safety must be investigated. The purpose of this study is to investigate the feasibility and safety of a synchronous oneon-one tele-yoga intervention for individuals with cervical dystonia.

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James-Palmer AM and Daneault J-F (2022) Tele-Yoga for the Management of Cervical Dystonia: A Safety and Feasibility Trial. Dystonia 1:10015. doi: 10.3389/dyst.2021.10015 **Methods:** Individuals with cervical dystonia were enrolled in a single group pilot feasibility study consisting of a 6-weeks tele-yoga intervention bookended by two assessment sessions, ending with a 6-weeks follow-up period and associated final assessment session. The live one-on-one tele-yoga intervention consisted of breathing, postures, and relaxation and was delivered for 30 min twice weekly. Primary outcomes included adherence, adverse events, technological challenges, and usability. Secondary outcomes included enjoyment, yoga status at follow-up, clinically relevant questionnaires, and functional measures.

**Results:** Of the fifteen individuals enrolled, one did not complete the follow-up assessment. Intervention adherence was 93%. No significant adverse events related to the intervention occurred. Manageable technological challenges occurred. Mean usability and enjoyment were high.

**Conclusions:** The implementation of a one-on-one tele-yoga intervention for individuals with cervical dystonia is safe and feasible thus, efficacy trials should be initiated.

Clinical Trial Registration: https://www.clinicaltrials.gov/ct2/show/NCT04348669, NCT04348669

Keywords: dystonia, complementary therapy, yoga, exercise, tele-rehabilitation

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# INTRODUCTION

Cervical dystonia (CD), the most common form of adult-onset focal dystonia, is vastly understudied. With an approximate prevalence of 28-183 cases per million (1), CD is characterized by its cardinal motor impairments including abnormal head and neck posture, decreased range of motion, tremor, and muscle spasms (2). Additionally, individuals with CD often experience comorbid mental health conditions such as symptoms of anxiety, depression, and/or sleep dysfunction (3). Pain, in combination with these non-motor symptoms, often leads to decreased quality of life (3). The current gold standard treatment of botulinum toxin injections (4) focuses primarily on improving motor impairments (5) and pain (6). Additionally, this treatment, while generally safe and effective, often includes bouts of recurring symptoms between injections (7) with decreased satisfaction of treatment occurring prior to reinjections (7). In a survey, out of 1,071 respondents, 25% reported being fairly/very dissatisfied with their care (8). Of 400 respondents who were specifically dissatisfied with their botulinum toxin treatment, 46% reported it was due to non-responsiveness and 33% reported it was due to side effects (8). Thus, there is a need for safe supplementary interventions that can be paired with botulinum-toxin injections and have the potential to address both motor and non-motor impairments of CD while having minimal side effects.

Yoga has the potential to impact both motor and non-motor impairments and has been sought out by individuals with chronic health conditions to manage care (9). Yoga, which can be low impact, may be especially useful for individuals with CD, who report that fatigue and motor symptoms impede participation in physical activity (10). While, to our knowledge, there is no published research involving the implementation of yoga for individuals with CD, yoga has been shown to be beneficial for individuals with chronic neck pain (11) and for others with movement disorders, such as those with Parkinson's disease (PD) (12–15).

Despite potential benefits of yoga, barriers may make inperson yoga inaccessible for individuals with chronic conditions such as CD. Disease-related mobility impairments make travel to in-person interventions challenging (10). Even if mobility impairments do not impede travel, transportation difficulties including lack of access to a vehicle, distance to the intervention, and costliness of transportation may impede access (16). Individuals with CD also report that a lack of reachable knowledgeable professionals inhibits participation (10). Stigma (10) and the fear of being watched by others may also impede participation in community based in-person yoga classes. Therefore, there is a need for an intervention delivery method that can address these barriers by limiting the need for travel, facilitating access to knowledgeable professionals, and providing the opportunity to engage in interventions in a judgement free environment such as one's own home.

Delivering yoga remotely *via* technological means (tele-yoga) may address some of these barriers. Tele-yoga, similarly, to telemedicine and telerehabilitation, can be delivered synchronously (in real-time) or asynchronously (through prerecorded materials). Telemedicine has been used successfully

for the evaluation of individuals with CD (17). There is currently an interest in telerehabilitation approaches for this population as indicated in a recently published paper demonstrating telerehabilitation exercises for individuals with CD (18). There is, however, support in the literature for both synchronous and asynchronous tele-interventions for individuals with other movement disorders such as PD (19–21). The work thus far has shown that physical tele-interventions may be safe and beneficial for individuals with PD, however there is less known about other movement disorder populations and tele-yoga specifically.

