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ORIGINAL RESEARCH ARTICLE

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The immediate effect of peripheral stimulation on cervical dystonia disease outcomes: single-case experimental design

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ABSTRACT

Introduction: Limited cervical range of motion (CROM) and pain are among the most common symptoms of cervical dystonia (CD), which are treated with botulinum neurotoxin (BoNT) injections. However, adjunctive interventions may offer additional benefit. Therefore, this study evaluated the immediate effects of peripheral stimulation comprising conventional transcutaneous electrical nerve stimulation (TENS), muscle stimulation (NMES), and muscle vibration on CROM and pain in patients with CD.

Methods: A single-case experiment design (SCED), alternating treatment design was conducted with 19 CD patients, recruited at the Antwerp University Hospital (UZA). Participants randomly received conventional TENS, NMES and muscle vibration on three different days. Main outcome measures were CROM, measured using a 3D-stereophotogrammetric scanning system (3dMD Inc., GA, USA) and pain, quantified by Visual Analog Scale (VAS).

Results: Mean age of participants was 59.87 ± 8.9 years. Baseline CROM for rotation ranged from 87° to 137.6° and from 79° to 132.4° for flexion/extension. Improvements in neck rotation were observed in 5 (33.3%) participants after conventional TENS, 8 (42.1%) after NMES, and 8 (42.1%) after muscle vibration. Similarly, improvement in flexion and extension was observed in 10 (55.6%) after conventional TENS, 8 (53.3%) after NMES, and 7 (46.7%) after muscle vibration. Significant reductions in pain shortly after the three interventions were observed in patients experiencing pain.

Conclusions: These findings suggest that peripheral stimulation may provide immediate symptomatic relief in CD, particularly for improving CROM and reducing pain. While promising, these results should be considered as exploratory and warrant further investigation in larger, controlled studies.

Keywords: Cervical dystonia, Electric stimulation, Muscle vibration, Transcutaneous electric nerve stimulation, TENS

What's already known about this topic?

- *The dystonic contraction of cervical muscles resulted in limited cervical range of motion (CROM), pain, and disability.*
- *Some patients reported a sub-optimal level of satisfaction with the intramuscular injections of botulinum neurotoxin (BoNT), which is the mainstay of treatment.*

What does the study add?

- *Peripheral stimulation comprising conventional transcutaneous electrical nerve stimulation (TENS), muscle stimulation (NMES), and muscle vibration provided immediate symptomatic relief, particularly by improving cervical mobility and reducing pain.*

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Introduction

Cervical dystonia (CD) is characterized by involuntary sustained or intermittent muscle contractions leading to repetitive twisting movements or sustained positions (1). The dystonic contraction of cervical muscles leads to abnormal head posture, which can result in limited cervical range of motion (CROM), pain, and myofascial contractures (2). Pain, when present, significantly contributes to functional limitation (3), disability (4), and serves as the primary reason to seek treatment.

The primary choice of treatment for CD is the administration of intramuscular injections of botulinum neurotoxin (BoNT) (5). The injection is repeated every three to four months and results in significant improvement in the severity of torticollis and head position (6) and health-related quality of life (7). However, some patients reported a sub-optimal level of satisfaction (8), which may be due to temporarily reduced neck muscle strength and muscle tone, which usually returned to pre-injection levels within two months (9).

In addition to BoNT injection, alternative non-invasive interventions are emerging. A review finding on the effectiveness of physiotherapy on CD outcome highlighted its beneficial effect (10). Non-invasive peripheral stimulation techniques such as transcutaneous electrical nerve stimulation (TENS), muscle stimulation (NMES), and muscle vibration target the modulation of neural mechanisms and are emerging as an alternative treatment. TENS is a transcutaneous use of electrical stimulation to produce analgesia with a portable device which generates mild pulsed electrical stimulation using electrode pads with varies frequency, intensity, and duration (11). The intensity can be of different levels, including sub-sensory, sensory, motor, and noxious (12). Conventional TENS is applied with high frequency (50-130 Hz), low intensity (comfortable and not painful), and small pulse duration (50-200 µs). In muscle stimulation (motor level TENS), the pulse intensity is increased to produce motor contraction (13).

Studies reported the therapeutic use of TENS for symptomatic relief of pain (14-16), for functional movement disorders (17), stroke (18), and writer's cramp dystonia (19). TENS modulates nociceptive input at peripheral, segmental, and extra-segmental sites. Stimulation of low-threshold large-diameter non-noxious cutaneous afferents reduces the activity of central nociceptive transmission cells (20). Electrical muscle stimulation and Vibro-Tactile Stimulation (VTS) are the other alternative treatment approaches, where the signals from stimulated muscles modulate cortical activity (21).

Despite advances in the management of CD, such as BoNT injections, there remains a significant gap in understanding the efficacy and clinical applicability of non-invasive interventions like TENS, muscle stimulation, and vibration. Previous studies on peripheral stimulation have largely focused on TENS or reported on single-case observations, leaving a lack of robust evidence to guide sustained management strategies. Thus, this study aimed to evaluate the immediate effects of peripheral stimulation (conventional TENS, electrical muscle stimulation, and muscle vibration) on CROM and pain in CD.

