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" An Overview of Rehabilitation Approaches for Focal Hand Dystonia in Musicians: A Scoping Review"

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ABSTRACT

Objective

To provide a comprehensive overview of rehabilitation treatment strategies for Focal Hand Dystonia in musicians, examining their evolution and effectiveness.

Data Sources

A systematic search of five databases, PubMed, PEDro, Cochrane Library, Trip, and Google Scholar, to identify relevant articles on Focal Hand Dystonia rehabilitation. The last search was performed on 20/12/2023.

Methods

Inclusion criteria were applied to 190 initially identified articles, resulting in 17 articles for review. Exclusions were made for duplicates, irrelevant titles, abstracts, and non-rehabilitation interventions.

Results

Ten different rehabilitation approaches were identified over 20 years. While no definitive intervention protocol exists, a multimodal approach is commonly recommended.

Conclusions

This scoping review underscores the diversity of rehabilitation strategies for Focal Hand Dystonia. It suggests the potential of multimodal approaches, emphasizing the need for further large-scale clinical efficacy studies.

Keywords: musician's dystonia, rehabilitation, motor retraining, task specific training.

INTRODUCTION

Focal Hand Dystonia in musicians also known as Musician's Focal Dystonia or musician's cramp, is a task-specific movement disorder characterized by involuntary loss of control and coordination of finger movements during instrument playing. It is generally painless and associated with extensive and vigorous use of the fingers. The etiology of Focal Hand Dystonia in musicians is still considered idiopathic but is likely multifactorial(1–3). The co-contraction of agonist and antagonist hand muscles in affected musicians is associated with inadequate reorganization of cortical finger representation areas and reduced sensory perception, resulting in temporal-spatial discrimination anomalies. It can develop in musicians with genetic predisposition, alterations in the basal ganglia circuit, or after prolonged and repetitive instrument practice, possibly associated with psychological factors such as anxiety disorders, perfectionism, and stress(4-6). Currently, the prevalence of dystonia in musicians is estimated to be 0.2-0.5%, with guitarists, pianists, and wind instrument players being the most commonly affected, accounting for 70% of patients. The onset of dysfunction rarely occurs during the initial development of instrumental technique but typically emerges after years of instrument practice when the musician is at the peak of their career. Affected musicians experience a sudden or insidious deterioration of their sensorimotor abilities, reporting a lack of coordination during performance often accompanied by involuntary flexion or extension of the fingers during musical passages that require rapid movements and control. Symptoms include cramping sensation, abnormal posture of the affected hand, finger curvature, and loss of coordination during specific activities(7). The loss of coordination is characterized by involuntary flexion of dystonic fingers or extension of adjacent fingers to compensate for abnormal movement, along with prolonged cocontraction of flexor and extensor muscles(8–11). This disorder most commonly affects fingers 3, 4, and 5 of the hand. Focal Hand Dystonia in musicians is a highly disabling condition, often impacting musicians' careers. Multiple strategies have been explored to manage dystonia in musicians, with highly variable results. The currently available data do not provide guidance for choosing a rehabilitative intervention plan. The objective of this scoping review is to present a map of the current treatment strategies and their temporal evolution, examining how rehabilitation approaches have adapted over time to integrate into increasingly effective therapeutic contexts.

METHODS

The present scoping review was conducted following the JBI methodology(12)for scoping reviews. The Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR)(13) Checklist for reporting was used. Research team: To support robust and clinically relevant results, the research team included authors with expertise in evidence synthesis, quantitative and qualitative research methodology, sport and musculoskeletal rehabilitation, experts in rehabilitation of the hand. Review question: We formulated the following research question: "What are the rehabilitation strategies and their temporal evolution for Focal Hand Dystonia in musicians, and how have they been integrated into increasingly effective therapeutic management contexts?". Eligibility criteria: Studies were eligible for inclusion if they met the following Population, Concept, and Context (PCC) criteria.

