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Review

Therapeutic Exercise Holds the Key to Improve Hand and Upper Limbs Dystonia Rehabilitation Programs: A Systematic Review

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Abstract

Upper limb dystonia is a focal locomotion disorder affecting arm, forearm, and hand muscles, causing abnormal movements given by repeated, steady, and intercontinuous contractions. There are different types of dystonia and the multifaced nature of this pathology is challenging in the treatment management, leading to a worsening of affected patients' life quality, mainly from a psychological point of view, but also from a functional perspective. This work examines the present literature regarding upper limb dystonia rehabilitation and treatment with a glance at recent approaches and new treatment strategies. This systematic review was carried out according to the PRISMA statement. The investigation used PubMed, Cochrane, and Google Scholar, including papers from the last ten years. The search yielded 1608 records, all undergone careful screening. A total of 15 papers were selected. The data highlight the importance of combined and customized treatments. The most common strategy included botulinum toxin. Evidence has shown the association between botulinum toxin and therapeutic exercise or occupational therapy. Other approaches involved: tDCS, rTMS and orthosis use, always combined with rehabilitation programs. This pathology requires a multidimensional approach combining personalized therapeutic exercise and other treatments. Nevertheless, further investigations are needed, with a larger population and standardized outcomes to improve dystonia patients' quality of life and motor abilities.

Keywords

Hand dystonia; focal dystonia; writer's cramp; upper limbs dystonia; rehabilitation; therapeutic exercise

1. Introduction

Dystonia is a movement disorder characterized by repeated, steady and involuntary muscle contractions that lead to abnormal postures. These movements are generally schematic, tortuous, and may be jerky [1]. They are typically caused by the co-contraction of agonist muscles, which are responsible for the intended action, and antagonist muscles. Due to the diverse manifestations of the disorder, its overall epidemiology remains poorly defined [2]. According to the existing literature, focal dystonia is the most common form. It primarily affects individuals over 50, with an estimated prevalence of 732 cases per 100.000, classifying it as a disease predominantly affecting adults [3]. The condition is more frequently observed in females [4], and there appears to be a genetic component, as 20% of dystonia patients report a family history of the disorder [5].

Among the most common focal dystonias are cervical dystonia (69%) and blepharospasm (17%), followed by limb dystonia (3-7%), spasmodic dysphonia (1-3%), musician's (hand) dystonia (3%), and oromandibular dystonia (1%) [6]. While they constitute only 3-7% of all focal dystonias, the one involving the hand and upper limbs represents a significant clinical challenge for patients, as it often impairs their work and dramatically burdens their psychological state. A distinguishing feature of upper limb dystonia compared to other forms of focal dystonia is its task specificity. Typically, upper extremity dystonia occurs during selective and repetitive movements, with no motor impairment in the execution of other fewer skills demanding activities [7]. While initially focal dystonia exclusively involves the affected segment, over time, if left untreated, the condition can progress and become less specific and more diffuse, involving adjacent muscle groups and thus, leading to increased disability. Indeed, approximately 16% of primary focal hand dystonia may spread to proximal muscles and contralateral limbs or even develop a generalized form within a few years. This deterioration may be further complicated by the onset of dystonic tremor [8, 9]. Task-specific focal dystonia affects the hand or upper limb muscles, including conditions such as writer's, typist's, or musician's cramp. The predominant manifestations include excessive flexion of the fingers and wrist, pronation of the forearm, and humerus abduction, occasionally accompanied by trembling while performing highly skilled actions [10]. Diagnosis is typically clinical [11].

From a therapeutic perspective, the literature remains inconclusive, and there are no established protocols to guide the treatment of this aggravating and disabling condition. Oral medications such as baclofen and anticholinergics are sometimes utilized to alleviate symptoms of primary upper limb dystonia. However, their effectiveness is often limited, and the dosage of these drugs should always be well-calibrated to avoid side effects [12]. On the other hand, botulinum toxin A injections are commonly used to denervate the affected muscles and reduce symptoms. Still, it is essential to avoid any functional impairment resulting from secondary weakness [13]. Neurosurgical options, such as deep brain stimulation, are under investigation as potential treatments for dystonia, but the precise brain targets remain unidentified [14]. Recent literature underscores the importance of therapeutic exercise and occupational therapy in managing dystonia, both as stand-alone interventions during the early stages of the disease and in combination with pharmacological and non-pharmacological treatments in more advanced stages [15]. The goal of this systematic review is to synthesize the most recent literature from the past decade, to provide a foundation for future research that could contribute to the development of evidence-based treatment protocols integrating specific exercises and drug therapies to improve patients' outcomes and quality of life.

2. Materials and Methods

This work was carried out following the guidelines set by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement and received approval from PROSPERO, registered under the number: CRD42023475730. It intended to study all the existing literature regarding upper limb dystonia pharmacological and physiotherapy treatment, respectively, with botulinum toxin type A and occupational therapy. The literature was searched for on PubMed, Google Scholar and Cochrane based on the alignment of research objectives with the strengths of the selected databases, by three independent authors, considering articles published between 2013 and 2023, thus of the last 10 years. The eligibility criteria were structured using the PICOS: population (patients affected with hand or upper limb dystonia), intervention (therapeutic exercise), comparison (other treatments like BonT, rTMS, Tdcs…), outcome (functionality scale specific for dystonia), and study type (RC, RCT) framework, The search Boolean query was "hand dystonia" OR "upper limb dystonia" AND "rehabilitation" OR "therapeutic exercises." The search strategy employed for the PubMed database integrated the MeSH terminology. Once potential articles were gathered, a subsequent selection process was conducted according to the following exclusion criteria: full text not available, articles not in English language, articles in which botulinum toxin type A was associated with other pharmacological therapies as the aim was to highlight its potential combined with occupational therapy, protocols and in vitro studies or animal models to focus attention on achievements in human being. A revised version of the Jadad Scale was utilized to evaluate the risk of bias and the quality of the clinical trials across all the chosen studies (Table 1).

Table 1 The revised version of the Jaded quality scale.