Methods

Study design

A Single Case Experimental Design (SCED), alternating treatment design was used. A Single-Case Reporting guideline In BEhavioral intervention (SCRIBE) was followed (22). Participants randomly received the three types of peripheral stimulation on three different days. The primary outcome measure, CROM, was evaluated at baseline (T0), immediately after intervention (T1), and 10 minutes post-intervention (T2). Pain was assessed before and after the interventions.

Participants

Adult participants with the diagnosis of CD were informed about the study by providing an information leaflet during their routine follow-up visits for BoNT injections. Patients with additional neurological conditions, previous cervical spine surgery, rheumatoid arthritis, head trauma or skin conditions at the site of stimulation were excluded from the study. Detailed demographic and clinical characteristics of participants are presented in Table 1.

Setting

Participants were recruited from a tertiary center for movement disorders, the neurology department of the Antwerp University Hospital, Belgium (UZA).

Measures and materials

Volunteer patients were scheduled for the experimental session either within a week after a new round of BoNT injection or immediately before their next treatment follow-up. During this time, it is assumed that the effect of BoNT had worn off after administration (23). At baseline, CROM (full range of neck rotation and neck flexion/extension), the Toronto Western Spasmodic Rating Scale (TWSTRS), Dystonia Non-Motor Symptoms Questionnaire (DNMSQuest), Tsui scale, and pain, measured by the Visual Analog Scale (VAS), were taken. Similarly, at end-line, CROM (full range of neck rotation and neck flexion/extension), VAS for pain, and the Global Perceived Effect scale (GPE) were used to assess participants' subjective response for intervention. TWSTRS is a disease-specific rating scale that covers physical findings (severity subscale), disability, and pain. The severity subscale consists of maximal excursion, duration factor, effect of sensory trick, shoulder elevation/anterior displacement, range of motion, and time. The sum of items amounts to a maximum score of 35, with a higher score indicating the severity of the symptoms. The reliability (24) and validity have been established (25). The DNMSQuest is a 14-item self- administered questionnaire to assess the presence of a range of non-motor symptoms (NMS) during the past month (26). The total score is the sum of positive responses, with a maximum possible score of 14, with a higher score indicating the severity of the symptoms. The tool was found to be reliable, reproducible, robust, and valid in clinical settings for patients with CD (26, 27). Furthermore, the severity of the tremor was measured using the Tsui scale (6). It is a 4-item scale evaluating the presence and frequency of head tremor, with the higher score indicating worsening of symptoms. The tool showed good inter-observer reliability in CD patients (28). For pain, patients were asked to mark the point on a 10-centimeter (cm) line that represents the severity of the pain or symptoms in cm. The longer the line, the greater the severity of the pain or symptom; 0 cm indicates absence of pain or symptoms. The tool has previously been used to measure outcomes in patients with cervical disease (29). A 7-point

TABLE 1 - Socio-demographic and baseline clinical characteristics of study participants

ID	Gender	Age (year)	Predominant type of CD	Duration of illness	Tremor	Total TWSTRS score	DNMS score	Baseline neck rotation (⁰)		Baseline neck flexion/ extension (⁰)		Baseline VAS score (cm)
								Mean (SD)	SEM	Mean (SD)	SEM	Mean (SD)
1	F	73	Left torticollis	20	No	35.0	7	87 (8.0)	4.6	79 (4.2)	4.2	2.5 (1.5)
2	F	59	Left torticollis	19	No	22.0	6	126.9 (8.1)	4.7	97.8 (2.5)	1.4	1.8 (0.6)
3	M	65	Left torticollis	5	No	28.0	9	87.8 (5.4)	2.7	106.8 (4.8)	2.4	1.8 (0.5)
4	F	56	Laterocollis	7	No	36.0	5	112.7 (8.7)	3.3	81.6 (14.8)	5.6	3.2 (0.1)
5	M	34	Right torticollis	2	No	21.0	5	130.4 (7.5)	2.5	120.3 (8.5)	2.9	-
6	F	73	Left torticollis	25	Yes	24.0	2	112.4 (8.4)	2.8	100 (4.6)	1.5	2 (0.7)
7	F	59	Right torticollis	9	Yes	31.0	5	131.5 (3.4)	1.4	109.8 (7.9)	2.7	4.6 (2.6)
8	F	61	Right torticollis	10	Yes	22.3	4	110.7 (8.1)	2.7	103 (19.4)	6.5	1.3(0)
9	F	58	Right torticollis	23	Yes	20.3	7	129.2 (2.6)	1.0	113.3(5.9)	2.4	3 (0.1)
10	M	64	Laterocollis	18	Yes	22.3	10	125.6 (3.2)	1.1	114.2 (10.9)	3.6	1.3 (0)
11	F	57	Right torticollis	10	Yes	16.3	3	137.6 (7.7)	2.6	116.7 (4.7)	1.6	2.7 (0.4)
12	F	59	Laterocollis	12	Yes	29.3	9	114.4 (3.0)	1.0	121.4 (9.4)	3.1	3.7 (2.6)
13	F	64	Left torticollis	4	Yes	20	5	127.2 (6.2)	2.1	110.3 (6)	2	2.4 (0.6)
14	F	55	Right torticollis	2	Yes	34	1	116.7 (9.4)	3.1	117.7 (10.5)	3.5	5.1 (0.7)
15	F	61	Right torticollis	7	No	40.25	6	92.3 (2.5)	0.8	78 (1.4)	0.5	3 (0.4)

