Rehabilitation of focal hand dystonia in musicians: a systematic review of the studies

Rita Chiaramonte, Michele Vecchio

Introduction. Focal hand dystonia in musicians is a task-specific movement disorder characterized by an involuntary loss of control and coordination of finger movements during instrumental playing.

Materials and methods. Literature searches with the keywords 'dystonia' AND 'musician' AND 'finger' OR 'treatment' OR 'therapy' OR 'rehabilitation' were conducted in PubMed, EMBASE, Cochrane Library and Web of Science to perform the systematic review about the several strategies used to treat dystonia in musicians. The search was performed independently by two authors (R.C. and M.V.) from 6 April 2020 till 6 June 2020. The research identified a total of 423 articles. Seventy-seven selected articles were analysed by the reviewers. Thirty-six publications met the inclusion criteria and were included in the systematic review.

Results. The systematic review was performed to identify the main used treatments for dystonia in musicians. We defined the several techniques to better guide the physician to delineate a rehabilitation protocol adopting the better strategies described in the current literature.

Conclusion. This systematic review tried to provide to the reader a complete overview of the literature of all possible different treatments for dystonia in musicians. A correct protocol could permit to improve the motor performance and the quality of life of musicians.

Key words. Dystonia. Fingers. Musicians. Quality of life. Rehabilitation. Systematic review.

Department of Biomedical and Biotechnological Sciences; Section of Pharmacology; University of Catania (R. Chiaramonte, M. Vecchio). Rehabilitation Unit; AOU Policlinico Vittorio Emanuele; Catania, Italia (M. Vecchio).

Correspondence:
Dra. Rita Chiaramonte. Department of Biomedical and Biotechnological Sciences. Section of Pharmacology. University of Catania. Piazza Università, 2. 95124 Catania, Italy.

E-mail:
ritachiaramd@gmail.com

Accepted:
04.03.21.

How to cite this paper:
Chiaramonte R, Vecchio M. Rehabilitation of focal hand dystonia in musicians: a systematic review of the studies. Rev Neurol 2021; 72: 269-82. doi: 10.33588/rn.7208.2020421.

Versión en español disponible en neurologia.com

Introduction

Focal hand dystonia in musicians

Focal hand dystonia in musicians is a task-specific movement disorder characterized by an involuntary loss of control and coordination of finger movements during instrumental playing correlated to extensive and forceful use of the digits [1]. It is correlated to a co-contractions of agonist and antagonist muscles due to maladaptive cortical reorganisation and altered sensory perception with abnormalities in temporal and spatial discrimination [2,3].

The dystonia in musicians became a field of investigation only in the last 40 years, even if some authors asserted that previously famous composers were affected. In 1999 Richard J. Lederman [4] in his paper 'Robert Schumann' hypothesized that Schumann was affected by this disease. In 1988, Frank Wilson [5] had written 'musician in full flight is an operational miracle, but a miracle with peculiar and occasionally unpredictable vulnerabilities' and in 1989 he had published a pioneering article [6] on dystonia in musicians in which he sustained 'although the relationship of music and medicine is measured in millennia, it is only within the past few years that it has developed genuine vitality'.

Nowadays, we know that the prevalence of dystonia in musicians is 0.2% to 0.5% [1]. Every instrument group is affected, but guitarists, pianists and woodwind players are most commonly affected, representing 70% of patients [1].

The musicians have a sudden or insidious deterioration of sensorimotor skills, often only during instrument playing. Musicians often report incoordination while playing that is frequently accompanied by involuntary flexion or extension of fingers during music passages that emphasize rapid, forceful finger movements [7]. They presented involuntary spasms, cramping sensations, abnormal hand posture, finger curling, loss of coordination during specific task [1]. It more often involves digits 3, 4 and 5 (D3, D4, D5) of the hand and is thought to be related to the intense and prolonged practice of rapid, alternating, and highly precise finger movement patterns [1].

Disability condition and the role of treatment

The dystonia in musicians can be disabling to compromise the professional career. This condition does not need to be severe to exercise a significant impact on musician's career. Multiple strategies were explored to manage dystonia in musicians with highly variable outcomes. Rehabilitation goals include improvement of control of the movement and of sensibility perception. No specific program of therapy has been defined. The treatment is needed because cortical changes, together with emergent neurological dysfunction, can be redressed by context-specific treatment [8]. In addition, the use of a movement intervention capable of producing measurable changes in the cortical organization of sensory areas underscores the tight relationship between sensory and motor systems [8].

For musicians with dystonia, maintaining a good motor performance is hard and it has consequences on their careers. The presence of dystonia in musicians may constitute a negative factor in their quality of life.

Purpose

This study aimed to perform a systematic review of the several strategies for dystonia in musicians to investigate the different therapy intervention, aimed at normalising movement patterns, in musicians affected by dystonia. Unfortunately, the current literature does not show robust evidences that indicate which treatments work better, when they should start, their best duration or intensity. Until now, there are no specific guidelines.