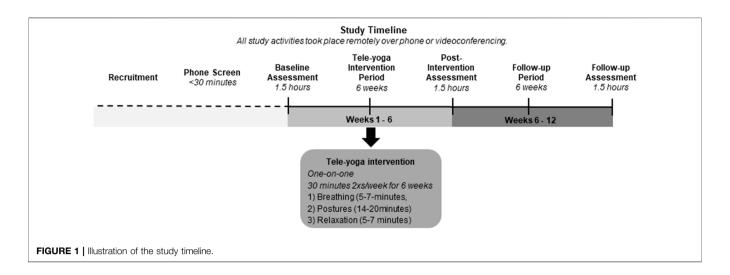
There are currently no tele-yoga studies specifically investigating individuals with movement disorders, but there is a small body of work regarding tele-yoga in general. Overall, the existing studies consist primarily of asynchronous delivery with a few studies examining synchronous delivery of tele-yoga. However, synchronous tele-yoga may afford opportunities to optimize safety, symptom assessment/management, and participant understanding. Three studies to date have addressed the implementation of synchronous tele-yoga in adult populations, including women with breast cancer (22), individuals with cardiopulmonary conditions (23), and veterans (24). These few existing studies showed high enjoyment, satisfaction, and improvement in mental health outcomes (22-25). Despite potential benefits, the literature is sparse, not specific to CD, and presents some current challenges including technological difficulties and low adherence (22, 23) which may be addressed through changes in the technology used and different approaches to scheduling. This current study aims to build on existing literature by investigating the feasibility and safety of a synchronous one-onone tele-yoga intervention for individuals with CD. Based on the available literature, we hypothesized that the synchronous tele-yoga intervention would be feasible and safe for individuals with CD.

# MATERIALS AND METHODS

This study was a non-randomized single-group pilot intervention study. It was completed entirely remotely from participants' residences through videoconferencing (April 2020 and May 2021). It was unblinded and consisted of: 1) baseline assessment session, 2) 6-weeks synchronous tele-yoga intervention period (participants completed twice weekly live yoga sessions one-onone with a certified instructor), 3) post-intervention assessment session, 4) 6-weeks follow-up period (participants continued their regular activities with no interaction from the study team), and 5) final follow-up assessment session (see **Figure 1**). This study was registered with ClinicalTrials.gov (NCT04348669) and approved by the Institutional Review Board at Rutgers University. It was reported in accordance with the STROBE guidelines for Cohort Studies (26), and the CONSORT extension for pilot and feasibility trials (27).

### **Participants**

A convenience sample of 15 individuals with CD were recruited using a study information flyer circulated on online platforms including dystonia-related social media support groups and dystonia-related organization websites. Recruitment ceased when



sufficient information regarding feasibility and safety was obtained from enrolled participants. Eligibility was assessed during a phone screen. Individuals were eligible to participate if they were 1)  $\geq$ 18 years old, 2) English-speaking, 3) diagnosed with CD, 4) able to communicate with the study team and yoga instructor such that they could indicate pain, answer questions, and provide feedback either verbally or through written communication and body gestures, 5) able to access WiFi via an applicable technological device, and 6) agreeable to use Zoom and to be video recorded. People were not eligible if they had 1) unmanaged major depressive disorder (self-reported), 2) a condition that could prevent the ability to engage in physical activity, 3) major cognitive impairment, 4) past yoga experience of five or more times within the last 2 months or 5) were pregnant (self-report). During the first live videoconferencing assessment session eligibility was confirmed and informed consent was obtained.

### Intervention

The tele-yoga intervention was delivered to participants' residences by a certified yoga instructor remotely and synchronously through the videoconferencing platform Zoom individually for 30-min two times a week for 6-weeks. The intervention was developed using the previously published "eight domains for developing a yoga protocol" (28). Each session included three key yogic elements in the following sequence 1) breathing exercises (5–7 min), 2) yoga postures (14–20 min), and 3) meditation exercises (5–7 min). Postures included seated, supine, quadruped, standing postures, and mindful movement of the head and neck depending on the participants' abilities. The intervention was delivered and modified in real-time to fit the needs of each participant.