Authors	Was the treatment assigned at random?	Was the randomization method explained and deemed suitable?	Were there any details provided about withdrawals and dropouts?	Was there a distinct explanation of the criteria for inclusion and exclusion?	Were the methods of statistical analysis outlined?	Jadad score (0-5)
	No	No	Yes	Yes	Yes	3
	No	No	Yes	Yes	Yes	3
	Yes	No	Yes	Yes	Yes	4
	Yes	Yes	Yes	Yes	Yes	5
	No	No	No	Yes	Yes	2
	Yes	Yes	Yes	Yes	Yes	5
	No	No	Yes	Yes	Yes	3
	Yes	Yes	Yes	Yes	Yes	5
	No	No	Yes	Yes	Yes	3
	No	No	No	Yes	Yes	2

Three independent evaluators assessed the titles and abstracts of the articles based on the inclusion and exclusion criteria, and the results were compared. Conflicts were resolved through the consensus of all authors. Full-text articles were retrieved and further evaluated for inclusion based on the eligibility criteria. As a systematic review, this study is exempt from Institutional Review Board approval.

3. Results

According to the inclusion and exclusion criteria outlined earlier, a detailed analysis of the search results yielded in 15 articles (11 of them were found in both PubMed and Google Scholars, one only in PubMed and 3 in Cochrane Library), which can be categorized into three main groups: seven randomized controlled trials, one clinical trial, and seven case reports (Figure 1, Table 2).

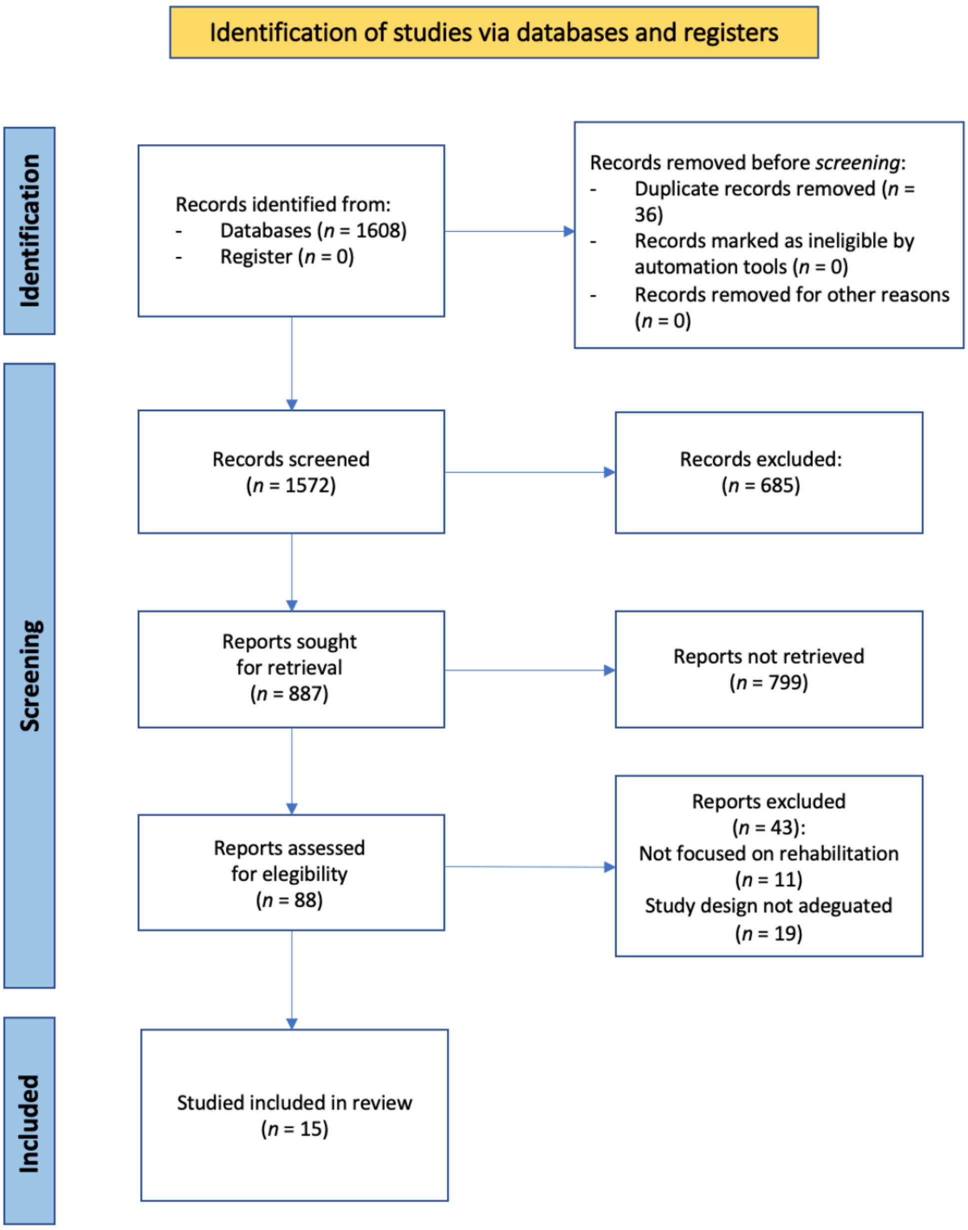

Figure 1 Prism a Flow Diagram.

Table 2 Summary characteristics of reviewed studies.