This review defined the several techniques for the treatment of dystonia in musicians to better guide the physician to delineate a therapeutic protocol adopting the better strategies described in the current literature.

Materials and methods

Search strategy

The search was carried out on the following medical electronic databases: PubMed, EMBASE, Cochrane Library and Scopus Web of Science. The review was conducted from 6 April 2020 till 6 June 2020. We searched for the following terms and keywords: 'dystonia' AND 'musician' AND 'finger' OR 'therapy' OR 'treatment' OR 'rehabilitation'.

Selection criteria and data extraction

From 1989 to 2020, searches in the database produced for the keywords 'dystonia' AND 'musician' AND 'finger' yielded 60 references and for the search 'dystonia' AND 'musician' AND 'treatment' OR 'therapy' OR 'rehabilitation' respectively 159, 135 and 66 references, whose titles and abstracts were screened by the reviewers.

The selected papers remained for full-text screening were 77 and the eligibility of the study inclusion was assessed independently. Thirty-six publications met the inclusion criteria and were included in the systematic review. Forty-one articles were excluded for the following reasons: 20 studies were about different topics than ours, 8 described a different disorder from finger dystonia, 13 treated on a different professional category than musicians (Figure).

We included original articles about finger dystonia in musicians. We excluded animal studies and participants with other disorders. We also excluded all of the remaining duplicates.

Two reviewers independently screened the titles and abstracts from the initial search to identify relevant records and to identify eligible studies based on title and abstract. In the case of conflicting opinions, a consensus was reached after discussion between the authors. Selected full texts were then reviewed and included in the systematic review following the PRISMA protocol [9] and in accordance with the PICOS [10] (population, intervention, comparison, outcome, and study design) criteria, shown in Table I: participants were aging adult musicians; intervention was based on rehabilitation therapy; comparator was any comparator; outcomes included clinical assessments, diagnostic scales and instrumental tools; and study design was RCTs, case series and case report, retrospective studies.

Results

Description of the studies

The number of studies produced at each stage of the search for the systematic review is shown in figure.

A total of 36 studies were included in our systematic review with the sample characteristics and details of the design of each included study in Table I.

Variations of experimental conditions across the studies

The selected 36 articles were described on the basis of the several treatments used for dystonia in musicians. Characteristics of the studies are shown in table I.

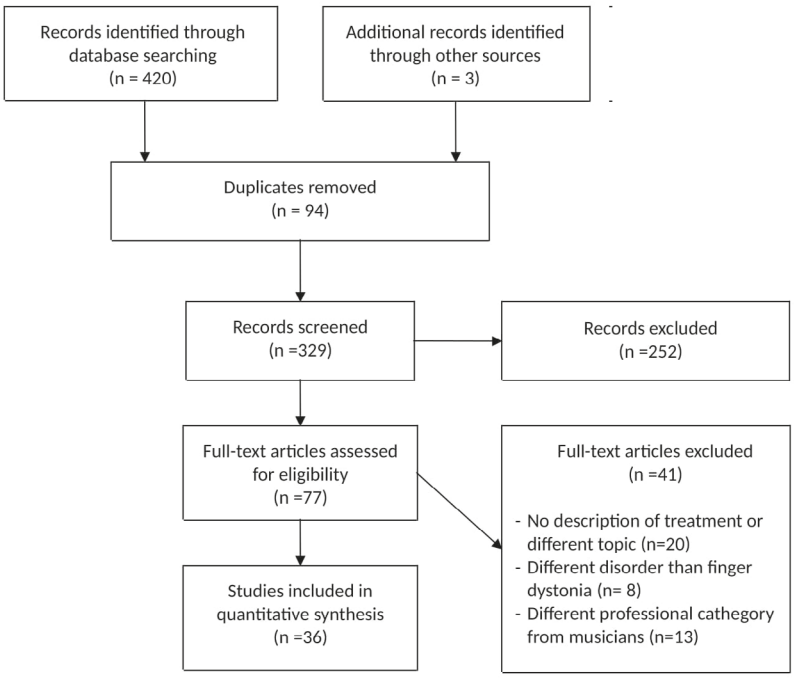
All study groups were not homogeneous for relevant general clinical features as affected fingers, duration of disease, kinds of diagnostic measures, severity of symptoms, rehabilitation therapy, time of starting therapy, duration of treatment (Table I).

Rehabilitation treatment

We showed all methods of the current literature used for the treatment of dystonia in musicians in Table II.

Botulinum injection is the most used therapy in literature [11-20]. Sensory re-education and sensory motor returning [21-25] and the combination of task specific motor training and neuromodulation [21,25-28] were used in five studies. Behaviour treatment [8,29-31], immobilization of fingers with splints in combination with specific finger exercises [25,29,30,32], transcranial direct current stimulation [26,27,33,34] were used in several articles too. Other treatments were less used, such as the constraint-induced therapy in combination with slowdown exercises [1,35,36], oral anticholinergic medication [14,37], proprioceptive training [38,39]. In only one study the following therapies were described: kinesiotaping [40], motor fatigue exercises [41], oral intake of Δ9-tetrahydrocannabinol [37], surgical treatment [42]. The outcomes of the different types of therapy are resumed in Table I.