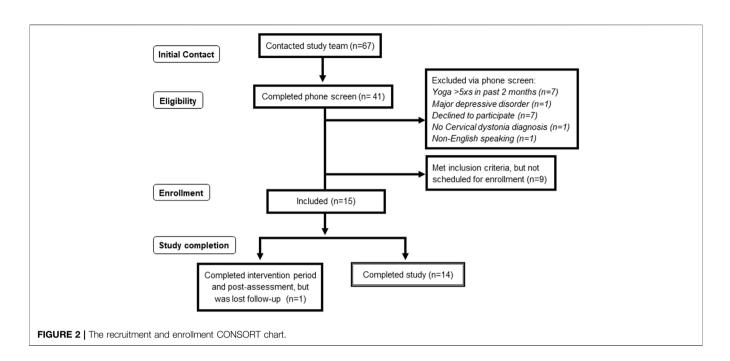
### **Outcome Measures**

Primary outcome measures included recruitment rate, retention rate, adherence, adverse events, technological challenges, and usability. Recruitment rate was calculated by computing the number of participants who met the inclusion criteria and were enrolled in the study compared to the number of individuals who engaged in the phone screening process. Retention rate was calculated by computing the number of participants who completed the study compared to those who were enrolled. Adherence was measured *via* yoga session and assessment session attendance. Adverse events were recorded with their potential relationship to the study and their severity. For technological challenges, anything perceived as a challenge by the yoga instructor or participant was documented with severity of the challenge measured by time disrupted as *mild* (1–3 min), *moderate* (3–10 min), or *severe* (>10 min). Usability was assessed using the ten item Systems Usability Scale (SUS) (29).

Secondary outcome measures included enjoyment, yoga status at follow-up, clinically relevant questionnaires, and functional measures. Enjoyment of the intervention was assessed using a 0-10-point scale for the overall study experience, the overall yoga intervention, and the three yogic elements (breathing, postures, and relaxation/meditation). At the follow-up assessment, participants completed a questionnaire about their yoga practice over the 6-weeks follow-up period after the synchronous tele-yoga intervention had ended. Disease-specific quality of life was assessed by the Craniocervical Dystonia Questionnaire (CDQ-24) (30). General quality of life was assessed with the SF-36 (31). Anxiety and depression were assessed using the Beck Anxiety Inventory (BAI) (32) and Beck Depression Inventory-II (BDI) (33). respectively. Sleep dysfunction was assessed using the Pittsburgh Sleep Quality Index (PSQI) (34). Dystonia symptom severity, disability, and pain were assessed using the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) (35). The TWSTRS was administered in real-time over videoconferencing. The session was video recorded and the TWSTRS was scored at a later date using the recorded session. It has been shown that there is excellent agreement between the TWSTRS motor severity domain when it is administered in-person vs. when it is administered during a telemedicine evaluation (17). Lastly, physical function was assessed using the five times sit to stand (FTSTS) (36). Permission and licenses were obtained for questionnaires as applicable.

### **Data and Statistical Analyses**

Score computation and statistical analysis was completed using R and R studio (37, 38). Total scores and available subscale scores



| TABLE 1 | Participant demographics. |  |
|---------|---------------------------|--|
|         |                           |  |

| Sex   | Female ( <i>n</i> = 11), Male<br>( <i>n</i> = 4) |
|---|--|
| Age   | 51.13 years ± 13.55                              |
| Race  | White $(n = 14)$ , Bangla $(n = 1)$              |
| Weight  | 168.11 lbs ± 38.68                               |
| Height  | 67.13 in. ± 3.76                                 |
| Body Mass index                               | 25.86 ± 4.23                                     |
| Years since diagnosis                         | $9.03 \pm 8.09$                                  |
| Participants managing CD with botulinum toxin | <i>n</i> = 10                                    |

Abbreviations: CD, cervical dystonia.

were computed for all applicable outcome measures. Missing items were addressed using item mean imputation (i.e., the mean of the specific missing item for the relevant time period across all subjects). Descriptive statistics of all outcomes were calculated. For the relevant clinical outcome measures with normal distributions, repeated measures ANOVAs were computed, and effect sizes (partial eta squared) were calculated. For the relevant clinical outcome measures with non-normal distribution, Friedman tests were computed, and effect sizes (Kendall's W) were calculated. A secondary analysis was completed in which participants who had received botulinum toxin injections over the course of the study (participants 6, 8, 11, 13, 14, and 15) were excluded from analysis. The methods for the secondary analysis were the same as those for the primary analysis described above.

### RESULTS

Of the 67 participants who contacted the study team, 15 participants were enrolled in the study, 14 of which completed

the study in its entirety (see **Figure 2**). Individuals were able to remotely participate from all over the world including Australia (n = 5), Canada (n = 2), and the United States (n = 8). See **Table 1** for participant characteristics. Supplementary management techniques for CD symptoms at baseline included: oral pharmaceutical management (n = 5), deep brain stimulation (n = 2), physical activity (n = 7), hypnosis therapy (n = 1), and Bowen therapy (n = 1).

Participants 1 and 2 were overdue for botulinum toxin injections at the start of the study by 5 and 2 months, respectively. Due to the pandemic, they did not receive any injections over the course of the study. Participant 11 was 1 month overdue and received a botulinum toxin injection between the first and second yoga session. Participant 15 received a botulinum toxin injection between yoga sessions five and six and was 1 week overdue for the injection at that time. Participant 8 received his first botulinum toxin injection between yoga sessions seven and eight. Participant 6 had not received an injection in years (unable to explicitly quantify) and received one during the 3rd week of the follow-up period. Participant 12 received an injection 1 week prior to the baseline assessment. Participant 13 received an injection 2 months prior to the baseline assessment and then between yoga sessions three and four. Participant 14 received an injection between the baseline assessment and the first yoga session. Participant 10 did not report when they received their most recent injection and did not receive any injections during the study period. All other participants (participants 3, 4, 5, 7, and 9) did not use botulinum toxin injections to manage CD symptoms.