CROM <80% of age appropriate values,-: no pain at baseline[1]
[1] ID: Identification, CD: Cervical Dystonia, TWSTRS: Toronto Western Spasmodic Torticollis Rating Scale, DNMS: Dystonia Non-Motor Symptom, VAS: Visual Analog Scale, cm: centimeter, SD: Standard Deviation, CROM: Cervical range of motion, (⁰) : angle

GPE scale rates participants' perception on how much their condition is improved or worsened. The tool is easy and quick to administer and it has several qualities. The higher score indicating slighter improvement of symptoms. The VAS scale was administered before and after each intervention. CROM was measured using a three-dimensional stereophotogrammetry system (3dMD Inc., GA, USA) (Fig. 1).

FIGURE 1 - Three-dimensional scanning using 3dMD.

CROM: Procedure for measurement

Participants were seated in a chair with their head remained fully within the field of view of the 3dMD cameras during all movements of the head and neck. The device was calibrated before each acquisition. The participants were requested to remove any jewelry (necklaces, earrings and facial piercings) that might interfere with the imaging process. If necessary, a wig cap was used to keep the ears visible without interference from hair. Blue self-adhesive markers were placed on the tip of the nose, both cheekbones, and the distal point of the processus spinosus of C7. Each recording for CROM was conducted three times with the eyes closed. Participants were instructed to move as far as possible from flexion to extension and from right to left rotation to assess maximal CROM. The 3dMD head scanners provide 360-degree full head and face coverage. It captures a range of head positions in 3D over time. Recordings were performed for a maximum of 20 seconds at a rate of 10 frames per second. From each session, the frame showing the maximal position was selected for 3D reconstruction. Per acquisition time, three 3D models were generated for further analysis: neutral, left, and right neck rotation and flexion/extension. The Vultus software, the image analysis package from 3dMD, was used for image alignment, registration, landmark placement, and calculation of coordinate points. The 3dMD scanner has been used in the clinical population involving head, face, and hand (30). Its reliability was tested in children with torticollis and found to be moderately accurate compared to the conventional tests to measure head tilt (31).

A Microsoft (MS) Excel sheet capturing the x, y, and z coordinate points and angles of each type of movement (neutral head position, left and right head rotation, and head flexion-extension) was prepared and used for the data collection process. The range of motion was calculated for rotation (x), flexion-extension (y), and lateral flexion

(z) during each of the movements. The full range of x-angle was calculated to determine the full range of rotation (angle x-neutral to the right plus angle x-neutral to the left). The full range of y-angle was calculated to determine the full range of flexion and extension (angle y-neutral flexion plus angle y-neutral extension).

Interventions

Participants were randomly assigned to receive peripheral stimulation (conventional TENS, NMES, and muscle vibration) on three separate days. Prior to each intervention, patients rated their pain, and CROM was assessed. Following the baseline measurements, the skin was prepared.

In the case of conventional TENS, two medium-adhesive electrodes were placed dorsally along the cervical spines, with one electrode placed at the level of C2 and the other at C7. A conventional 100 Hz pulse frequency was used for 20 minutes, with the voltage or amplitude increased until the patient reported intense but non-painful stimulation, without muscle contraction (13). Participants were requested to report a perceived reduction in the intensity of the stimulus during the intervention. The intensity of amplitude was maintained at a consistent level throughout the procedure. The conventional TENS activates the A-β sensory fibers, which reduces the transmission of the noxious stimulus from the C-fibers, through the spinal cord and to the higher centers (32).

For muscle stimulation, medium adhesive electrodes were applied to the antagonist muscles of the dystonic muscles, e.g., the contralateral descending part of trapezius, sternocleidomastoid, and/or splenius capitis. The positive effect of antagonist muscle stimulation in improving clinical outcomes of stroke patents had been reported (33). A combination of a low-frequency TENS and biphasic surge neuro-muscular electro stimulation (NMES) was used. Low-frequency TENS was employed to activate the antagonist muscle for two minutes, followed by a biphasic surge NMES to strengthen the antagonist muscle for six minutes (34). NMES uses electrical currents to stimulate the motor points, thereby facilitating a contraction and thus strengthening the muscle. This is mainly by mimicking the signals of the central nervous system to induce muscle contraction, which further enhances muscle control, reduces pain, and improves range of motion (35). For muscle vibration, two stimulators were applied with adhesive tape to the dystonic muscles, such as the sternocleidomastoid, trapezius, or splenius muscles. A 100Hz pulse frequency was used. The researchers were guided by the physical examination and the findings of the TWSTRS to identify the target muscle for electrode and stimulator placement.