Figure. Flowchart of the process of initial literature search and extraction of studies meeting the inclusion criteria.



Discussion

Treatment of musicians' dystonia

Hand dystonia seems to be related to the excessive performance of repetitive activities and has been treated in different ways. Treatment is based on using multiple strategies for the management of the dystonia, with variable results. Although a specific therapy has not been defined, several positive outcomes are shown.

Task-specific motor training and neuromodulation

Task-specific motor training combined with neuromodulation was employed in dystonia in musicians to promote proper body alignment and prevent dystonic patterns during motor performance [26-28,34].

Sensory re-education and sensory motor returning

Significant improvement in motor control, accuracy, sensory discrimination and physical performance was described after specific exercises of sensory re-education and sensory motor returning [21-25,35,43]. Sensory motor returning (SMR) could be used both as only intervention [30,31,36] or in combination with other approaches [1,35]. Candia et al [30] developed a novel treatment intervention, a combination of sensory motor retuning (SMR), behavioural treatment and constraint-induced therapy. By immobilising one or several digits 'compensating' for the dystonic movements, the 'dystonic' finger was required to carry out repetitive exercises in coordination with the remaining free digits [30]. Even if improvement in motor performance were described, the program was not sufficient to return musicians to professional levels of performance [22].

Table I. Treatment of dystonia in musicians: characteristics and outcomes of studies included in the systematic review.

	Study design	Dystonia location	Dystonia duration	Participants	Intervention/ treatment	Outcome measures	Intervention frequency	Clinical results
Berque et al, 2010	Case series	D2, D3, D4, D5	~ 9.2 y	2 guitars, 2 flutes, 2 bagpipes, 1 oboe player, 1 accordion	CIMT and SDE at slow speed	Video recordings, FAM, change in metronome speed	Up to 30 min/d for 12 m. Only the first w of retraining involved CIMT, for 2 h/d. Each subject would play each sequence continuously for 10 min, with a 2-min rest between sequences. A 10-min of rest was given after the first series of 5 sequences was completed. Speed of metronome was modified every 2 min since the abnormal movement pattern occurring	Significant ↓ in the number of abnormal movements per second of instrumental playing over the 12-month period.
Berque et al, 2013	Case series	D2, D3, D4, D5	~ 11.6 y	2 guitarist, 2 flutes players	CIMT and SDE at slow speed	Video recordings, FAM, TCS, ADDS, change in metronome speed	Up to 30 min/d for 12 m	Significant ↓ in the number of abnormal movements per second of instrumental playing over 36 m.
Betti et al, 2018	Case study	D1, D2, D3	~ 3 y	1 guitarist	rTMS	Movement time (time interval between onset and offset), MGA (maximum distance reached by the 3D coordinates of the thumb and index finger) TMGA (time at which the distance between the 3D coordinates of the thumb and index finger was maximum from movement onset) TMGV (time at which the tangential velocity of the 3D coordinates of the thumb and index finger was maximum from movement onset) TMWH (time at which the 3D coordinates of the wrist were maximum from movement onset) TMWD (time at which the deceleration of the 3D coordinates of the wrist was maximum from movement onset) DG (time interval between the onset of the wrist movement and the onset of fingers' opening) Motor threshold	1 Hz of rTMS for 5 d	Significant difference in Finger Flexion Task, TMGV, TMWD, DG.
Bravi et al, 2019	CT	D1, D2, D3, D4, D5	8.7±9.7 y	1 harpsichord, 1 guitarist, 1 violinist, 3 pianist, 1 clarinettist	Correction Kinesiotaping versus sham Kinesiotaping	EMG	–	No significant differences in effects between Correction Kinesiotaping and Sham Kinesiotaping were reported by the experts, either for general performance or for fingers' posture. Musicians estimated that Correction Kinesiotaping was ineffective in improving their musical abilities

Table I. Treatment of dystonia in musicians: characteristics and outcomes of studies included in the systematic review *(cont.)*.