Title	Authors	Sample	Variables of the Study	Study limitation	Results
Botulinum toxin and occupational therapy for Writer's Cramp (WC)	Park JE, Shamim EA, Panyakaew P, Mathew P, Toro C, Sackett J, Karp B, Lungu C, Alter K, Wu T, Ahmad OF, Villegas M, Auh S, Hallett M. Botulinum toxin and occupational therapy for Writer's cramp. Toxicon. 2019 Nov; 169: 12-17. doi: 10.1016/j.toxicon.2019.07.010. Epub 2019 Jul 24. PMID: 31351085; PMCID: PMC6754272.	12 patients with writing difficulties. **Group 1**: 6 individuals received only botulinum toxin (BoNT) therapy. **Group 2**: 6 individuals received both BoNT therapy and occupational therapy.	Main Results: Subjective measures assessed by patients at 20 weeks; Writer's Cramp Rating Scale (WCRS); Writer's Cramp Impairment Scale (WCIS); Writer's Cramp Disability Scale (WCDS); Handgrip strength;Dynamic parameters	-differences in baseline disease severity between the two groups; -sample size;	Subjective Evaluation: There were no significant differences between the groups at 20 weeks. Objective Improvement: BoNT & Occupational Therapy Group: 28% reduction in WCIS scores.
Effectiveness of Botulinum Toxin on Pain in Stroke Patients Suffering from Upper Limb Spastic Dystonia	Trompetto C, Marinelli L, Mori L, Puce L, Avanti C, Saretti E, Biasotti G, Amella R, Cotellessa F, Restivo DA, Currà A. Effectiveness of Botulinum Toxin on Pain in Stroke Patients Suffering from Upper Limb Spastic Dystonia.	-Participants: 41 individuals with upper limb spastic dystonia. -Treatment: Administered incobotulinum toxin-A therapy.	-Time Points: -T0: Immediately before treatment. -T1: One-month post-treatment with botulinum toxin A. -Evaluations: -Pain Assessment: NRS (Numeric Rating Scale) for	-observational study; -small sample size; -pain reduction could be due to a placebo effect;	Treatment: Administration of Botulinum toxin type A (BoNT-A). Effects: Decreased discomfort across all joints, including the shoulder region. Nociceptive discomfort is shared in a significant subset of individuals with upper limb spastic dystonia. Results:

	Toxins (Basel). 2022 Jan 5; 14(1): 39. doi: 10.3390/toxins14010039. PMID: 35051017; PMCID: PMC8780435.		pain at rest and muscle tone. -Neuropathic Pain Identification: Douleur Neuropathique 4 (DN4) questionnaire. -Patient Request: -Identify the joint experiencing the most significant pain.		BoNT-A reduced spastic dystonia and discomfort in all joints, except the shoulder. Pain relief in the shoulder may be influenced by improved abnormal postures affecting surrounding joints.
Assessment of Botulinum Neurotoxin Injection for Dystonic Hand Tremor: A Randomized Clinical Trial	Rajan R, Srivastava AK, Anandapadmanabhan R, Saini A, Upadhyay A, Gupta A, Vishnu VY, Pandit AK, Vibha D, Singh MB, Bhatia R, Goyal V, Dwivedi SN, Srivastava P, Prasad K. Assessment of Botulinum Neurotoxin Injection for Dystonic Hand Tremor: A Randomized Clinical Trial. JAMA Neurol. 2021 Mar 1; 78(3): 302-311. doi: 10.1001/jamaneurol.2020.4766. PMID:	Participants: 30 adults. Group Assignment: BoNT Group: Received botulinum toxin treatment. Placebo Group: Received a placebo treatment	Outcome: Fahn-Tolosa-Marin Tremor Rating Scale.	-Patients were included regardless of the dystonia distribution; -Potentially placebo effect; -outcomes not assessed earlier than 6 weeks, possible underestimation of hand weakness	-Patient Condition: -Dystonic tremor of the upper extremities. -Treatment Method: -Electromyography-guided personalized BoNT injections. -Outcomes: -Effective tremor management. -No significant hand weakness was observed.

	33346814; PMCID: PMC7754081.				
Effect of botulinum toxin A & task-specific training on upper limb function in post-stroke focal dystonia.	Umar M, Masood T, Badshah M. Effect of botulinum toxin A & task-specific training on upper limb function in post-stroke focal dystonia. J Pak Med Assoc. 2018 Apr; 68(4): 526-531. PMID: 29808039.	A total of 43 participants were divided into two groups: Experimental Group: Received BoNT-A injections along with task-specific training. Control Group: Received only task-specific training. Duration: 8 weeks.	Data Collection Timeline: Baseline After 4 weeks After 8 weeks Assessment Tools: Upper Extremity Components of the Motor Assessment Scale Upper limb section of the Fugl-Meyer Assessment	Not randomized; not blinded;	Cohorts: Both groups showed significant improvements in: Motor Assessment Scale Fugl-Meyer Assessment Differences: No significant differences at: Baseline After 4 weeks After 8 weeks Outcome: Eight weeks of task-oriented training improved upper limb functionality in patients with post-stroke focal dystonia.
Effects of Low-frequency Repetitive Transcranial Magnetic Stimulation on Focal Hand Dystonia: A Case Report.	Furukawa T, Kanke H, Masakado Y. Effects of Low-frequency Repetitive Transcranial Magnetic Stimulation on Focal Hand Dystonia: A Case Report. Tokai J Exp Clin Med. 2021 Apr 20; 46(1): 44-50. PMID: 33835475.	Patient: 40-year-old female Diagnosis: Right-hand dystonia Treatment: Repetitive transcranial magnetic stimulation (rTMS) therapy Stimulation Details:	Evaluation Summary: Improvement Observed: After 350 repetitions Assessment Tools: Simple Test for Evaluating Hand Function (STEF) Finger Flexion and Writing Movements Monitoring Method: Cerebral Blood Flow	-single case report;	Results Summary: After 150 rTMS Sessions: Significant improvements in separation movements of the right arm and fingers. After 350 rTMS Sessions: Movements became notably swift, approaching left-side speed.

		Target Area: Motor cortex for the right upper limb Frequency: 1 Hz Repetitions: 350 to 500 Intensity: 1.2 times the determined motor threshold	changes are assessed via Near-Infrared Spectroscopy (NIRS) Measurements of: Motor Evoked Potential (MEP) Cortical Silent Period (CSP) Short-interval Intracortical Inhibition (SICI) Timing of Measurements: Before TMS After 150 repetitions After 350 repetitions Additional Documentation: Arm movements recorded via video.		Marked enhancements in STEF and written expression. MEP Observations: No substantial changes were recorded. Increased CSP latency noted. Significant reduction in SICI ratio. NIRS Assessments: Minimal changes in relative hemoglobin concentrations in the left motor cortex (responsible for right finger movements) compared to pre-stimulation. Notable decrease in hemoglobin levels in: Left premotor cortex Left prefrontal cortex The decline in writing movements was observed in the left motor, premotor, and prefrontal regions. Clinical Outcome: Improvement in clinical symptoms remained stable over an extended period with low-frequency rTMS application.
Effect of Transcranial Direct Current Stimulation on Neurorehabilitation of Task-Specific Dystonia: A	Rosset-Llobet J, Fàbregas-Molas S, Pascual-Leone Á. Effect of Transcranial Direct Current Stimulation on Neurorehabilitation of Task-Specific Dystonia:	Study Design Summary: Type: Parallel, double-masked randomized Participants: 30 musicians Diagnosis: Right-hand primary focal dystonia	Intervention Summary: Duration: 2-week neurorehabilitation program Therapy Components:	-subjective outcomes measuremente s; -tDCS not examined alone;	Outcome Summary: Improvement: Both cohorts showed a marked enhancement in dystonia severity scores over the 2-week period. Treatment Group: Demonstrated a statistically significant greater level of