Statistical analyses

The CROM and VAS data from the MS Excel spreadsheet were imported into Statistical Package for Social Sciences (SPSS) version 24.0 (SPSS Inc, Chicago, IL) for analysis. Descriptive statistics and percentage change in CROM were computed. The standard error of the mean from the baseline CROM measurements was calculated to define improvement after intervention. Participants were categorized as “improved” if the change in CROM was larger than the Standard Error of Mean (SEM) or “not improved” if the change in CROM was smaller than the SEM. Individual patient values were visualized in graphs to illustrate trends in CROM over time.

Results

Of the 22 participants initially enrolled in the study, three were lost in follow-up, and thus, 19 completed the three interventions. Four participants were excluded from the analysis, as they had no pain or restriction on ROM (Fig. 2). This was intended to ensure a more accurate assessment of the immediate effect. The sequence of interventions is detailed in the supplementary file (Table S1)

Socio-demographic and clinical characteristics

The predominant type of CD was torticollis, laterocollis, and tremor. The mean age of the participants was 59.87 ± 8.9 years, ranging from 34 to 73 years. The mean duration

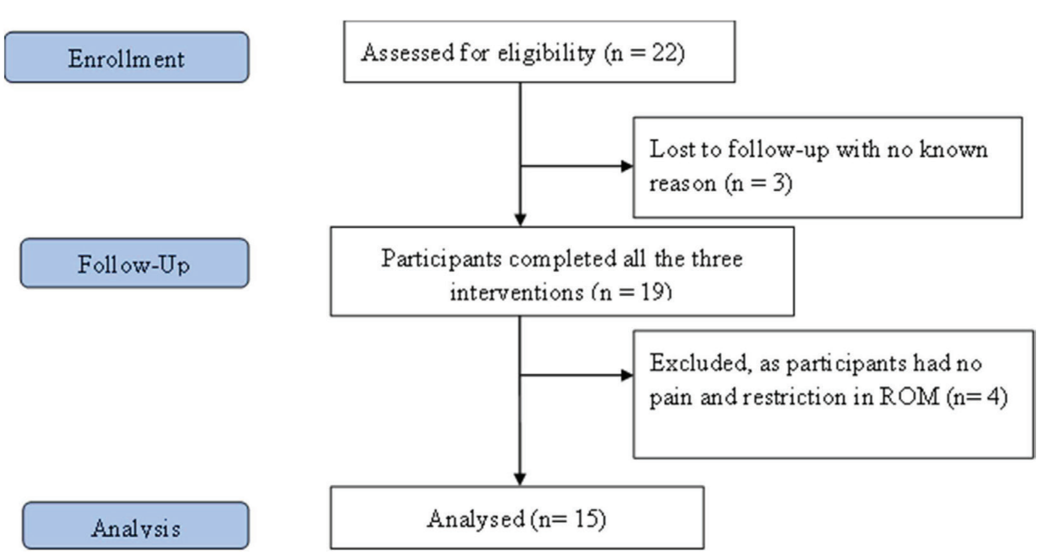

FIGURE 2 - Flow diagram showing the participants’ enrollment and follow-up status.

of the illness at the moment of intake was 8.8 years, ranging from 2 to 25 years. The baseline score of TWSTRS ranged from 16.3 to 40.3, and the DNMS score ranged from 1 to 10. About 40 % of the participants had no tremors. As to CROM, the baseline mean score of the full range of neck rotation ranged from 92.3° to 137.6°; and it ranged from 78° to 132.4° for neck flexion/extension. Compared to age-related normative values (36), six of the participants had a CROM not age-appropriate (<80% CROM of the normal age-related CROM) (Table 1).

Descriptive and one-level analysis of the outcome

The evolution of individual full range neck rotation is presented at different time points. Visual inspection of patients' neck rotation indicated variability in their responses to different interventions. Observing the individual graphs, most patients demonstrate a persistent increase in range of motion during muscle stimulation and muscle vibration immediately after the intervention. A schematic presentation indicates the variability in range of motion over different time points (Fig. 3).

Immediate response to conventional TENS

Subjective perceived effect

Fourteen (93.3%) participants reported a slight improvement of symptoms with the 7-point Likert GPE scale, and one participant (Id: 4) reported marked improvement of symptoms (Table 2).

Immediate response to pain

In participants experiencing pain, a favorable pain reduction was observed, varying from 8.3% to 100% and one participant (Id: 7) reported a worsening of pain by 23% from the baseline (Table 2).