	Study design	Dystonia location	Dystonia duration	Participants	Intervention/ treatment	Outcome measures	Intervention frequency	Clinical results
Butler et al, 2018	CT	D3, D4, D5	~ 2.5 m	4 pianist, 2 guitarist, 1 flutist	Sensory–motor task-specific dystonia rehabilitative program	ADDS, TCS, BIPQ, EQ-5D 5L, CGI	Six therapy sessions over a 6-m period. Forearm muscles stretch to decrease myofascial tension, shoulder exercises to increase freedom of movement in the shoulder girdle, hand strengthening exercises to increase proprioceptive awareness and strength of intrinsic hand muscles.	Improvements occurred across measures and were typically larger at the 6-m
Buttkus et al, 2010	Case report	Finger flexion dystonia	~ 15 y	1 piano	A: Retraining strategies B: Retraining strategies + tDCS	MIDI-based scale analysis.	20' per d, 5 d consecutively for 6 w	Beneficial effect of retraining on fine motor control enhanced by cathodal tDCS
Byl et al, 2000	Case series	Fingers	1-5 y	12 p, whose 5 musicians (not specified the instruments)	Sensory training with biofeedback, mirror therapy, mental practice techniques	A battery of sensory, motor, physical and functional performance tests	Once a w for 6 m	Significant gains in pain, posture, balance, neural tension, strength and range of motion of fingers
Byl et al, 2003	Case series	D3, D4, D5	–	2 flutists, 1 bagpipe player	SDE and motor training	Magnetoencephalography and clinical sensory and motor tests task-specific motor control: Purdue Pegboard test, digital reaction time, line tracing accuracy and time Sensory discrimination: localization, 2-point discrimination, graphesthesia, stereognosis, kinesthesia musculoskeletal performance: ROM, posture, neural tension, independence: functional independence, work status	1p: 2 times a w for 12 w and reinforced daily at home. 1 p: 1 d for 2 w 1 p: 1 w for 17 w	Improved an average of 86.8% on somatosensory hand representation, 117% on target-specific performance, 23.9% on fine motor skills, 22.7% on sensory discrimination, 31.9% on musculoskeletal skills, and 32.3% on independence.
Byl et al, 2009	RS	–	>3 y	8 musicians	Home program of fitness and learning-based sensorimotor and memory training	Functional independence and strength	8 w	Task practice plus learning based memory and sensorimotor training can improve MD
Candia et al, 1999	Case series	–	–	3 pianists, 2 guitarists	CIMT	Non-invasive neuroimaging technique (magnetic source imaging), DES		Significant ↑ in the smoothness of movements after treatment
Candia et al, 2002	Case series	D2, D3, D4, D5	~ 7 y	6 pianists, 2 guirarists, 2 flutes, 1 oboe	SMR	DES, Dexterity and Displacement Device	2 h per day over a period of 8 consecutive days 1-month follow-up	SMR is of value for the treatment of focal hand dystonia in pianists and guitarists
Candia et al, 2003	Case series	Fingers	~ 3 m	11 pianists	Behavioural treatment	Oldfield handedness questionnaire and DES	8 consecutive d for 1.5 to 2.5 h per sessions	Significant ↑ in DES scores and in smoothness of movement
Candia et al, 2005	Case series	Fingers	–	101 musicians: pianists, guitarist, flautists, oboist	SMR, hand splint	DES	8 consecutive d for 1.5 to 2.5 h per session	Pianists and guitarists visibly improved the performance. The wind players did not improve

Table I. Treatment of dystonia in musicians: characteristics and outcomes of studies included in the systematic review *(cont.)*.

	Study design	Dystonia location	Dystonia duration	Participants	Intervention/ treatment	Outcome measures	Intervention frequency	Clinical results
Chen et al, 1998	RS	*Frontalis* or *abductor digiti minimi*	69 m	1 musician, 81 y	Botulinum toxin injections	5-point scale of benefits, VAS	23 injections, 130 U.	Benefits of botulinum injections for 6 m
Cohen et al, 1989	RS	D1, D2, D2, D4, D5	~ 7 y	1 pianist, 2 guitarist, 1 clarinet	Botulinum toxin injections	EMG	Every 2 w increasing doses (up to 20 U the first w, up to 40 U the second w, and up to 80 U the third w)	Benefit lasted between 1 and 6 m and was reproducible.
Cole et al, 1995	CT	D3	~ 6.5 y	1 bagpipes player, 1 pianist	Botulinum toxin injections	MRC scale, videotaped performance	3 injection with 5 and 10 U respectively for each patient	The efficacy of botulinum toxin lasted for 1 to 4 months
Furuya et al, 2014	RS	D2, D3, D4	~ 7.9 y	20 pianists	TMS	Rhythmic variability of the key strokes at pretest, post-test, and testing baseline	5 sessions	Therapeutic effects of behavioral training assisted by bihemispheric, noninvasive brain stimulation restored fine motor control
Horisawa et al, 2013	RS	D1, D2, D3, D4, D5	8.3 ± 8.2 y	1 koto player, 7 guitars, 2 shamisen, 3 pianists, 1 flutist 1 violinist	Ventro-oral thalamotomy	Tubiana's musician's dystonia scale	Surgery	Ventro-oral thalamotomy remarkably improved MD and the effect persisted for a long time
Jabusch et al, 2004	Case report	D3, D4, D5	~ 10 y	1 Pianist	THC	MIDI	After administration of THC or placebo, the patient played the piano for 3 h with short intermissions every 45 min.	Significant ↑ of motor control. After administration of placebo, motor control of the affected hand showed no improvement
Jabusch et al, 2005	RS	*Flexor digitorum superficial, profundus, flexor carpi radialis, flexor pollicis longus, extensor digitorum, extensor indicis, interosseus palmaris*	~ 9 y	144 musicians: keyboard players, woodwind players, guitarists	Botulinum toxin, Dysport (n. 71 p, 48%), Trihexyphenidyl (n. 69 p, 49%) Pedagogical retraining (n. 24 p, 17%) Ergonomic changes (n. 51p, 35%) Nonspecific exercises on the instrument (n.78 p, 54%)	Standardized questionnaire about the nature and duration of therapies	Trihexyphenidyl per 9±7 m: dosage 10 mg/day±1-13 BT: In 18 p one injection In 53 p (37%): 5.7±12 injections per patients; range, 2–25 Duration:16±29 m. Interval between treatment sessions of 4.2±12 m. Dosage per treatment session:128±218 U. Ergonomic changes for 35 m Pedagogical retraining for 28 m	In 77 p (54%): alleviation of symptoms: In 33% with trihexyphenidyl, 49% with BT, 63% with ergonomic changes 50% with pedagogical retraining, 56% with unmonitored technical exercises,
Jabusch et al, 2009	Case series	–	–	19 pianists	Instruments practice	Standardized questionnaire about the nature and duration of therapies and musical instrument digital interface-based scale analysis	27 m	Maintenance of motor skills was significantly influenced by practice quantity
Karp et al, 1994	RS	–	5±1 y	4 pianists, 4 guitarists, 2 drum players, 2 violinists, 2 trump players, 2 flute players, 1 clarinettist, 1 organist, 1bagpipes player	Botulinurn injections	Performance scale, MRC	The initial dose of BT was 2.5 to 20 U per muscle. The mean dose of BT used in a session was 26 ± 2 U. Benefit from each session lasted a mean of 6 ± 1 m. Weakness lasted a mean of 3 m.	BT injection is safe and effective for the long-term management of focal hand dystonia.