Population:

- Studies involving musicians diagnosed with Focal Hand Dystonia.
- Studies focusing on musicians from various instrument groups.

Concept:

- Studies investigating rehabilitation strategies or interventions for Focal Hand Dystonia in musicians in musicians.
- Studies exploring the effectiveness, outcomes, or impacts of rehabilitation approaches.
- Studies describing the evolution or changes in rehabilitation strategies over time.

Context:

- Studies conducted in any geographical location.
- Studies published in peer-reviewed journals.
- Studies published in the English language.
- Studies with available full-text articles.

Exclusion criteria: Studies that did not meet the specific PCC criteria were excluded. Search strategy: The following electronic databases were utilized: PubMed, PEDro, Cochrane Library, Trip, and Google Scholar. Additional studies were identified through bibliographic references and related articles. Contact was made with authors via email to identify additional sources and gain access to full-text articles if not readily available. In addition, grey literature (e.g. Google Scholar, direct contacts with experts in the field) and reference lists of all relevant studies were also searched. Searches were conducted on 20 December 2023 with no date limitation. The summary of these articles is presented in Table 1. The article selection process is depicted in a flow chart (Figure I). The entire selection process and reasons for the exclusion were recorded and reported according to the latest published version of the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA 2020) flow diagram. Data extraction and data synthesis : Data extraction was conducted using an ad-hoc data extraction form which was developed a priori, based on the JBI data extraction tool. Key information (authors, country, year of publication, study design, patients characteristics, PFD, type of intervention and related procedures) on the selected articles were collected. Studies identified and included were reported as frequency and percentage, and the description of the search decision process was mapped. In addition, extracted data were summarized in tabular form according to the main characteristics.

RESULTS

As presented in the PRISMA 2020-flow diagram (Figure <u>1</u>), from 190 records identified by the initial literature searches, 173 were excluded and 14 articles were included(Table1). Study selection: A total of 190 articles were identified through the search of the aforementioned electronic databases. These articles were initially screened based on their titles and abstracts, resulting in the exclusion of

172 articles that did not meet the eligibility criteria or were duplicates. The remaining 18 articles underwent a full-text review, during which 4 articles were excluded for the following reasons: the study population did not consist of musicians or the intervention was not relevant to rehabilitation. After this process, 11 articles were deemed eligible for inclusion. Additionally, 3 articles were obtained through manual searching of related articles and contacting the authors for additional information, bringing the total number of included articles to 14 in this review. The summary of the combined results from these 14 studies explored various treatment techniques for focal hand dystonia in musicians, focusing on motor and sensory recovery. Most showed improvements in motor control and reduction of dystonia symptoms. Techniques ranged from sensorimotor rehabilitation and orthosis use to mirror therapy and memorybased training. These personalized approaches suggest potential in reorganizing sensorimotor neural networks and enhancing musical performance.

Fig.1 Preferred reporting items for systematic reviews and meta-analyses 2020 (PRISMA) flow-diagram

Identification of studies via databases and registers

Identification



Screening



Studies included in review (n = 14)

Table.1 Main characteristics of included studies

Authors / Title	Year Study Type Population	Intervention	Outcome
Victor Candia et			Pianists and Guitarists:
	2002	Sensory Motor	
al. (14) "Sensory			Dystonia Evaluation Scale 2.4
	Case Series	Retuning	
Motor			to 3.9; Wind Players: Dystonia

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
Retuning: A	11 Musicians (6	Dystonia Evaluation	Evaluation Scale <1.4 (Similar
Behavioral	Pianists, 2 Guitarists, 3	Scale <1.8 (Severe to	to Pre-Treatment), No Benefit
Treatment for	Wind Players)	Mild Dystonia)	for Wind Players
Focal Hand		Pianists and	
Dystonia of		Guitarists: Dystonia	
Pianists and		Evaluation Scale >2.5	
Guitarists"		to 3.8; Wind Players:	
		DES 0.8 to 1.9 (Similar	
		to Pre-Treatment)	
Nancy N. Byl et			Significant improvements in
al. (15) "Effect of			somatosensory evoked
Sensory	2002	Soncorry	potentials (>86.8%), hand
Discrimination	2003		representation area (increased
Training on			over 20%), response
Structure and	3 Wind Musicians	Training	amplitude (improved by
Function in			40%), sensory discrimination
Patients With			(>22%), musculoskeletal