Immediate response on CROM

Immediately following conventional TENS, the CROM changed from −9° to +22°. The increase in rotation varied from 1° to 7.1°, or 0.79% to 6.37% of the CROM in some participants. Of the six participants with decreased CROM to rotation, two showed increased neck rotation immediately following conventional TENS. A change of −23° to +23° was measured 10 minutes post-intervention. The increase in rotation varied from 1° to 23° or 1.09% to 19.5% of the CROM. However, seven participants (Id: 1, 5, 7, 10, 11, 12, and 13) had a slight reduction in neck rotation immediately following conventional TENS. The reduction ranged from −1° to −9° or −0.7% to −7.8%. Similarly, 10 minutes after the same intervention, seven participants (Id: 1, 5, 7, 10, 11, 13, and 15) had a slight reduction in ROM during rotation, ranging from −1° to -23°, or −0.7% to −18% (Table 2). Regarding full CROM flexion-extension, the CROM changed from −13° to +22° immediately after conventional TENS. The increase in ROM varied from 1° to 22° or 1% to 29% of the CROM. All the participants with a decreased CROM to flexion/extension showed improvements in mobility. Ten minutes after the intervention, the CROM to flexion/extension was increased from 1° to 17° or 1% to 22% compared to baseline CROM

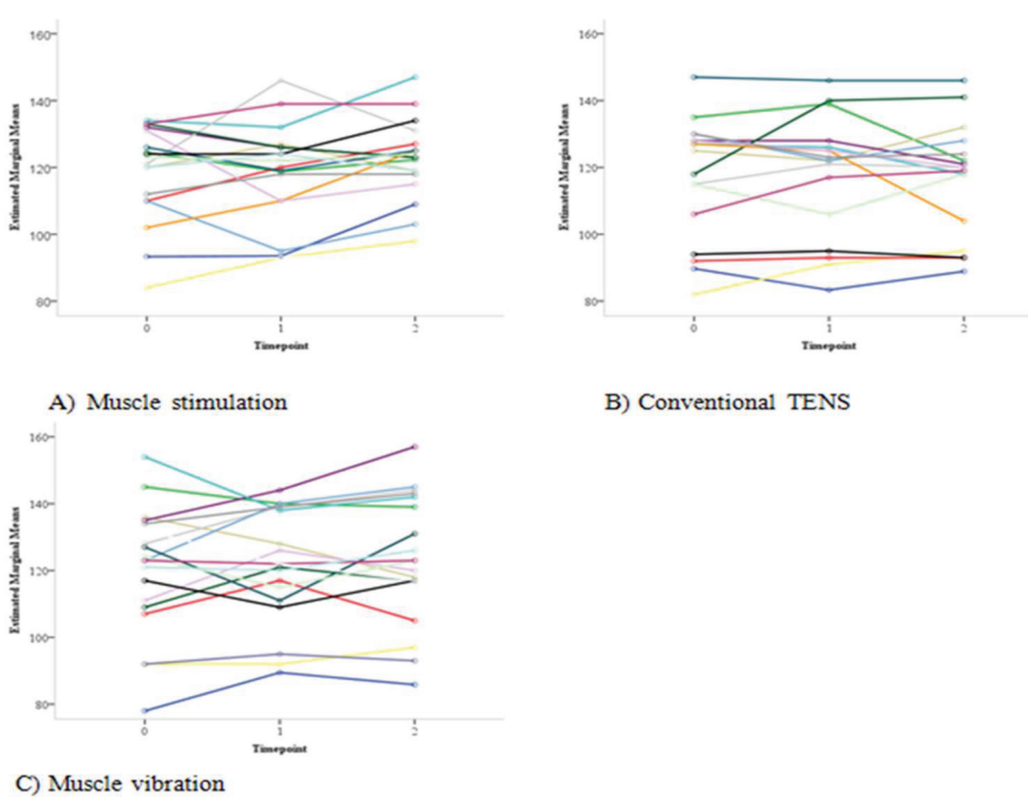

FIGURE 3 - Individual mean score of the full range of neck rotation at baseline (0), immediately post stimulation (1), 10 min. post stimulation (2) for the different interventions.

TABLE 2 - Participants' values and percentage change of CROM, pain, and subjective perceived response after conventional TENS, muscle stimulation, and muscle vibration