Table I. Treatment of dystonia in musicians: characteristics and outcomes of studies included in the systematic review *(cont.)*.

	Study design	Dystonia location	Dystonia duration	Participants	Intervention/ treatment	Outcome measures	Intervention frequency	Clinical results
Lungu et al, 2011	RS	–	>20 y	2 pianists, 1 guitarist, 1 drum player, 1 trumpet player	BT	Self-reported scales of benefit and weakness	Period between injections 19.9±12.4 m The patients received a higher mean dose at the end of the follow-up period compared to the initial treatment (49.9 vs. 24.9 U)	Botulinum injections are safe and effective after more than a decade of treatment
McKenzie et al, 2009	CT	–	1-10 y	14 p	Sensorimotor training	Sensory discrimination and fine motor speed	8 w	Effectiveness of the treatment
Pesenti et al, 2001	RS	–	2.8 ± 0.4 y	4 guitarists, 1 violinist, 3 pianists, 1 accordionist, 1 harpist	Motor fatigue	Handgrip test	Handgrip contraction lasts a mean of 2.13 ± 0.47 min	In dystonic musicians, a fatiguing contraction significantly improved motor performance. Five minutes after the contraction ended the benefit progressively disappeared.
Priori et al, 2001	RS	–	3.62 ± 2.36 y	4 guitarists 1 pianist 2 drummers	Limb Immobilization	ADDS, TCS, Self-rating scale	Patients wore the splint throughout the 24 h and removed it only once a w for brief (10 min) local hygiene. Duration 4.5 w	Four w after removal of the splint, all the patients had regained normal voluntary control of the hand with normal strength
Rosenkranz et al, 2008	Case series	D4	–	5 pianists	Proprioceptive training	SICI, BMF, TCS	15 m/d	Proprioceptive training improves hand motor function
Rosenkranz et al, 2008	Case series	D2	~ 7.8 y	1 trumpet/ pianist, 1 violinist/ pianist 1 violinist, 2 frenchhorn / pianists, 1 clarinettist	Proprioceptive training	BFM scale	Time spent playing h/d 3.33	Behavioral proprioceptive training improves hand motor function
Ross et al, 1996	RS	D2, D3	~ 5.8 y	6 pianists, 2 drummers, 1 horn player, 1 saxophonist, 1 guitarist	BT	EMG	Smaller muscles injected with 5–15 U. Larger muscles injected with 60–100 U. The dose was administered in one injection site, and the concentration of botulinum toxin was always 5 U/0.1 cc.	BT had success to treat MD
Rosset-Llobet et al, 2011	Case report	D3, D4, D5	10 y	1 pianist	SMR	Performance score, EMG, brain NMR	Every day for a y.	The patient returned to high-level piano playing, and after 8 y of follow-up, performance remains normal
Rosset-Llobet et al, 2015	CT	D2, D3, D4	18.77± 10.8 y	16 guitarists, 10 guitarists	tDCS	DSR	2-w course of neurorehabilitation based on sensory motor retuning therapy coupled with either real or sham tDCS for the first 30 min of each daily 1-h therapy session (total 10 sessions)	Both groups significantly improved their dystonia severity score during the 2 w of therapy.

Table I. Treatment of dystonia in musicians: characteristics and outcomes of studies included in the systematic review *(cont.)*.