Double-Blind, Randomized Clinical Trial.	A Double-Blind, Randomized Clinical Trial. Med Probl Perform Art. 2015 Sep; 30(3): 178-184. doi: 10.21091/mppa.2015.3033. PMID: 26395620.		Sensory-motor retuning therapy Active or sham transcranial direct current stimulation (tDCS) Session Details: Each session: 1 hour tDCS administered during the first 30 minutes of each session Total Sessions: 10 Blinding: Both therapist and participant were blinded to the tDCS condition Assessment: The dystonia severity score was evaluated before and after the 2-week intervention.	-no follow-up after two weeks of treatment	improvement compared to the control group.
Non-invasive brain stimulation and kinesiotherapy for treatment of focal dystonia:	De Oliveira Souza C, Goulardins J, Coelho DB, Casagrande S, Conti J, Limongi JCP, Barbosa ER, Monte-Silva K,	Study Summary: Patient Diagnoses: Cervical Dystonia (CD)	Evaluation Summary: **Cervical Dystonia (CD) Assessment**: Tools Used:	-3 cases, open-label clinical observation; -intervention mechanism not	Key Conclusion: Integration of Intervention Strategies: Enhances clinical outcomes in neurological disorders.

Instrumental analysis of three cases.	Tanaka C. Non-invasive brain stimulation and kinesiotherapy for treatment of focal dystonia: Instrumental analysis of three cases. J Clin Neurosci. 2020 Jun; 76: 208-210. doi: 10.1016/j.jocn.2020.04.025. Epub 2020 Apr 10. PMID: 32284289.	Hand Focal Dystonia (HFD) Treatment Regimen: Daily sessions Total Sessions: 15 Therapies Administered: CD Patients: Transcranial Direct Current Stimulation (tDCS) HFD Patients: Repetitive Transcranial Magnetic Stimulation (rTMS) Combined Treatment: Non-invasive brain stimulation (NIBS) with kinesiotherapy.	Modified Toronto Scale for Cervical Dystonia Assessment (MTS) Quiet Balance Test Visual Postural Assessment **Hand Focal Dystonia (HFD) Assessment**: Tools Used: Handwriting Quality Analysis Tremor Acceleration Amplitudes Writer's Cramp Rating Scale (WCRS) **Assessment Timing**: Conducted at: Pre-treatment Immediately post-treatment Three months post-treatment	completely understood	Predicts favorable results in the challenging clinical management of: Cervical Dystonia (CD) Hand Focal Dystonia (HFD).
Cathodal transcranial direct current stimulation in children with dystonia: a sham-controlled study	Young SJ, Bertucco M, Sanger TD. Cathodal transcranial direct current stimulation in children with dystonia: a sham-controlled study. J Child Neurol.	Type of Study: Double-blind Sham-controlled Crossover design Participants: 14 pediatric subjects Diagnosis: Dystonia	Session Details: Duration: Two separate sessions, each lasting 2 hours Stimulation Types:	-sample size -minor effects of cathodal stimulation; -placebo effect could have	Key Findings: Observation: There is a notable decrease in overflow following actual stimulation. Specific Context:

	2014 Feb; 29(2): 232-239. doi: 10.1177/0883073813492385. Epub 2013 Jun 11. PMID: 23760989.		Real Transcranial Direct Current Stimulation (tDCS) in one session Sham Stimulation in the other session Session Sequence: Randomized order of sessions Assessment Tool: Barry-Albright Dystonia Scale Assessment Timing: At the beginning and conclusion of each session.	influenced the results	Observed when participants performed the experimental task with the hand opposite to the cathode.
KinesioTaping Reduces Pain and Modulates Sensory Function in Patients with Focal Dystonia: A Randomized Crossover Pilot Study: neurorehabilitation and Neural Repair.	Pelosin E, Avanzino L, Marchese R, Stramesi P, Bilanci M, Trompetto C, Abbruzzese G. Kinesiotaping reduces pain and modulates sensory function in patients with focal dystonia: a randomized crossover pilot study. Neurorehabil Neural Repair. 2013 Oct; 27(8): 722-731. doi: 10.1177/154596831349	Study Overview: **Participants**: 25 patients with dystonia 14 with Cervical Dystonia (CD) 11 with Focal Hand Dystonia (FHD) Study Design: Randomized crossover pilot investigation Intervention Duration: 14-day treatment Treatment Types: Kinesio Taping	Assessment Summary: Pain Assessment: Visual Analog Scale (VAS) used to evaluate: Typical Pain Maximum Pain Pain Relief Disease Severity Measurements: Cervical Dystonia (CD): Toronto Western Spasmodic Torticollis Rating Scale	-no follow-up; -poor sensitivity in measuring tiny changes in the used evaluation scales	Key Findings: Kinesio Tape Administration: Resulted in: Reduction of perceived pain levels Change in sensory discrimination capabilities Sham Taping: Did not produce observable effects. Correlation Observed: Significant positive correlation in both patient groups: Enhancement of pain perception is linked to a decrease in somatosensory temporal discrimination threshold values