### Adherence

Study adherence was high with 14 out of 15 participants completing all three assessment sessions. Yoga-intervention adherence was also high (93%) with 168 sessions attended out

of 180 sessions scheduled. Mean attendance was 11.2 ± 1.86 sessions. Eleven participants received 100% of the intended dosage, two of these participants completed the intended 12 sessions within 7 weeks due to rescheduling. Three participants completed at least 10 of the intended sessions, while only one participant completed less than 10 sessions. Reasons for rescheduling included: resting following botulinum injections (n = 1), migraine (n = 1), participant illness (n = 7), instructor illness (n = 3), instructor internet connectivity (n =3), participant overslept (n = 1), participant scheduling conflict (n = 6), instructor scheduling conflict (n = 3), participant vacation (n = 3). Reasons for cancelations included unrelated exacerbation of CD (n = 4), instructor power-outage (n = 1), participant noshow with reason not-provided (n = 3), and participant scheduling conflicts (n = 3). Participants were encouraged to engage in home practice between intervention sessions consisting of the exercises taught in each session. Participants reported engaging in home practice inconsistently and were generally unable to quantify or specify what they did.

### Safety

No significant adverse events were attributed to the intervention. As expected, all participants reported baseline pain including CDrelated (neck/shoulder) pain (n = 15), hip pain (n = 3), low back pain (n = 3), and ankle pain (n = 1). Seven participants reported temporary increases in pain (one-point) during specific postures. At the end of one session a participant reported a one-point pain increase, but resolved the pain through self-massage. Other mild events related to the intervention included transient dizziness with a position change (n = 1), headache following a position change (n = 1), as well as temporary increases in symptoms in supine position (n = 1) and with breathing exercises (n = 1); all of which abated when the activities were stopped. Five events (resulting in modification of the yoga sessions) with a possible causal relationship to the intervention included development of new soreness/pulling at the shoulders (n = 2), increased rigidity/ soreness of the back of the neck (n = 1), new hip pain (n = 1), and new low back pain (n = 1) during the course of the intervention, all of which resolved prior to the end of the study. Two participants reported transient mood fluctuations, one unrelated between sessions, and one during the relaxation exercises which resolved prior to the session's end. Mild unrelated events were successfully managed by participants and/or their neurologists as applicable. These mild unrelated events included injection site pain following botulinum toxin injections (n = 1), increased whole body shaking attributed to a medication error (n = 1), and chest tightness attributed to asthma (n = 1). One moderate event unrelated to the intervention (mild symptoms of the Sars-CoV-2 virus) resulted in the rescheduling of one session. Lastly, one severe unrelated event occurred during the follow-up period in which a participant was hospitalized for differential diagnosis of an occipital migraine.

### **Technological Challenges**

Various technological challenges occurred during the yoga sessions (**Table 2**). Audio and visual delays, the most common challenge, did not greatly disrupt time but did impact session

quality. A few mild interruptions not caused by technological challenges disrupted time from the study. None of the mild or moderate challenges/interruptions impacted the amount of yoga received, as time disruption was accommodated within affected sessions. Most severe challenges were also able to be accommodated. However, in two instances, the instructor had difficulty connecting to the internet and needed to reschedule the sessions. Despite these technological challenges, scores on the SUS (89.67  $\pm$  7.67) indicated good usability.

# **Enjoyment and Yoga Status at Follow-Up**

Mean enjoyment scores were high for the remote study experience (9.5  $\pm$  0.94), overall yoga intervention (9.2  $\pm$  1.37), breathing exercises (8.6  $\pm$  2.61), postures (9.07  $\pm$  1.22), and relaxation exercises (8.73 ± 2.25). Despite high enjoyment levels, not all participants continued to practice yoga consistently after the intervention period ended during the 6-weeks follow-up period. Participants reported varying levels of practice including inconsistent practice (n = 5), one time per week (n = 1), twothree times per week (n = 4), and greater than three times per week (n = 4). Participants reported practicing at home without any instruction (n = 11), via live videoconferencing (n = 2), and through online videos (n = 1). Participants reported practicing postures, breathing, and meditation (n = 8), postures and breathing (n = 3), postures and meditation (n = 1), and meditation only (n = 1)1). Two participants reported not practicing as much as they desired because they did not have access to an instructor while another attributed lack of practice to low energy levels.