ID	Gender	Age (year)	Predominant type of CD	Duration of illness (year)	Tremor	TWSTRS score	Conventional TENS						NMES: neuromuscular stimulation						Muscle vibration					
							Neck rotation outcome		Neck flexion/ extension outcome		Pain (post-pre)	GPE	Neck rotation outcome		Neck flexion/ extension outcome		Pain (post-pre)	GPE	Neck rotation outcome		Neck flexion/ extension outcome		Pain (post-pre)	GPE
							Immediate	10 min post	Immediate	10 min post			Immediate	10 min post	Immediate	10 min post			Immediate	10 min post	Immediate	10 min post		
1	F	73	Left torticollis	20	No	35.0			Δ 22°, 29%	Δ 17°, 22%	∇ 0.1	3		Δ16°, 17%		Δ 5°, 6%	∇ 2.6	3	Δ 11°, 15%	Δ 8°, 10%	Δ 12°, 13%		∇ 0.3	3
2	F	59	Left torticollis	19	No	22.0		Δ 7°, 6%	Δ 3°, 3%	Δ 6°, 6%		3	Δ 7°, 6%			Δ 11°, 12%	∇ 0.6	4			Δ 9°, 9%			3
3	M	65	Left torticollis	5	No	28.0	Δ 9°, 11%	Δ 13°, 16%			∇ 0.2	3	Δ 4°, 4%	Δ 9°, 10%	Δ 9°, 9%	Δ 9°, 9%	∇ 0.4	3		Δ 5°, 5%			∇ 0.8	2
4	F	56	Laterocollis	7	No	36.0			Δ 6°, 7%	Δ 13°, 16%	∇ 0.5	4	Δ 10°, 9%	Δ 17°, 15%	Δ 29°, 40%	Δ 24°, 33%	∇ 1.8	3	Δ 10°, 9%					3
5	M	34	Right torticollis	2	No	21.0			Δ 13°, 13%	Δ 6°, 5%	∇ 0.1	3		Δ 13°, 9%	Δ 4°, 4%	Δ 4°, 4%		3	Δ 9°, 7%	Δ 14°, 11%		Δ 4°, 3%		3
6	F	73	Left torticollis	25	Yes	24.0	Δ 3°, 3%		Δ 3°, 3%	Δ 2°, 2%		3			Δ 6°, 6%	Δ 3°, 3%	∇ 1.2	3	Δ 12°, 11%	Δ 8°, 7%			∇ 1.7	3
7	F	59	Right torticollis	9	Yes	31.0			Δ 25°, 21%		Δ1.7	3					∇ 0.95	3			Δ 6°, 6%		∇ 0.4	3
8	F	61	Right torticollis	10	Yes	22.3	Δ 18°, 15%	Δ 23°, 19%	Δ 9°, 7%	Δ 13°, 10%	∇0.2	3	Δ 8°, 8%	Δ 22°, 22%	Δ 10°, 12%	Δ 13°, 16%	∇ 1	2	Δ 14°, 13%	Δ 9°, 8%	Δ 14°, 14%			4
9	F	58	Right torticollis	23	Yes	20.3						3					∇ 0.8	3		Δ 4°, 3%			∇ 0.1	2
10	M	64	Laterocollis	18	Yes	22.3					Δ1.45	3					∇ 0.25	3					∇ 0.3	3
11	F	57	Right torticollis	10	Yes	16.3						3	Δ 5°, 4%	Δ 5°, 4%				4	Δ 8°, 5%	Δ 10°, 8%				3
12	F	59	Laterocollis	12	Yes	29.3		Δ 3°, 2.6%		Δ 8°, 7%	∇ 0.5	3	Δ 6°, 5%	Δ 6°, 5%	Δ 8°, 7%		∇0.3	3					∇ 1.7	3
13	F	64	Left torticollis	4	Yes	20					∇ 1.5	3	Δ 4°, 3%				∇0.45	4					∇ 0.9	3
14	F	55	Right torticollis	2	Yes	34	Δ 11°, 10%	Δ 13°, 12%	Δ 9°, 8%		∇ 1.6	3		Δ 1°, 0.8%			Δ3.2	3		Δ 6°, 5%			∇ 1.9	3
15	F	61	Right torticollis	7	No	40.25	Δ 2°, 2%		Δ 4°, 5%	Δ 3°, 4%	∇ 2.6	3	Δ 1°, 1%	Δ 3°, 3%	Δ 5°, 6%	Δ 5°, 6%	∇1.3	2	Δ 3°, 3%	Δ 2°, 2%	Δ 4°, 5%	Δ 3°, 4%	∇ 0.3	3

Δ: increase in the value of the angle of movement and percentage change observed, improved, not improved 10 mm improvement in VAS score for pain: improved, not improved, no pain
Subjective improvement on the Likert scale: 1-3: symptoms have slightly improved to disappear, 4-7: not improved[2]

[2]ID: Identification, CD: Cervical Dystonia, GPE: Global Perceived Scale, NMES: Neuromuscular Electrical Stimulation, TWSTRS: Toronto Western Spasmodic Torticollis Rating Scale, , TENS: Transcutaneous Electric Nerve Stimulation, (0) : angle, Δ : Increase in the value of angle of movement, Δ: Reduction in the value of angle of movement

(Table 2). Overall, improvements of CROM were observed in five (33.3%) and nine (60%) participants immediately after conventional TENS for neck rotation and neck flexion/extension, respectively (Fig. 4).

Immediate response to neuromotor electrical stimulation

Subjective perceived effect

Ten (66.7%) participants reported slight improvement of symptoms, two participants (Id: 8 and 15) reported no change of symptoms, and three participants (Id: 2, 11, and 13) reported marked improvement of symptoms (Table 2).

Immediate response to pain

In participants experiencing pain, a favorable pain reduction was observed, varying from 19% to 100%. One participant (Id: 14) reported a worsening of pain by 105% from the baseline (Table 2).