	1010. Epub 2013 Jun 13. PMID: 23764884.	Sham Taping Application Sites: CD: Neck FHD: Forearm muscles Washout Phase: 30-day period before receiving the alternative treatment.	Focal Hand Dystonia (FHD): Writer's Cramp Rating Scale (WCRS) Additional Assessment: Somatosensory Temporal Discrimination Threshold evaluated.		associated with Kinesio Taping intervention.
Assessment of the effects of Kinesiotaping on musical motor performance in musicians suffering from focal hand dystonia: a pilot study.	Bravi R, Ioannou CI, Minciacchi D, Altenmüller E. Assessment of the effects of Kinesiotaping on musical motor performance in musicians suffering from focal hand dystonia: a pilot study. Clin Rehabil. 2019 Oct; 33(10): 1636-1648. doi: 10.1177/0269215519852408. Epub 2019 Jun 4. PMID: 31159569.	Study Overview: Participants: 7 musicians diagnosed with Focal Hand Dystonia (FHD) Experimental Conditions: Without Kinesio taping During corrective kinesiotaping intervention Immediately after the removal of the corrective tape During sham kinesiotaping intervention Immediately after the removal of sham tape (Block 2)	Intervention Summary: Type: Customized kinesiotaping intervention Application: Applied to the affected fingers Tailored to individual dystonic patterns exhibited by each patient during performance.	Small sample size; pilot study; no control group; lack of a standardized and objective assessment tool to evaluate heterogeneous groups of musicians; each patient underwent both the Correction and the Sham Kinesiotaping interventions	Key Findings: Statistical Analysis: No statistically significant differences between: Corrective kinesiotaping and sham Kinesio taping Overall performance ($p > 0.05$) Finger posture ($p > 0.05$) Effect of Taping: Minor advantages from corrective kinesiotaping dissipated after tape removal. Musician Assessment: Musicians considered corrective kinesiotaping ineffective for enhancing musical performance. Coactivation Index:

		Randomization: Experimental blocks were randomly assigned to participants.			No significant alterations were observed across different conditions ($p > 0.05$).
A simple orthosis solves a problem in a patient with a dystonic finger after a stroke.	Vercelli S, Ferriero G, Bravini E, Al Yazeedi W, Salgovic L, Caligari M, Sartorio F. A simple orthosis solves a problem in a patient with a dystonic finger after stroke. J Hand Ther. 2017 Jan-Mar; 30(1): 113-115. doi: 10.1016/j.jht.2016.04.003. Epub 2016 Nov 25. PMID: 27894678.	Case Overview: Intervention Type: Basic static hand orthosis Purpose: Manage atypical finger movements during gripping activities Design: Low-temperature, custom-fabricated thermoplastic orthosis Specifically designed to: Prevent metacarpal hyperextension Improve grip strength	Clinical observation and analytical reasoning.	-case report; -only clinical observation	Patient Outcomes: Effect of Orthotic Device: Enabled extension of: Proximal Interphalangeal (PIP) joints Distal Interphalangeal (DIP) joints Enhanced ability to extend fingers during object manipulation Comfort: No discomfort was reported during hospitalization Follow-Up: At one-year follow-up assessment: The patient continued to use the orthosis
Two single cases were treated by a new pseudoelastic upper-limb orthosis for secondary dystonia of the young.	Garavaglia L, Pagliano E, Arnoldi MT, LoMauro A, Zanin R, Baranello G, Aliverti A, Pittaccio S. Two single cases treated by a new pseudoelastic upper-limb orthosis for secondary dystonia of the young. IEEE Int Conf	Study Overview: Participants: 2 males diagnosed with upper-limb dystonia 6 age-matched healthy controls Intervention: Development of tailored assistive devices	Evaluation Methods: Clinical Scales Used: Modified Ashworth Score Melbourne Upper Limb Assessment Pediatric Quality of Life Inventory (PedsQL) Additional Assessment Methods:	-case report	Key Findings: Reference Data: Normal kinematics were established from the control group. Kinematic Analysis Results: Notable alterations in movement patterns for both patients, including: Increased range of motion (ROM) in initially rigid segments

	Rehabil Robot. 2017 Jul; 2017: 1260-1265. doi: 10.1109/ICORR.2017.8009422. PMID: 28813994.	specifically designed for the patients.	Structured interviews Optoelectronic kinematic analysis		Improvements in posture Development of multi-joint strategies Clinical Scale Observations: Trends in clinical scales did not consistently align between the two cases.
Botulinum toxin treatment of spasticity targeted to muscle endplates: an international, randomized, evaluator-blinded study comparing two different botulinum toxin injection strategies for treating upper limb spasticity.	Rekand T, Biering-Sörensen B, He J, Vilholm OJ, Christensen PB, Ulfarsson T, Belusa R, Ström T, Myrenfors P, Maisonobe P, Dalager T. Botulinum toxin treatment of spasticity targeted to muscle endplates: an international, randomized, evaluator-blinded study comparing two different botulinum toxin injection strategies for the treatment of upper limb spasticity. BMJ Open. 2019 May 5; 9(5): e024340. doi: 10.1136/bmjopen-2018-024340. PMID:	Study Overview: Study Design: Prospective, open-label study with evaluator blinding Conducted across 20 medical centers Participants: Patients aged over 18 years Diagnosis: Spasticity in upper limbs Indicated by Modified Ashworth Scale (MAS) score of 2 or 3 Conditions: The following stroke Following traumatic brain injury Treatment History: Had undergone a minimum of two	Outcome Measures Summary: Primary Outcome Measure: The proportion of patients with at least a one-point decline in Modified Ashworth Scale (MAS) scores (range 0 to 4). Baseline MAS scores for elbow flexors assessed at: Baseline 4 weeks 12 weeks post-injection A decrease of ≥1 point from baseline is considered clinically significant. Secondary Outcome Measures: Intensity of Spasticity-Related Pain:	-sample size; -patients not consulted to design the study;	Due to the restricted participant pool, the assessment of non-inferiority for NMJ-targeted injections could not be established. Nevertheless, no statistically significant differences were observed between the groups.