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
Focal Hand			performance (>31.9%), and
Dystonia: A			fine motor control (average
Case Series"			improvement 23.9%). Two
			out of three musicians
			returned to their musical
			careers.
			Scale for Quality of Effort:
			Improvement in non-dystonic
			hand from 3.12 to 3.78 points,
Rae de Lisle et			dystonic hand from 2.32 to
al. (16)	2006		3.54 points. Difficulty in
"Pianism	Case Series	Pianism Retraining	Hand Instrumental Exercise:
Retraining: A	3 Pianists		Significant decrease in correct
Case Series"			identification of dystonic
			hand by blind listeners from
			pre-treatment (79%) to post-
			treatment (28%). TRE test:

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			Increase from pre to post- treatment of 1.0 points on average, with variations among subjects. VER test: Overall increase of 1.3 points, with individual improvements detailed.
Katherine Butler et al. (17) "Focal Hand Dystonia Affecting Musicians. Part II: An Overview Of Current Rehabilitative	2006 Descriptive Study Musicians with Focal Hand Dystonia	Overview of Rehabilitation Interventions	Identified three main categories of rehabilitation approaches: 1) Multidisciplinary approach including sensory and motor treatments, rest from triggering activities, and occupational therapy. 2) Use of orthoses. 3) Supportive therapeutic approaches such

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
Treatment			as instrument modifications,
Techniques"			Feldenkrais and Alexander
			techniques, biofeedback
			therapy, psychotherapy, and
			prevention through
			organized practice and rest
			schedules. Future research
			areas in focal hand dystonia
			highlighted.
Nancy N. Byl et			Improvements in spatial and
al. (18) "Focal	2008		sensory discrimination (88%),
Hand Dystonia:		T . I I	target-specific performance
Effectiveness of	Case Series	Learning-based	(75%), and motor speed
a Home	13 subjects with Focal	Sensorimotor and	(76.5%). Functional
Program of	Hand Dystonia,	Memory Training	independence, fine motor
Fitness and	including 7 musicians		skills, and hand strength did
Learning-based			not show statistically

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
Sensorimotor			significant scores. Six months
and Memory			post-treatment: Most subjects
Training"			returned to work, with
			functional independence
			scores equivalent to healthy
			controls for most participants.
			Scale for Quality of Effort:
			Improvement in non-dystonic
Rae de Lisle et			hand by 0.20 points (95% CI [-
al. (19)			0.9, 0.49], p=0.17) and
"Pianism	2009	Pianism Retraining via	dystonic hand by 0.68 points
Retraining via	Case Report	Video Conferencing	(95% CI [0.23, 1.13], p=0.0037)
Video	1 Pianist	Video concrenenz	during in-person training;
Conferencing:			similar improvements in
A Case Report"			video conferencing. Difficulty
			in Hand Instrumental
			Exercise: Identification of
	(] '	1	il de la constant de

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			dystonic hand decreased significantly post-treatment in both in-person (from 95% to 44%) and video conferencing
			(to 62%) settings.
Patrice Berque			Significant decrease in
et al. (20) "A			abnormal movements over 12
Combination of			months (F = 6.32, p < 0.001).
Constraint-	2010		Improvements noted in
induced	Case Series	Combination of	Toronto Western Spasmodic
Therapy and	8 Musicians (6	Constraint-induced	Torticollis Rating Scale (F =
Motor Control	professionals 2 non-	Therapy and Motor	4.96, p < 0.001) and Arm
Retraining in	professionals)	Control Retraining	Dystonia Disability Scale (F =
the Treatment	professionals)		3.60, p = 0.004). Increased
of Focal Hand			performance speed noted in
Dystonia in			the first 6 months, with
Musicians"			variations in later months.