Immediate response on CROM

Immediately following muscle stimulation, the CROM changed from −21° to +10°. The increase in rotation varied from 1^0 to 10^0, or 1% to 9% of the CROM in some participants. A change of −16° to +23° was measured 10 minutes post-intervention. The increase in rotation varied from 3° to 22.5° or 3.26% to 23% of the CROM. However, seven participants (Id: 2, 5, 6, 7, 9, 10, and 13) had a decrease in neck rotation immediately following muscle stimulation, ranging from −1° to −21°, or −0.8% to −16%. Similarly, 10 minutes after the same intervention, five participants (Id: 6, 7, 9, 10, and 13) had decreased in ROM during rotation, ranging from −1° to −16°, or −0.8% to −12% (Table 2). Regarding full CROM flexion-extension, the CROM changed from −24° to +29° immediately after muscle stimulation. The increase in ROM varied from 1° to 40° or 0.8% to 40% of the CROM. Ten minutes after the intervention, the CROM to flexion/extension was increased from 1° to 24° or 1% to 33% compared to baseline CROM (Table 2). Overall, improvements of CROM were observed in eight (53.3%) and seven (46.67%) participants immediately after NMES for neck rotation and neck flexion/extension, respectively (Fig. 4).

Immediate response to muscle vibration

Subjective perceived effect

Eleven (73.3%) participants reported a slight improvement in symptoms, two participants (Id: 3 and 9) reported no change in symptoms, and one participant (Id: 8) reported marked improvement of symptoms (Table 2).

Immediate response to pain

In participants experiencing pain, a favorable pain reduction was observed, varying from 3% to 73%. One participant (Id: 2) reported a worsening of pain by 8% from the baseline, while two participants (Id: 4 and 8) reported no change in pain levels (Table 2).

Immediate response on CROM

Immediately following muscle vibration, the CROM changed from −16° to +15°. The increase in rotation varied from 3° to 15°, or 3.26% to 13.5% of the CROM in some participants. A change of −13° to +22° was measured 10 minutes post-intervention. The increase in rotation varied from 1° to 22.5° or 1% to 23% of the CROM. However, seven participants (Id: 2, 7, 9, 10, 12, 13, and 14) had a decrease in neck rotation immediately following muscle vibration, ranging from −1° to −16°, or −0.8% to −12.6%. Similarly, 10 minutes after the same intervention, three participants (Id: 2, 10, and 13) had decreased in ROM during rotation, ranging from −1° to −18°, or −0.8% to 13% (Table 2). Regarding full CROM flexion/extension, the CROM changed from −19° to +16.22° immediately after muscle vibration. The increase in ROM varied from 2° to 16.22° or 1.69% to 22.4% of the CROM. Ten minutes after the intervention, the CROM to flexion/extension was increased from 1° to 12° or 1-13% compared to baseline CROM (Table 2). Overall, improvements of CROM were observed in seven (46.67%) and five (33.3%) participants immediately after muscle vibration for neck rotation and neck flexion/extension, respectively (Fig. 4).

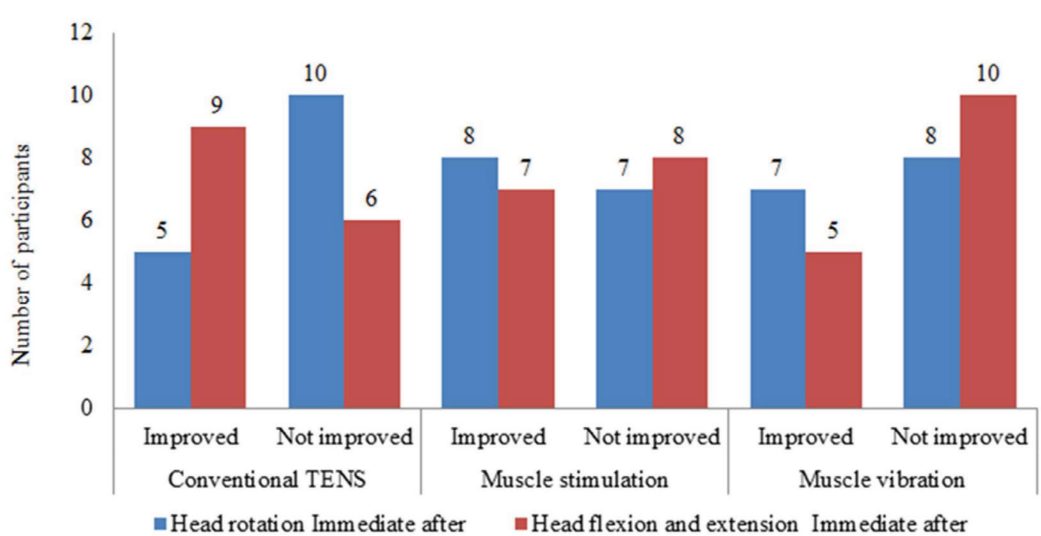

FIGURE 4 - Improvement status in CROM neck rotation and neck flexion/extension immediate after the interventions.

Tremor presence had little immediate response on neck rotation improvement after TENS, 33.3% improvement was observed in participants with and without tremor. However, it was observed that a higher number of participants (66.67% with no tremor and 44.4% with tremor) had an improvement in neck rotation immediately after muscle stimulation. Similarly, 33.3% with tremor and 66.67% without tremor had an improvement in neck rotation immediately after muscle vibration. In participants below the age-appropriate CROM, NMES and muscle vibration showed the best improvements in ROM. Regarding the relationship between pain and neck rotation, two participants (11.1%) following conventional TENS, one participant (5.3%) after muscle stimulation, and two participants (10.5%) after muscle vibration achieved minimal clinical important changes in pain score (37), along with corresponding improvements in neck rotation.