	Study Design	Dystonia location	Dystonia duration	Participants	Intervention/ treatment	Outcome measures	Intervention frequency	Clinical results
Schuele et al, 2004	Case series	Forearm flexors	~ 10.6	12 strings, 20 keyboards, 25 guitars, 25 woodwinds, 2 brass	BT	Standardized questionnaire about the nature and duration of therapies, self-rating scale, six-step self-sating Score	Mean duration of treatment: 23 m Mean interval between injections 3.8 m. Number of injections per patient 7.4 Number of muscles injected per treatment: 2.5 (range 1 to 6). Total dose per treatment was 126.9 U (range 5 to 420) at the initial visit and 112.2 U (range 3 to 1,000) at the last visit. Average dose per muscle group at last visit was 55 U for shoulder muscles (only 2 patients), 47.4 units for forearm flexors, 31.4 for forearm extensors, and 17.7 for hand muscles	In 58 p: improvement after the injections. In 36% of p: long-term benefit
Van Vugt et al, 2013	RS	–	~ 10.1 y	54 pianists	BTXA (53%), trihexyphenildyl (51%)	Standardized questionnaire about the type and effectiveness of therapies	BT (53%), 4.1±3.0 injection session for 25.4±21.8 m, 24.1±17.6 U per session Trihexyphenidyl (51%), 21.5±17.8 m (5.02±3.7 mg/d) Retraining (87%) 41.0±38.9 sessions in 38.2±29.5 m (1.4±1.1 sessions per m) Hand therapy (42%) Relaxation techniques (38%) Physiotherapy (30%) Psychotherapy (23%) Acupuncture (21%) Body techniques (21%).	50% of patients improved task performance following participation in a variety of intervention strategies, but subjectively, 80% of subjects reported improvement after retrained and BT more than Trihexyphenidyl.
Vecchio et al, 2012	Case report	D4	~ 4 m	1 guitarist	Levocarnitine + BT	EMG, CT, MRI, transcranial magnetic stimulation, Fahn dystonia scale	levocarnitine (3 g daily for 3 months) + BT injected into the superficial (20 IU) and deep fl exor (20 IU) muscles	The contractions diminished 5 d later the BT injection. The beneficial effect of BT lasted for about 8 m
Yoshie et al, 2015	Case report	D3, D4, D5	~ 4 m	1 pianist	Slow-Down Exercise	EMG	30 min per d for 12 m.	Effectiveness of slowdown exercise. Behavioral intervention can reverse the reorganization of sensorimotor neural networks
Zeuner et al, 2005	Case series	–	14.6±7.0 y	1 guitarist, 9 writers	Motor training program and splint.	Kinematic Analysis, EEG, Transcranial Magnetic Stimulation	Finger flexor splints and others extensor finger splints. In addition, they were instructed to train each finger individually by making clockwise and counter clockwise circles, the letters II and mm on a piece of paper. During the first week, they practiced 25 min per d, 5 min with each finger, in the remaining 3 w 50 min/d with 10 min per finger.	Improvement after training

ADDS: arm dystonia disability scale; BFM: Burke-Fahn-Marsden; BT: botulinum toxin A; CIMT: constraint-induced movement therapy; CT: clinical trial; d: day; D: finger; DES: dystonia evaluation scale; DG: delay grasping; DSR: dystonia severity rating; f: females; FAM: frequency of abnormal movements scale; FHD: focal hand dystonia; h: hours; HC: healthy controls; m: males; MGA: maximum grip aperture; MIDI: MIDI-based scale analysis; min: minutes; MRC: Medical Research Council; n.: number; OS: observational study; p: patients; ROM: range of motion; RS: retrospective study; RT: retrospective study; rTMS: repetitive transcranial magnetic stimulation; SDE: motor control retraining; SICI: ahort interval intracortical inhibition; SMR: sensory motor retuning; TCS: Tubiana and Chamagne scale; tDCS: transcranial direct current stimulation; THC: Δ9-tetrahydrocannabinol; TMGA: time of maximum grip aperture; TMGV: time of maximum grip velocity; TMWD: time of maximum wrist deceleration; TMWH: time of maximum wrist height; U: units; VAS: visual analogue scale; w: week; y: year/s.

Table II. Treatments of focal hand dystonia in musicians.