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			Dystonia Evaluation Rating:
			Improved from 1 (worst
			dystonia) pre-treatment to 2
			(slightly improved) post-
			treatment, and 4 (almost
Rae de Lisle et			normal) at follow-up.
al. (21)			Variable for Control of
"Rehabilitation	2012	I., . (Response: Improved from 1
of a cellist	Case Report	Datasinin a	pre-treatment to 2 post-
whose vibrato	1 Cellist	Kettaning	treatment (irregular sound on
was affected by			long notes), and 3 at follow-
focal dystonia"			up (controlled sound on long
			notes at steady speed).
			Violoncellist's Questionnaire
			Evaluation: Improved from
			1.14 (SD 0.52) pre-treatment
			to 2.82 (SD 0.86) post-

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			treatment, with sustained
			improvement to an average
			score of 2.75 (SD 0.89,
			p<0.0001) at follow-up.
Patrice Berque			Frequency of Abnormal
et al. (3) "A			Movements: 80% reduction in
Combination of			abnormal movements over
Constraint-			time for both combined pieces
Induced	2012	Combination of	(F=7.85, df=8, p<0.001).
Therapy and			Toronto Clinical Scoring
Motor Control	Case Series, Long-term	Constraint-Induced	System: No statistically
Retraining in	4 Musiciana Classical	Control Botacining	significant changes between
the Treatment	4 Musicians Classical	Control Ketraining	12 months and 4 years (p
of Focal Hand			values ranging from <0.001 to
Dystonia in			0.043). Arm Dystonia
Musicians A			Disability Scale: No
Long-term			statistically significant

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
Follow-up			changes between 12 months
Study"			and 4 years (p values ranging
			from <0.001 to 0.045).
			Metronome speed achieved
			by subjects without abnormal
			movements increased over
			time from about 30-100bpm
			for both pieces (F=20.45, df=8,
			p<0.001), with consistent
			improvement over time
			except in year 4 for the
			medium difficulty piece.
Katherine	2018		Improvement in scores at 3
Butler et al. (22)	Randomized	Sensory-motor rehabilitation therapy	and 6 months compared to
"Sensory–motor	Controlled Trial		baseline: Arm Dystonia
rehabilitation	15 subjects: 8		Disability Scale (0-100% scale,
therapy for	musicians with		higher scores indicate less

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
task-specific	Musician's Dystonia, 7		disability) improved from
focal hand	with Writer's Dystonia		66.07 to 70.0 (3 months) and
dystonia: A			69.25 (6 months); Toronto
feasibility			Western Spasmodic
study"			Torticollis Rating Scale (0-5
			scale, higher scores indicate
			less disability) improved from
			2.86 to 3.0 (3 months) and 3.43
			(6 months); Brief Illness
			Perception Questionnaire (0-
			80 scale, 80 indicates the most
			threatening disease
			perception) improved from
			50.67 to 47.25 (3 months) and
			44.33 (6 months); EQ-5D 5L
			(0-1 scale, 1 indicates full
			health) improved from 0.79 to

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			0.84 (3 months) and 0.88 (6
			months); Video (1-4 scale, 4
			indicates greater impairment)
			improved from 2.5 to 1.68 (6
			months); Clinical Global
			Impression Scale, 0-7, 7
			indicates much worse)
			improved from 3.08 (3
			months) to 2.58 (6 months).
			Qualitative interviews
			indicated effectiveness
			percentages for Sensory-
			motor reprogramming (86%),
			Slow-down exercise (71%),
			Mirror Therapy (43%),
			Shoulder and hand exercises
			(43%), while soft tissue