	31061021; PMCID: PMC6502046.	consecutive treatment cycles with BoNT-A Required retreatment using the same approach as previous cycles.	Assessed by Visual Analog Scale (VAS) (range 0 to 10) at: Baseline 4 weeks 12 weeks Pain Associated with Injection: Evaluated via VAS during the initial visit. Goal Attainment Scale (GAS): Scores range from -2 to 2 at either 4 or 12 weeks. Subject's Overall Evaluation of Treatment Effectiveness: Assessed after the study.		
Functional recovery from chronic writer's cramp by brain-computer interface rehabilitation: a case report.	Hashimoto Y, Ota T, Mukaino M, Liu M, Ushiba J. Functional recovery from chronic writer's cramp by brain-computer interface rehabilitation: a case report. BMC Neurosci. 2014 Sep 1; 15: 103. doi: 10.1186/1471-2202-15-103. PMID:	Case Report Summary: Patient Diagnosis: Writer's cramp Intervention: Biweekly one-hour training sessions Duration: Five months	Study Overview: Procedure: EEG recordings conducted over the bilateral sensorimotor cortex Patient: Individual with chronic Writer's cramps	Single case report without control; -pilot study	The present pilot investigation indicates that a brain-computer interface may provide real-time feedback on cortical excitability to individuals with dystonia, enabling them to modulate excessive neural activity and facilitate functional recovery.

	25179667; PMCID: PMC4158043.	No adverse effects reported Key Findings: Notable reduction in beta frequency component at EEGs during handwriting Correlation with marked functional improvement..	Task: Subject instructed to: Diminish an amplified beta frequency component in the EEG Extend the hand during the task		
Improvement of Writer's Cramp from an Old Lesion in the Contralateral Hemisphere with Transient Gait Disturbance After Thalamotomy.	Asahi T, Taira T, Ikeda K, Horisawa S, Yamamoto J, Tsubono H, Sato S. Improvement of Writer's Cramp from an Old Lesion in the Contralateral Hemisphere with Transient Gait Disturbance After Thalamotomy. World Neurosurg. 2019 Jul; 127: 8-10. doi: 10.1016/j.wneu.2019.03.199. Epub 2019 Mar 28. PMID: 30928593.	Case Report Overview: Patient: 43-year-old individual Diagnosis: Writer's cramp Clinical Background: Developed following a hemorrhagic event in the left basal ganglia Accompanied by right hemiparesis.	Clinical observation	Case report with only clinical observation	Clinical findings indicated that following a rehabilitation program, the patient's ambulation significantly improved, returning to pre-surgical levels within three months, and the symptoms of Writer's cramp resolved ultimately.

An analysis of the data collected from our research showed that rehabilitative treatment, often combined with other therapeutic approaches, is commonly employed for patients affected by upper limb dystonia, with a particular emphasis on musicians' hand dystonia.

According to these findings, in recent years, a significant consensus has emerged from various studies regarding the imperative to combine therapeutic interventions with specific therapeutic exercises in the rehabilitation of patients. This integration is essential for optimizing recovery and enhancing overall patient outcomes. By focusing on tailored exercise regimens, it is possible to significantly improve the likelihood of patients returning to a lifestyle that closely mirrors that of healthy individuals. This discussion centers on the critical role that these specific exercises play in shaping rehabilitation efforts and transforming patient lives for the better.

Specifically, the combination of botulinum toxin A injections and occupational therapy has been reported to demonstrate the most significant potential for improvement in individuals suffering from writer's cramps. A work conducted in 2019 showed the effectiveness of this combined approach, as it resulted in a notable objective improvement in patients compared to botulinum toxin injections alone [16]. The occupational therapy employed in this research included an isometric splint designed to help patients execute finger movements that counteract dystonic movements while writing. Participants were advised to wear the splint during daily writing sessions lasting 30 minutes for 20 weeks. All patients received guidance from the same physical therapist throughout the study. Although the results are promising, the study's small sample size, consisting of only 12 patients, and its short follow-up period of 20 weeks, are notable limitations. The study demonstrated the positive impact of botulinum toxin on post-stroke patients, not only by reducing muscle spasticity but also by alleviating tremors and improving the quality of life [17]. Based on a recent article, individuals who have experienced a stroke should undergo task-oriented training for 60 minutes each day, three times a week, over 12 weeks, commencing one week after receiving the botulinum toxin injection. Each daily session should be divided into 3 sets of 20 repetitions for each task. As the patient makes progress, the difficulty of the exercises should be increased. Suggested exercises for each week include: week I and II involve reaching for a glass on the table, pushing a bottle aside, and rolling a can with the hand; during week III and IV, patients will focus on keeping the elbow straight while tapping a bottle with their wrist, bringing a glass to their mouth, and performing bimanual activities like pouring and transferring water; in week V and VI, activities include finger tapping, towel folding, card picking, drawing dots and lines, lifting blocks of various colors, and turning objects on and off; for weeks VII and VIII, tasks consist of turning newspaper pages, swiping coins, picking up ludo pieces, and catching and throwing a ball; finally, from week IX and XII, exercises will feature sorting beans of different colors into corresponding bowls, holding a pencil to write words, and stacking various coins on top of each other [18]. A randomized clinical trial compared the benefits of therapeutic exercise alone versus exercise combined with botulinum toxin in 43 patients with upper limb dystonia after stroke [19]. The results showed that a specific training program for 8 weeks had better outcomes than the combined treatment. However, the exercises used in the program were not reported, making it challenging to replicate them. Additionally, post-stroke dystonia may have unique characteristics and may only be a subset of a more significant problem. Nevertheless, this study is highly promising and underscores the significance of therapeutic exercise in this patient population.

Other recurrent therapeutic modalities for upper-limb dystonia include transcranial direct current stimulation (tDCS) and repetitive transcranial magnetic stimulation (rTMS). Notably, a case has been documented involving a patient in her 40s with right-hand dystonia who experienced substantial improvement following exclusive rTMS treatment [20]. However, most existing literature indicates that these interventions are often employed with rehabilitation strategies. A double-masked randomized clinical trial explored the effects of tDCS combined with a 2-week sensory motor retuning therapy-based neurorehabilitation program [21]. Thirty patients with primary focal dystonia of the right hand were divided into groups of 15. Each patient participated in a 2-week neurorehabilitation program, receiving either active or sham tDCS for 30 minutes daily during their 1-hour therapy session. The administration of biparietal tDCS, utilizing a left-sided cathode throughout 10 sessions, was deemed a safe intervention for this patient cohort. The combination of tDCS and kinesiotherapy has demonstrated a longer-term improvement in dystonia symptoms, and this effect appears to be significant in both pediatric [22] and adult patients [23]. While the sample size was small and the rehabilitation used by the patients was not precisely defined, these findings provide a promising outlook and motivate further research into the development of a defined rehabilitation plan for pediatric and adult patients with dystonia.