### **Clinically Relevant Measures**

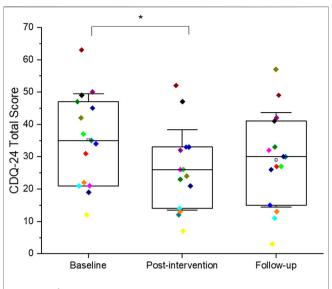
There were 10, 4, and 140 items missing for the baseline, postintervention, and follow-up assessment periods respectively (Supplementary Table S1). Overall disease related quality of life and the subdomains of stigma and emotional well-being demonstrated preliminary efficacy with post-hoc testing indicating significant differences between the baseline and post-assessment periods for these measures. In addition to disease-related quality of life, statistically significant improvements were also seen in CD severity as shown by the severity subscale on the TWSTRS, with post-hoc testing revealing improvements between baseline and post intervention. No other outcomes demonstrated statistically significant differences. See Table 3 and Supplementary Table S2 for detailed results, including effect sizes which ranged from small to large. See Figures 3, 4 for a visual representation of the total scores for the CDQ-24 and TWSTRS, respectively. Additionally, a supplementary analysis of the data, excluding all participants who received a botulinum toxin injection was also performed to minimize the possibility that the improvements observed here were solely related to the effect of the injection in those participants. While minor differences were observed in the magnitude of the effect sizes across the different outcome measures, statistically significant differences remained for the CDQ-24 total score, stigma subscale, and emotional wellbeing subscale as well as for the TWSTRS symptom severity subscale. An additional statistically significant difference also emerged for the SF-36 general health subscale. Detailed results of this

#### TABLE 2 | Technological challenges during yoga sessions.

|                          | Mild (1–3 min)   | Moderate (3–10 min)   | Severe (>10 min)   |
|--------------------------|--|---|--|
| Audio challenges         | -Difficulty hearing speaker ( $n = 4$ )  | None  | -Volume on low $(n = 1)$   |
|                          | -Unable to connect to audio $(n = 1)$  |   | -Unable to connect to audio $(n = 1)$                            |
| Video challenges         | -One screen freeze $(n = 2)$<br>-Screen freeze + video reboot $(n = 1)$<br>-Recurring screen freezing $(n = 1)$              | None  | -Difficulty turning on video ( $n = 1$ )                         |
| Audio/video<br>delay     | (27 sessions)  | None  | None   |
| Difficulty<br>logging in | -Received incorrect link $(n = 1)$   | -Unable to exit waiting room into session $(n = 1)$<br>- Participant wanted to change devices $(n = 1)$   | -Participant stuck in waiting room, instructor unaware $(n = 1)$ |
| Disruptions              | -Misc. interruptions (cause unknown) ( <i>n</i> = 3)<br>-People in the house ( <i>n</i> = 3)<br>-Phone alarm ( <i>n</i> = 1) | -Computer updated ( $n = 1$ )   | None   |
| Internet<br>connectivity | -Dropped call(s) (One/session $n = 10$ , two/<br>session $n = 1$ )   | <ul> <li>Difficulty connecting to internet (n = 3)</li> <li>Dropped call(s) (one/session n = 2, two/session n = 1, nine/session n = 1)</li> </ul> | -Difficulty connecting to internet $(n = 3)$                     |