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			massage and ultrasound were
			not effective.
			Experts reported no
Riccardo Bravi			significant differences in
et al. (23)			effects between corrective
"Assessment of			Kinesiotaping and Sham for
the effects of			both criteria: "General
Kinesiotaping			Performance" and "Fingers
on musical	2019		Posture". Musicians self-
motor	Pilot Study	Kinesiotaping	assessed that Kinesiotaping
performance in	7 musicians		was ineffective in improving
musicians			general performance. Some
suffering from			subjects reported improved
focal hand			fine control during
dystonia a			performance, which might be
pilot″			attributed to the "sensory
			trick" phenomenon.

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			Results showed
			improvements in Toronto
			Western Spasmodic
			Torticollis Rating Scale
			(z=2.449, p<0.05) and Arm
Marina			Dystonia Disability Scale
Ramella, et al.			(z=2.000, p<0.05), but not in
(24) "Modified	2021	Madified Creded	Dystonia Evaluation Scale
Graded Motor	Case Series	Modified Graded	(z=1.414, p>0.05). At the end
Imagery for	6 musicians		of treatment, musicians
Musicians'			evaluated their overall
Focal Dystonia"			performances, selected pieces
			, and a C major scale as
			improved: Global
			Performance Score (z=2,214,
			p<0.05), Piece Performance
			Score (z=2,207, p<0.05), Scale

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			Performance Score (z=2,207,
			p<0.05).
			Tubiana rating scale scores
			improved for all participants.
			Participant 1: Pre-treatment
			score 3, post-treatment score 5
			(returned to concert
Bronwen			repertoire). Participant 2: Pre-
Ackermann et	2021	MusAARP: Anatomy-	treatment score 3, post-
al. (25)	Pilot Study	based Retraining	treatment score 5 (resumed
"MusAARP:	4 musicians	Program	playing and preparing for
Pilot Study"			exams). Participant 3: Pre-
			treatment score 3, post-
			treatment score 4 (took a
			break at six months due to
			breath-holding during
			practice, then continued with

	Year		
Authors / Title	Study Type	Intervention	Outcome
	Population		
			meditation and online retraining). Participant 4: Pre- treatment score 2, post- treatment score 5 (playing
			returned to normal).
G. Mohan			
Kumar et al.			Likert Scale Pre-test Scores:
(26) "Impact of			Group A - 38.86, Group B -
Alexander			38.92, Group C - 38.78 (no
technique	2022	Alexander Technique	significant difference pre-test
mirror therapy	2022 Comparative study 150 musicians	Mirror Therapy,	among groups). Post-test
versus			Scores: Group A - 22.76,
conventional		Conventional Inclupy	Group B - 32.62, Group C -
therapy on			28.32 (significant differences
musician's			post-test among groups,
cramp in			P≤0.001).
guitarists″			

The studies explored various rehabilitative approaches for musicians with focal hand dystonia. They include Sensory Motor Retuning, which was effective for pianists and guitarists, and a combined Constraint-induced Therapy and Motor Control Retraining approach, showing significant long-term improvements. Sensory Discrimination Training focused on sensory tasks, enhancing motor control. Learning-based Sensorimotor and Memory Training documented improvements in specific task performance. Instrumental Retraining adapted to different instruments showed promising outcomes. Sensory-motor rehabilitation therapy indicated gradual improvements. Kinesiotaping didn't show significant benefits, while the Alexander Technique seemed effective for guitarists. MusAARP and modified Graded Motor Imagery showed overall positive results in improving hand movements and posture. Each study presented a unique approach to managing focal hand dystonia, highlighting the complexity and diversity of treatment methods.