Two studies have been identified in the literature investigating the application of kinesiotaping in dystonic patients. While the first study, published in 2013, reported a reduction in painful hand sensation in patients following a 14-day treatment period and 30 days of washout [24], the second study, published in 2019, concluded that Kinesio taping was ineffective in reducing dystonic patterns or enhancing the performance capabilities of musicians suffering of focal hand dystonia [25]. In the initial case, a standard beige KinesioTape was employed universally, utilizing “I”-shaped strips, which are documented for their analgesic effects. KinesioTape was applied to the medial-superior region of the forearm’s dystonic musculature utilizing two “I-strips” for patients presenting with focal hand dystonia. Each strip was placed along the flexor carpi radialis and on the flexor carpi ulnaris muscles, with application-directed rostrocaudal while the forearm muscles were stretched [24]. In the second case, a 2.5 cm wide tape strip was affixed to the dorsal surface of the finger while maintaining full tension just proximal to the distal interphalangeal joint. This was accomplished by bifurcating the distal section of the strip into two 1.25 cm segments wrapped around the finger. Following this, the KinesioTape was fully extended to the midpoint of the strip and securely anchored over the metacarpal and the wrist regions without applying tension. The subsequent application involved direct placement of the tape on the dorsal surface of the dystonic finger with inward pressure to inhibit uncoordinated extension during performance. A second 2.5 cm wide tape strip was then applied to the palmar surface of the finger while the subject maintained it curved into the palm. This tape was first anchored to the distal interphalangeal joint fully extendedly and subsequently secured over the palm up to the wrist without applying tension. Finally, the strip was affixed to the palmar surface of a compensatory finger [25]. Given the discordance between these two studies, it is worth further investigating this topic as simple pain relief could be significant for affected patients. Indeed, a pain-free joint works better, and consequently, a pain-free dystonic hand or upper limb can tolerate more intense physiotherapy, resulting in longer-lasting and perhaps faster results.

Equally promising is the use of orthoses that can act as an aid in hand and upper limb dystonia even though scientific evidence is minimal. In 2017, Vercelli et al experimented a custom-fabricated orthotic device to improve hand motion and function for a patient affected by after stroke hand dystonia obtaining a satisfactory result [26]. In the same year, a dynamic wearable orthosis incorporating metallic components with non-linear mechanical properties was trialed in two young males with upper-limb dystonia [27]. After a month of use, kinematic assessments revealed alterations in motor patterns for both subjects, including an increased range of motion in previously rigid segments, improved posturing, and the development of multi-joint movement strategies.

The typical constant that emerges from all analyzed studies remains the need to combine any type of treatment with a specific therapeutic exercise. Finding optimal rehabilitation for these patients could be the key to returning them to a lifestyle as similar as possible to that of a healthy person. Therefore, the authors' focus will have to be placed precisely on specific exercises that can change the outcome of these patients.

4. Discussion

The data analysis of the presented research underscores the importance of adopting a multi-faceted approach to managing upper limb dystonia. Dystonia is a complex condition with diverse etiologies and manifestations, which requires a customized treatment strategy [28].

A systematic review conducted by R. Chiaramonte et al. in 2021 [11] focused on the rehabilitation of musician's dystonia. In contrast, the current review expands the scope to encompass all forms of focal dystonia affecting the upper limbs, regardless of their etiology. This broader perspective allows for a comprehensive evaluation of available treatment options, highlighting various therapeutic interventions such as exercise regimens and tailored rehabilitation programs.

This review aims to address a critical gap in the literature by emphasizing rehabilitation as a key component for improving patient outcomes across diverse clinical presentations of upper limb dystonia.