|        | Outcome         | Baseline $(n = 15)$ Post $(n = 15)$ (mean $\pm$ sd)(mean $\pm$ sd) | Post ( <i>n</i> = 15) | $\frac{\text{Follow-up } (n = 15^{\circ})}{(\text{mean } \pm \text{ sd})}$ | $\frac{\text{Effect size}}{{\eta_P}^2}$ (for normal distribution) |
|--------|-----------------|--|-----------------------|--|---|
|        |                 |  | (mean ± sd)           |  |   |
|        |                 |  |                       |  | W (for non-normal distribution)                                   |
| CDQ-24 | Stigma *        | (11.467 ± 6.278)   | (7.667 ± 6.253)       | (8.600 ± 6.479)  | $\eta_{\rm P}^2 = 0.309$  |
|        | Pain            | (4.867 ± 3.357)  | $(3.600 \pm 2.324)$   | (3.467 ± 2.475)  | $\eta_{\rm P}^2 = 0.129$  |
|        | ADLs            | $(10.600 \pm 4.188)$   | (9.533 ± 3.114)       | (10.267 ± 4.183)   | W = 0.084   |
|        | EWB *           | (6.000 ± 3.855)  | (3.467 ± 2.800)       | (4.133 ± 3.335)  | $\eta_{\rm P}^2 = 0.300$  |
|        | Social          | $(2.267 \pm 2.344)$  | $(1.667 \pm 2.267)$   | $(2.600 \pm 2.501)$  | W = 0.0760  |
|        | Total *         | (35.200 ± 14.304)  | (25.933 ± 12.418)     | (29.067 ± 14.621)  | ${\eta_{P}}^{2} = 0.316$  |
| SF-36  | Physical Funct. | (57.000 ± 16.013)  | (56.333 ± 20.131)     | (54.000 ± 22.536)  | $\eta_{\rm P}^2 = 0.023$  |
|        | RLPH            | (31.667 ± 30.570)  | (30.000 ± 34.330)     | (38.333 ± 37.639)  | W = 0.011   |
|        | RLEP            | $(60.000 \pm 44.005)$  | (80.000 ± 37.374)     | (71.111 ± 45.192)  | W = 0.113   |
|        | General         | (62.000 ± 22.504)  | (56.000 ± 25.579)     | (56.667 ± 22.414)  | W = 0.106   |
|        | Fatigue         | (48.667 ± 18.074)  | (52.667 ± 17.715)     | (47.000 ± 18.107)  | $\eta_{\rm P}^2 = 0.127$  |
|        | Social          | (75.000 ± 18.900)  | (73.333 ± 21.582)     | (65.000 ± 24.640)  | $\eta_{\rm P}^2 = 0.079$  |
|        | Pain            | (41.500 ± 28.075)  | (52.500 ± 26.271)     | (43.333 ± 21.395)  | $\eta_{\rm P}^2 = 0.186$  |
|        | EWB             | (68.800 ± 16.984)  | (74.133 ± 13.169)     | (69.333 ± 16.676)  | $\eta_{\rm P}^2 = 0.170$  |
| TWSTRS | Severity *      | (18.467 ± 5.097)   | (15.867 ± 6.490)      | (16.333 ± 5.802)   | $\eta_{\rm P}^2 = 0.252$  |
|        | Disability      | (14.200 ± 5.570)   | (14.933 ± 5.147)      | (15.533 ± 5.235)   | $\eta_{P}^{2} = 0.153$  |
|        | Pain            | (10.583 ± 5.203)   | (10.300 ± 4.827)      | (11.283 ± 5.148)   | $\eta_{\rm P}^2 = 0.060$  |
|        | Total           | (43.250 ± 12.784)  | (41.100 ± 14.16)      | (43.150 ± 13.817)  | $\eta_{\rm P}^2 = 0.066$  |
| PSQI   | Quality         | (0.933 ± 0.704)  | (1.133 ± 0.516)       | (1.200 ± 0.561)  | W = 0.061   |
|        | Latency         | (1.267 ± 1.100)  | (1.133 ± 0.915)       | (1.200 ± 1.146)  | W = 0.006   |
|        | Duration        | (0.733 ± 0.884)  | (0.933 ± 0.703)       | (0.933 ± 1.033)  | W = 0.043   |
|        | Efficiency      | (0.600 ± 0.910)  | (0.800 ± 0.94)1       | (1.000 ± 1.195)  | W = 0.054   |
|        | Disturbance     | (1.267 ± 0.704)  | $(1.000 \pm 0.535)$   | (1.133 ± 0.640)  | W = 0.160   |
|        | Med. use        | (1.333 ± 1.345)  | (1.200 ± 1.373)       | (0.867 ± 1.366)  | W = 0.200   |
|        | Daytime Dysf.   | $(1.000 \pm 0.535)$  | $(1.000 \pm 0.655)$   | (0.933 ± 0.800)  | W = 0.007   |
|        | Total           | (7.133 ± 3.962)  | (7.200 ± 2.883)       | (7.267 ± 3.807)  | $\eta_{\rm P}^2 = 0.001$  |
| FTSTS  |                 | (13.919 ± 4.066)   | (13.617 ± 5.239)      | (12.117 ± 2.898)   | W = 0.093   |
| BAI    |                 | (11.133 ± 9.782)   | (10.400 ± 8.781)      | (10.733 ± 9.528)   | W = 0.011   |
| BDI    |                 | (8.600 ± 7.772)  | (7.333 ± 6.943)       | (9.333 ± 8.095)  | W = 0.101   |
|        |                 |  |                       |  |   |

Abbreviations: BAI, beck anxiety inventory; BDI, beck depression inventory; CDQ-24, craniocervical dystonia questionnaire-24, Daytime dysf., daytime dysfunction, EWB, emotional wellbeing; FTSTS, five times sit to stand, Physical funct., physical function, PSQI, pittsburgh sleep quality index; RLEP, role limitations due to emotional problems; RLPH, role limitation due to physical health, and SF-36, 36-Item Short Form Survey Instrument, TWSTRS, toronto western spasmodic torticollis rating scales. Asterisks identify statistically significant differences (p < .05).