DISCUSSION

The diverse rehabilitative approaches for Focal Hand Dystonia (FHD) in musicians indicate a preference for multimodal intervention. Sensory Motor Retuning (SMR) has shown effectiveness for pianists and guitarists but not for wind instrument musicians. The integration of SMR with Slow-Down Exercise (SDE) addressed this limitation, showing improvement in wind musicians when combined with instrument playing. Sensory Discrimination Training and Learning-based Sensorimotor and Memory Training (LBSMT) have also contributed valuable insights. The use of orthoses in methods like Sensory-Motor Rehabilitation Therapy and modified Graded Motor Imagery (mGMI) raises questions about its applicability based on dystonia severity. Instrumental Retraining by Rae De Lisle, although requiring musical knowledge, bridges the gap between rehabilitation and music. The ineffectiveness of Kinesiotaping in reducing dystonic patterns, despite some subjective improvements, suggests a 'sensory trick' phenomenon. The Alexander Technique, compared to Mirror Therapy and conventional therapy, showed improvements in guitarists. Collaboration between therapists and instrument teachers could enhance preventive aspects of Focal Hand Dystonia management. The literature suggests multimodal rehabilitation is preferable for treating Focal Hand Dystonia in musicians. Studies highlight the effectiveness of Sensory Motor Retuning for pianists and guitarists, but less so for wind musicians. Combining Sensory Motor Retuning with Slow-Down Exercise improved outcomes for wind musicians, potentially due to the integration of instrument playing in exercises. The use of orthoses in newer therapies like Sensory-Motor Rehabilitation Therapy and modified Graded Motor Imagery raises questions about its suitability based on dystonia severity. Rae De Lisle's Instrumental Retraining suggests the need for therapy approaches bridging rehabilitation and music. Bravi et al.'s study on Kinesiotaping showed limited effectiveness. The Alexander Technique outperformed mirror therapy and conventional therapy in guitarists. Overall, recent studies incline towards multimodal approaches, with promising results in varied therapies like mGMI and MusAARP. However, further research is needed to refine these approaches, considering the high expectations of affected musicians and the diversity of methodologies in existing studies. The comprehensive literature review on Focal Hand Dystonia in musicians provides an evidence-based, holistic approach to understanding the condition. It includes various studies, suggesting a need for individualized treatment, interdisciplinary collaboration, and preventive education. However, it lacks detailed implementation strategies, may not generalize across all clinical settings due to varying patient characteristics, and is limited by knowledge as of September 2021. Crucially, the absence of randomized controlled trials is a significant gap, hindering the development of definitive treatment guidelines and underscoring the need for high-quality research in this area. In clinical practice for managing Focal Hand Dystonia in musicians, it's important to conduct comprehensive assessments, adopt a multidisciplinary approach, and create individualized treatment plans. Rehabilitation interventions should incorporate evidence-based practices like sensory-motor retraining and motor imagery. Education on selfmanagement, regular monitoring, and collaboration with music educators are crucial. Continuous professional development is also key to stay informed on the latest research and treatment advancements in Focal Hand Dystonia. In conclusion, Focal Hand Dystonia necessitates a multidisciplinary approach for effective management. Individualized treatment plans tailored to the patient's needs are essential. Rehabilitation interventions, including sensory-motor retraining and feedback techniques, show promise in Focal Hand Dystonia in musicians. Patient education and self-management strategies play a crucial role in long-term success. Regular monitoring and collaboration with music educators can enhance outcomes. Continuous professional development is vital for staying updated in Focal Hand Dystonia in musicians management.

Clinical messages

For treating Focal Hand Dystonia in musicians, a multimodal rehabilitative intervention is recommended, combining various therapeutic approaches to achieve better results.

Rehabilitation strategies should be tailored to individual needs, considering the severity and specific characteristics of the dystonia in each musician.

Emerging treatments and collaborative efforts between rehabilitation experts and music educators can enhance therapy effectiveness and facilitate a smoother return to performance.

FOUNDING

Authors declaration no foundings

CONFLICT OF INTERESTS

The authors are doctoral students, clinicians, who have no financial relationships with organisations that might have an interest in the work presented in the last 3 years and have no other relationships or activities that might influence the work presented.

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