	Treatment	Aim of the therapy	References
Behaviour treatment	Finger exercises with feedback	Improvement of somatosensory cortical organization	Candia et al, 1999; Candia et al, 2002; Candia et al, 2003; Candia et al, 2005
Constraint-induced therapy + slow-down exercise therapy	Splint and slow-down exercise	Reversing of the reorganization of sensorimotor neural networks	Berque et al, 2010; Berque et al, 2013; Rosset et al, 2016
Immobilization of fingers	Immobilization with splints and combinations of specific finger exercises	Reducing co-contraction	Candia et al, 1999; Candia et al, 2002; Priori et al, 2001; Zeuener et al, 2005
Kinesiotaping	Passive method for muscle activity normalization	Decrease in pain, but no improvement in motor abilities	Bravi et al, 2019
Motor Fatigue exercises	Fatiguing handgrip contraction	Improvement in motor performance, finger coordination and movement accuracy	Pesenti et al, 2001
Pharmacologic therapy	Oral anticholinergic medication	Improvement in motor control	Jabush et al, 2004; Jabush et al, 2005
	Δ9-tetrahydrocannabinol	Improvement in motor control	Jabush et al, 2004
	Botulinum toxin injections	Control of muscle spasms	Chen et al, 1998; Cohen, 1989; Cole et al, 1994; Jabush et al, 2005; Karp et al, 1994; Lungu et al, 2011; Ross et al, 2005; Schuele et al, 2005; Van Vugt et al, 2014
	Botulinum toxin injections + levocarnitine	Beneficial effect in MD	Vecchio et al, 2012
Proprioceptive training	Vibratory stimulation	Enhancing of sensorimotor organization and proprioceptive awareness	Bluter et al, 2018; Rosenkranz et al, 2008
Sensory re-education and SMR	Exercises of palpating and visualising size, weight and texture of different objects, intensive sensory training or sensory deprivation. Positive reinforcement was provided by verbal, visual, tactile, or auditory feedback	Improvement in sensory discrimination. Promotion of somatosensory reorganization and subsequent motor improvement. Redefining spatial and temporal processing capacities in the sensory and motor cortices in order to restore task-specific skills	Byl et al, 2000; Byl et al, 2003, Byl et al, 2009; McKenzie et al, 2009; Zeuener et al, 2005
Surgical treatment	Ventro-oral thalamotomy	Improvement of finger movement	Horisawa et al, 2013
Task-specific motor training + Neuromodulation	Hand activities in the prone, supine, sitting, and standing position.	Promotion of body alignment and prevention of dystonic patterns. Normalising movement patterns	Buttkus et al, 2010; Byl et al, 2000; Furuya et al, 2014; Jabush et al, 2009; Zeuner et al, 2005
tDCS	Stimulating electrode over the primary motor cortex.	Support to training, increase rehabilitation effectiveness	Betti et al, 2018; Buttkus et al, 2010; Furuya et al, 2014; Rosset-Llobet et al, 2015

MD: musician's dystonia; SMR: sensory motor returning; tDCS: repeated transcranial direct current stimulation.

Behaviour treatment

Candia and his collaborators had a particular interest for the treatment of focal dystonia in musicians [8,29,31]. Since behavioural mechanisms apparently underlie both the cortical disorder and the involuntary incoordination of movement, it was thought to be possible that a behavioural intervention could be of value in reducing or eliminating these conditions [29].

Constraint-induced therapy, slow-down exercise therapy and immobilization

In two studies of Candia et al [29,30] and of Berque [1,35], splints were used to immobilise the wrist and the adjacent fingers to the dystonic one. The finger that trained was not splinted and retraining included combinations of specific finger exercises during playing the instruments [29,30].

Zeuner et al [25] and of Priori et al [32], described their task specific motor training with good outcome for dystonia in musicians. Priori et al [32] concluded that limb immobilisation was a simple, effective and safe treatment for focal dystonia, but the duration of the outcomes were variable.

However, prolonged immobilization could be associated with transient side effects and variable outcomes in writer's cramp and musician's dystonia [32,41].

A combination of intensive constraint-induced therapy and specific motor control retraining at slow speed is described as a successful strategy for the treatment of dystonia in musicians [1,35,36], but in this way, it is difficult to establish which aspect of treatment had the more beneficial effect [1].

A 1-year period of this intensive constraint-induced therapy and motor control retraining at slow speed could be a successful strategy with longterm, lasting effects for the treatment of dystonia in musicians [35].

Yoshie et al [44] confirmed the effectiveness of slowdown exercise for the treatment of focal hand dystonia and of behavioural intervention, these treatments could reverse the reorganization of sensorimotor neural networks in dystonic patients [44].

Proprioceptive training

Rosenkranz et al [39,45] tested the effect of vibratory stimulation on the pattern of sensorimotor organization in healthy non-musicians, healthy musicians, patients with musician's dystonia, and patients with writer's cramp. The proprioceptive training improved the dystonic pattern [39,45].

Bluter et al [38] described a rehabilitation program that included proprioceptive exercises. This intervention was feasible, with high retention, adherence and acceptability [38].

Repeated transcranial direct current stimulation (tDCS)

tDCS might be a safe and noninvasive tool to assist retraining and might improve retraining effects in pianists [26,27,33]. This technique does not interfere with the neurorehabilitation procedure and can increase therapy effectiveness in rehabilitation patients [34]. The study of Buttkus et al [26] and Furuya et al [27] has a sample very small (only a musician) to obtain significant results.

Comparison between tDCS with and without training revealed that tDCS alone is not effective for improving motor performance in musician's dystonia [27].

Kinesiotaping

Use of kinesiotape over affected muscles, a passive method for muscle activity normalization, led to significant decrease in pain and somatosensory temporal discrimination threshold in comparison to sham tape in cervical and focal hand dystonia [46]. The study of Pelosin et al [46] did not specify if the participants were musicians, for this reason the study was not included in the review.