Several therapeutic modalities have shown promise [29], and the discussion here delves into their implications, limitations, and potential areas for further research. The study on the writer's cramp highlights the potential benefits of combining botulinum toxin A injections with occupational therapy in enhancing patient outcomes [16]. Indeed, there is a wealth of studies in the literature that confirm the effectiveness of botulinum toxin type A in treating writer’s cramp and various forms of dystonia. Interestingly, in addition to the peripheral action that the toxin exerts on muscle spindles, recent research indicates that the toxin may also influence central mechanisms within the cerebral cortex [30]. Botulinum toxin A interrupts neuromuscular transmission between intrafusal muscle fibers and gamma-motoneuronal endings, consequently affecting the afferent signals from muscle spindles through Ia afferents [31]. This reduction in spinal excitability indirectly influences motor control centers in the brain, including the sensorimotor cortex [32]. This explains how peripheral botulinum toxin type A injections can lead to cortical reorganization and adaptive plasticity. In contrast, the impact of therapeutic exercise on promoting neuroplasticity is well-established, with a systematic review by Penna et al in 2021 highlighting the benefits of aerobic physical activity post-stroke [33]. Consequently, in addition to the established benefits associated with physical conditioning, functional capacity, mood enhancement, and cardiovascular health, therapeutic exercise potentiated the above-mentioned neuroplasticity process. For this reason, the synergic action of botulinum toxin A combined with targeted therapeutic exercise and, in the previously mentioned writer’s cramp, occupational therapy could be a successful approach for this category of patients. However, the limitations of a small sample size and restricted follow-up duration suggest the need for larger-scale and longer-term studies to validate these findings. It is also essential to investigate the sustainability of these improvements over time and the long-term impacts on patients' quality of life. The proven efficacy of botulinum toxin appears not merely to reduce muscle spasticity and develop neuroplasticity. A post-stroke dystonia study has demonstrated this therapy's success in reducing dystonic tremors, thus improving the overall quality of life [17]. Unfortunately, although the therapeutic exercise group displayed improved outcomes in the short term, the absence of exercise details makes it difficult to reproduce the results. Therefore, further research is necessary to identify the specific exercises that can provide the most benefit for after-stroke dystonia patients. A randomized clinical trial conducted in 2018 compared the use of botulinum toxin and propranolol in the treatment of essential and dystonic vocal tremors [33]. A total of fifteen patients were stratified into two groups based on the type of tremor and received both botulinum toxin and propranolol therapies at different intervals. This study showed that the two kinds of tremors respond differently to the two treatments, and specifically, dystonic tremors seem to be responsive only to botulinum toxin. Moreover, it has recently been reported in the literature that the type of tremor that better responds to botulinum toxin therapy is precisely dystonic tremor [34]. Significantly, reducing tremors in patients affected by dystonia could improve movements and optimize occupational therapy. However, it would be necessary to establish specific therapeutic exercises so that patients can undergo validated and reproducible rehabilitative programs. Additionally, transcranial direct current stimulation (tDCS) and repetitive transcranial magnetic stimulation (rTMS) demonstrate potential as complementary therapies in the management of dystonia. By 2021, a comprehensive review of the literature had further investigated the application of non-invasive stimulation techniques in treating movement disorders [35]. This work found that there was, until then, no recommendation for the use of non-invasive modulation in hand dystonia, although the results were promising. The best results were derived from employing low-frequency rTMS on the premotor cortex in focal hand dystonia and the cerebellum for dystonia tremor. Despite the positive findings reported in numerous studies and the robust theoretical justification for the application of non-invasive stimulation in focal upper limbs dystonia, further research is needed to optimize rTMS and tDCS protocols, ensuring their applicability in routine clinical practice and investigating their long-term effects [36]. To date, scientific evidence suggests combining tDCS with sensory motor retuning therapy-based neurorehabilitation since it is safe and effective [21]. This highlights the importance of incorporating non-invasive neuromodulation techniques with rehabilitation programs. Additional investigations are required to establish the ideal stimulation parameters and assess the long-term effects on the manifestations of dystonia. Two studies on the use of Kinesio taping for hand and upper limb dystonia have produced conflicting results. One study found a reduction in pain [24], while the other did not observe any significant improvement in dystonic patterns [25]. This discrepancy underscores the need for further investigation into the precise function of kinesiotaping in the administration of this type of dystonia. Factors such as patient selection, treatment duration, and application technique should be explored to clarify the effectiveness of kinesiotaping. In contrast to the discordant results found regarding kinesiotaping, there seems to be a unanimous view of the benefits obtained from the use of special orthotic devices. In 2013, a study tested a writing orthotic device for writer’s cramps in a population of fifteen patients for a time of two weeks [37]. After two weeks of home use of such a device, patients demonstrated improved writing, reduced dystonic tremor, and decreased pain perception. Moreover, from the previously analyzed studies, it was found that the use of specific orthotic devices is able both to improve hand movements in daily activities and exercises performed during rehabilitation programs [26] and, on the other hand, to improve the range of motion after a certain amount of time from the start of the wearing [27]. With the development of clinical engineering and research, it would be possible to obtain smaller and finer orthotic devices to improve patients' lifestyles and facilitate therapeutic exercise. Although the scientific evidence is limited, these findings are encouraging and warrant more comprehensive studies to assess the effectiveness of different orthotic designs and their long-term impact on dystonia patients. As mentioned, all therapeutic approaches for hand and upper limb dystonia require a concomitant therapeutic exercise program. Therapeutic exercise, indeed, plays a crucial role in the rehabilitation of dystonia patients.

This study is not free from limitations; while this systematic review provides valuable insights into the integration of therapeutic exercises with treatment interventions, several limitations should be acknowledged. First, the reliance on a limited number of studies, precisely 15 articles, may affect the generalizability of the findings. The variation in study designs, including the predominance of randomized controlled trials alongside case reports and clinical trials, introduces heterogeneity that can complicate comparisons and consensus on effective interventions. Additionally, the inclusion criteria may have excluded relevant studies due to specific restrictions, potentially leading to publication bias. Furthermore, the evaluators' subjective interpretations of inclusion and exclusion criteria could introduce bias despite efforts to reach a consensus. Lastly, given that this study did not seek Institutional Review Board approval due to the nature of a systematic review, ethical considerations surrounding the original research studies were not independently assessed, limiting the comprehensiveness of the evaluation. Future research should aim to include a broader range of studies and explore the effectiveness of therapeutic exercises in diverse patient populations to enhance understanding and applicability.

While specific exercises and training programs for dystonia patients have varied, the significance of rehabilitation in treatment plans is clear. To optimize outcomes, there is an urgent need for multicenter RCTs utilizing standardized protocols. These studies should also incorporate more extended follow-up periods to evaluate the sustained effectiveness of exercise regimens.

By identifying the most effective, patient-specific interventions, such research can enhance treatment efficacy and support patients in regaining functional independence and returning to a normal lifestyle. Ultimately, this will emphasize the critical role of comprehensive rehabilitation in managing dystonia.

5. Conclusions

In conclusion, the complex and heterogeneous nature of upper limb dystonia necessitates a multifaceted treatment approach. Botulinum toxin remains the most commonly used intervention; however, further investigations are needed to explore its efficacy across different subtypes of dystonia and at various stages of disease progression. Combining multiple therapeutic modalities may represent a promising strategy for more rapid and sustained improvements. Integrating botulinum toxin injections, therapeutic exercise, non-invasive neuromodulation techniques, kinesiotaping, orthoses, and occupational therapy offers the potential for a comprehensive and personalized rehabilitation plan. Moreover, many studies have relied on subjective outcome measures, underscoring the need for standardized, reproducible assessment tools to establish evidence-based management guidelines for this complex condition. While the studies reviewed present promising results, they also highlight the need for larger, longer-term, and more standardized investigations to refine and expand the understanding of these treatments and their benefits in the management of upper limb dystonia. Ultimately, the primary objective should be to improve the quality of life and functional abilities of affected patients and facilitate their reintegration into daily activities.

Author Contributions

Conceptualization, G.F., M.V.R. and A.B.; methodology, G.F., M.Me and F.A.; software, M.R. and M.Me.; validation, M.P.; formal analysis, L.D.A. and R.M.; investigation, M.V.R. and L.D.A.; resources, F.A. and R.M.; data curation, M.P. and A.B.; writing—original draft preparation, L.D.A. and M.V.R.; writing—review and editing, G.F. and Ri.M.; visualization, M.Me. and A.B.; supervision, M.Ma.; project administration, M.Ma. and A.B. All authors have read and agreed to the published version of the manuscript.

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