**FIGURE 3** Box plot with individual data points (with a differ color for each participant) for total scores on the CDQ-24 across the three different time points (baseline, post-intervention, and follow-up). The error bars represent one standard deviation. Statistically significant differences (p < 0.05) are indicated with \*.

secondary analysis can be found in the **Supplementary Tables S3**, **S4** as well as **Supplementary Figures S1**, **S2**.

### DISCUSSION

To our knowledge this study is the first to establish the feasibility and safety of a tele-yoga intervention for individuals with CD. Notably, it is also one of the few studies investigating a nonpharmaceutical intervention for individuals with CD.

### Safety

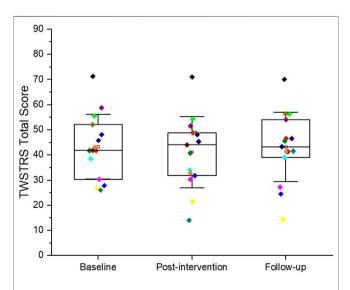
This study showed that a tele-yoga intervention for individuals with CD is safe, similar to previous findings showing yoga to be generally as safe as other exercise (39). Another synchronous tele-yoga study for individuals with chronic conditions also reported the intervention was safe (23). Asynchronous yoga interventions using pre-recorded videos also reported the interventions to be safe with no significant adverse events (40–42). For individuals with movement disorders such as PD, in-person yoga interventions have also not reported any significant adverse events (13, 43, 44). Thus, our results suggest that, as previously reported, yoga is a safe intervention that can be provided remotely *via* videoconference.

While no major adverse events occurred, some participants experienced varied one-point increases in pain during specific postures (e.g., side bends, chair posture, forward fold, cat/cow, warrior II, and baby cobra) which attenuated with modification or elimination of the relevant posture. This finding is similar to a yoga study involving individuals with PD who demonstrated knee pain in specific postures which resolved with posture modification (13). This emphasizes the importance of individualization of the yoga intervention. The one-on-one nature of this intervention allowed the instructor to accommodate each participant's reaction to the postures by modifying postures, slowing the speed of the session, and eliminating specific postures when needed. With these modifications, participants reported that their pain returned to baseline levels within an acceptable time. Future studies should investigate whether this individualized approach is more beneficial than group interventions where personalized feedback and instructors are not always possible.

### Feasibility

The current study showed that despite technological challenges, adherence was high and 14 participants achieved 80% or more of the intended dosage. This is in contrast to previous studies demonstrating that technological challenges impeded high adherence. Specifically, previous studies attributed feasibility challenges to scheduling difficulties (22), specific characteristics of the videoconferencing platform used (23), and technological literacy (23). We addressed those challenges by choosing to deliver the intervention individually according to the participant's availability, and by utilizing a readily available videoconferencing platform with an easy user interface. It must also be noted that this study took place during the Sars-CoV-2 pandemic in which the availability and use of videoconferencing platforms increased significantly. This general global increase in technological literacy may have impacted technological literacy of the participants and may positively impact future tele-rehabilitation studies.

Another factor that may have positively impacted feasibility was that the yoga instructor informed participants that challenges may occur and instructed them on how to navigate them if they arose. Additionally, the instructor modified the yoga sessions to accommodate video/audio delay by speaking in shorter bouts and



**FIGURE 4** Box plot with individual data points (with a differ color for each participant) for total scores on the TWSTRS across the three different time points (baseline, post-intervention, and follow-up). The error bars represent one standard deviation.

providing fewer cues, emphasizing safety cues. Only one participant reported that the technological challenges negatively impacted their feelings about the yoga classes.

Interestingly, while intervention adherence was high (93%) and assessment session adherence was also high with only one participant missing the final assessment session, yoga practice over the follow-up period was inconsistent. It is important to note that while participants were strongly encouraged to continue with some type of yoga practice over the 6-weeks follow-up period, similar to what they had completed during the intervention period, they were not explicitly instructed to practice for a set frequency or dosage. Nor were participants provided with any resources such as videos or handouts to practice. This may have contributed to the inconsistency of practice, as some participants noted that they did not practice because they did not have access to an instructor. Future studies could address this by providing a target frequency and dosage for yoga practice during the followup as well as by providing participants with practice resources such as videos, handouts, or contact information for other qualified voga instructors. Implementing these steps may help participant continue the yoga practice after the study has stopped and serve as a stepping-stone for positive behavior change.