As a summary, fourteen (93.3%), ten (66.7%), and eleven (73.3%) participants reported a slight improvement of symptom with the 7-point Likert GPE scale. Favorable pain reduction was observed, varying from 3% to 100% immediately after conventional TENS, NMES, and muscle vibration, respectively. Similarly, improvements in neck rotation were observed in 5 (33.3%) participants after conventional TENS, 8 (42.1%) after NMES, and 8 (42.1%) after muscle vibration. Furthermore, better improvement in neck rotation immediately after NMES and muscle vibration was observed in participants without head tremor.

Discussion

The aim of this study was to explore the immediate responses of peripheral stimulation comprising conventional TENS, NMES, and muscle vibration on CROM and pain in CD patients. The response/ influence of peripheral nerve stimulation, specifically TENS, has been the subject of clinical research since it was developed (13, 16). In this study, participants demonstrated diverse responses in neck rotation and pain for different types of interventions. The immediate response of conventional TENS, NMES, and muscle vibration are similar in improving CROM or reducing pain. In this population with diverse symptomatology, 83% of the participants with limited CROM showed marked improvements following the 3 types of intervention. More participants showed pain reduction following TENS and NMES than following muscle vibration.

In healthy individuals, studies reported the beneficial effect of peripheral stimulation on ROM outcomes. One review reported an improvement in muscle flexibility, blood flow, and muscle relaxation in healthy young adults (38), while another found increased lumbar flexion and curvature following the use of conventional TENS (39). In patients with musculoskeletal problems, studies reported mixed findings on ROM outcomes. An increase in active shoulder range of motion was observed in patients with subacromial pain syndrome following TENS (40). Similarly, the study by Shamsi et al. reported a significant increase in the ankle range of motion over time following TENS application (41).

Studies reported the beneficial effects of peripheral stimulation on neurological and stroke patients' disease outcomes. In chronic stroke patients, a combination of TENS on therapeutic exercise and sham taping reduced muscle spasticity and improved functional activity (42). An improvement in dystonic motor symptoms was observed following TENS application in writer's cramp dystonia (19). Furthermore, an improvement in motor plasticity (43) and an increase in muscle power and a decrease in spasticity had been reported (44).

The effect of TENS on neck pain has been investigated in a diverse group of patients. A review evaluating the efficacy of TENS on acute and chronic neck pain reported a reduction in pain intensity after the treatment (15). Consistent with the current finding, a study by Maayah et al. (45) revealed a pain relief of TENS for mild neck pain. Similarly, a study by Xu et al. demonstrated the potential short-term benefit of vibro-tactile stimulation (VTS) in the reduction of pain associated with CD (46). Furthermore, a case study on a CD patient reported a pain relief effect and alleviation in the extent of dystonic neck movements using VTS on the neck muscle (21). In contrast, the review report indicated that the short-term analgesic effect of conventional TENS remains uncertain (14), and a study by Sahin et al. did not reveal a significant difference in pain by conventional TENS (47). Multiple mechanisms involving complex brain and spinal cord networks are implicated. VTS activates cutaneous and muscular proprioceptive and tactile receptors (48). Furthermore, it modulates pain signal transmission (49) and the firing of cerebellar and cortical projection neural networks (50). As a strength, participants included in the study were evaluated by a neurologist specialized in movement disorders to minimize misdiagnosis. Precaution was taken in scheduling the experimental session to avoid the effect of BoNT, which was either two weeks before or one week after a new round of BoNT injection. The investigators received training, including on how to conduct 3dMD image scanning, image analysis, and how to apply the treatments according to the test protocol. A daily calibration of the 3dMD system was done immediately before each test day. To maximize the reliability of the findings, repeated measures were taken for each type of movement, and a mean value was calculated for each time point. Different limitations were recognized in this study. First, though 3dMD stereophotogrammetric imaging was used for the evaluation of different study outcomes, its validity and reliability in measuring CROM is not well established in adult CD patients. Secondly, this study assessed the short-term effect of TENS treatment, but no evidence is available regarding the longitudinal effect of the intervention. Third, the limited number of participants prevented drawing valid conclusions on the effect of the treatment at group level. Fourth, blinding of participants and using controls was not possible in this study. The lack of established evidence for evaluating the minimal clinically important difference in CROM and VAS in CD patients made it impossible to draw valid conclusions on the effect of the treatment in achieving meaningful clinical changes in the outcomes of interest. Furthermore, the potential for placebo effects cannot be ruled out, given the subjective nature of pain assessment and the absence of blinding.

In conclusion, over one-third of participants demonstrated immediate improvements in neck rotation and neck flexion-extension across all three treatment modalities. Short-term pain reductions were observed following muscle stimulation and vibration. Future research should explore the optimal frequency, duration, and combination of these peripheral stimulation modalities to maximize therapeutic benefit. Larger, controlled studies are needed to assess their integration with existing treatments such as BoNT injections, their long-term effects, and their clinically meaningful changes in pain and CROM

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