### **Preliminary Efficacy**

Interestingly, statistically significant improvements in diseasespecific quality of life (i.e., CDQ-24 stigma, emotional well-being, and total score) as well as in motor symptom severity as assessed by the TWSTRS were observed in the current study. Both the primary analysis including all participants and the secondary analysis including only participants who did not receive botulinum toxin injections over the course of the study support these findings. While there is limited information related to complementary therapies for individuals with CD, there appears to be evidence that physical therapy and exercise can lead to improvements in symptom severity (45-49). Other spheres of quality of life and TWSTRS sub-scores also exhibited moderate effect sizes, as supported in both the primary and secondary analyses, such that future studies could be powered to specifically examine the effect of tele-yoga on these clinically important factors. The effect sizes for sleep, physical function, anxiety, and depression were very small in the current study. However, our study sample did not exhibit any significant impairments in those spheres at baseline. Therefore, tele-yoga may still benefit individuals with CD that have those issues but future studies with more targeted inclusion criteria are needed. In fact, yoga has been shown to improve sleep quality (50), physical function (44), anxiety (13, 51), and depression (13, 51) in other populations.

It is important to note that this study did not show any significant improvements in pain either through the TWSTRS or SF-36. Pain is one of the major contributors to poor quality of life in people with CD and is an important symptom to address (52). The findings found here may be attributed to the small sample size as this study was not powered to assess the efficacy of the intervention on pain. Additionally, it is possible that the overall dosage of the intervention was too low to significantly improve pain. It is important to note that several studies in other populations have demonstrated the beneficial effect of yoga practice for pain management (53). Therefore, while the current study did not observe significant improvement in pain, future studies are needed to better establish the efficacy of a tele-yoga intervention on pain in people with CD. Additionally, future studies should also examine the interaction between botulinum toxin injections, which have been shown to significantly reduce pain in people with CD (6), and tele-yoga to identify whether one can potentiate the effect of the other.

### Limitations

We acknowledge that this study has limitations. This was a nonrandomized, single group, unblinded study with a small sample size. The primary purpose of this study was to assess safety and feasibility across a variety of individuals with CD and, as such, a pragmatic recruitment approach was implemented. Participants were included regardless of disease severity, disease duration, and botulinum injection status. These factors along with other confounding factors such as age and sex were not accounted for in the primary analysis as this was not a primary aim of the study. Yet, despite this heterogeneity in participant characteristics, the intervention was safe and feasible. Future randomized control trials should more specifically address confounding factors either through eligibility criteria or analysis. It is also important to note that while participants were asked to confirm that they had received a diagnosis of CD from a neurologist, this was not corroborated by a physician or with medical documentation. There may also be a selection bias such that individuals comfortable with technology and already engaged in or interested in physical activity may have expressed greater interest in study participation. We also acknowledge that the yoga instructor's scheduling flexibility along with the Sars CoV-2 pandemic and associated stay at home practices may have positively impacted adherence. Taken together these limitations indicate that results, especially those relating to preliminary efficacy, should be considered cautiously. Nonetheless, the results of this study are encouraging and can be used to power a larger randomized controlled study.

### **Future Implications**

As mentioned, current treatment strategies for individuals with CD focus on alleviating abnormal head postures through botulinum toxin injections but a significant proportion of patients are not satisfied with the outcome of this intervention. Developing telerehabilitation approaches that would lead to improved clinical outcomes, greater access to knowledgeable providers, and minimize overall burden could drastically change the prognosis of individuals diagnosed with CD. One could envision a hybrid model of care where patients can get in-person treatment (e.g., botulinum toxin injections) and virtual care with a multi-disciplinary team of physical and occupational therapists, psychologists, and others. To achieve this, future randomized trials informed by the information gathered here are warranted.

### Conclusion

This study shows that one-on-one tele-yoga is safe and feasible for individuals with CD. Furthermore, preliminary effectiveness shows statistically significant improvements in quality of life and symptom severity indicating that this complementary approach should be further studied for the management of individuals with CD.

# DATA AVAILABILITY STATEMENT

The datasets presented in this study can be found in online repositories. The names of the repository/repositories and accession number(s) can be found below: 10.7303/syn25891139 synapse.org.

# **ETHICS STATEMENT**

The studies involving human participants were reviewed and approved by Institutional Review Board at Rutgers University. The patients/participants provided their written informed consent to participate in this study.

# **AUTHOR CONTRIBUTIONS**

AJ-P participated in the research project conceptualization, methodology, organization, and execution (contacted/screened interested participants, conducted assessment sessions, and implemented intervention sessions); was involved in formal statistical analysis, design, execution, and review/critique; and assisted in writing the first draft of manuscript and reviewing and revising subsequent drafts. J-FD participated in the research project conceptualization, methodology, organization, and execution (supervising and assisting AJ-P) and project administration; was involved in formal statistical analysis design, execution, and review/critique; and co-wrote wrote the first draft of manuscript with AJ-P and supervised/participated in reviewing, editing and revising subsequent drafts.

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# **CONFLICT OF INTEREST**

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The remaining author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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# SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: https://www.frontierspartnerships.org/articles/10.3389/dyst.2021. 10015/full#supplementary-material

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