According to Bravi et al [40], musicians estimated that correction kinesiotaping was ineffective in improving musical abilities.

Motor fatigue exercises

According to the study of Pesenti et al [41], in dystonic musicians, after a fatiguing handgrip contraction lasting a few of minutes on the affected side, motor performance variably improved for a short time. Although the fatiguing task led to a subjective feeling of clumsiness of the arm and forearm, it improved finger coordination and movement accuracy in the affected fingers [41].

Pharmacologic therapy

Oral anticholinergic medication and Δ9-tetrahydrocannabinol

The results after anticholinergic medication administration were discouraging, with improvement being reported in only 33% of patients [14].

In the study of Jabusch et al [37], the results provide evidence that after single dose of 5 mg of Δ9-tetrahydrocannabinol (THC), motor control of the affected hand was significantly improved in a pianist suffering from focal dystonia. After administration of placebo, motor control of the affected hand showed no improvement [37].

Botulinum toxin injections

Uncontrollable muscle spasms can sometimes be controlled with repeated injections of botulinum toxin into the affected muscles to reduce the intensity of the contractions and relieve the discomfort of cramping [13,14,19]. One of the first article on the use of botulinum toxin injection in musicians'dystonia was the study of Cohen et al [12]. It provides preliminary evidence that botulinum toxin is helpful in treating many patients with hand cramps. Several studies [15,16,18] provided the evidence that botulinum toxin offers long-term benefit for some instrumental musicians with playing-provoked, focal limb dystonia [18].

Unfortunately, this type of treatment does not restore normal motor control, and patients often cannot return to previous jobs that require fine motor skills [14]. Improvement in performance was associated with some degree of weakness [12]. After the BT injection, 49% of patients reported an improvement and 57% received more than one injection [14]. Repeated injections are necessary [14].

Proper selection of the muscles to be injected and the use of electromyography are fundamental aspects to be taken into consideration. According to the study of Ross et al [17], the most common patterns of toxin spread were from *flexor digitorum sublimis* to *profundus, extensor carpi radialis* to *extensor digitorum communis*, and *extensor indicis proprius* to *extensor pollicis brevis*. Spread to adjacent uninjected muscles was a major factor contributing to suboptimal outcome for the consequent finger weakness [17]. In the study of Chen et al [11], *frontalis* and *abductor digiti minimi* were treated with botulinum toxin type F, with good outcomes. In the study of Vecchio et al [20] a guitarist with uncontrolled movement of the right index finger assumed levocarnitine (3 g daily for 3 months) to decrease cramps and botulinum toxin injected into the superficial (20 IU) and deep flexor (20 IU) muscles of the right index finger under electromyographic and ultrasonographic guidance. The contractions diminished 5 days later and the beneficial effect of botulinum toxin lasted for about 8 months [20].

Surgical treatment

According the study of Horisawa et al [42], the ventro-oral thalamotomy improved musician's dystonia and the effect persisted for a long duration.

Multidisciplinary team and psychological support

After a period of not playing, musicians must return playing with slow graded progression in duration and complexity of movements, often with psychological support.

The multidisciplinary team was proposed by several authors, including the participation a psychotherapeutic approach in many cases [14,34].

Summary of findings and Implication in rehabilitation

The rehabilitation for dystonia in musicians is variable in methodology and quality, intensity and duration. Also, there are no different specific protocols on the basis of the gravity of the disorder. The focus of therapy for mild, moderate and severe dystonia did not differ significantly for clinicians, even because musicians lose their virtuosity during the exhibitions even when the symptomatology is mild. Another non-homogeneous and very variable feature of the therapy is its beginning after the beginning of the disorder.

Musicians are extraordinarily motivated to pursue treatment, and they persevere in their search for an effective therapy. The treatment for dystonia in musicians could be very important to stimulate and promote the improvement of fine movements, to avoid the end of important carers with consequence worsening of quality of life, to sustain the mood of the subjects. The musicians need a complete recovery of the motor performance to recovery the lost virtuosity during their concerts.

Limitations

A lack of uniformity among the papers (measured parameters and assessment scale) may affect the outcomes of considered articles. The absence of information about some clinical characteristics, that could influence motor performance, represents another limitation, such as comorbidities affecting movements, the use of drugs, psychologic traits such as performance anxiety. Furthermore, in some articles the sample was very small. Several studies did not assess participants educational status, it could be a confounding factor and could influence the results.

Conclusion

Dystonia in musicians is a condition that leads to reduced performance levels and often the end of a musician's career. So, it is essential found a therapeutic solution. Several treatments, also in combi-

nation, could be promising tools with therapeutic potential. This systematic review tries to provide to the reader a comprehensive overview of the literature of all possible different treatments for dystonia in musicians.

Unfortunately the Wilson's theory [6], according whom are 'required months of lessons concentrating on rebuilding a 'healthy' movement strategy on the instrument before movements began to become normal', seems optimistic; many musicians have compromised their careers, as reported by Oliver Sacks in *Musicophilia* [47], such as the virtuosos of the piano Gary Graffmann, who definitively stopped his activity, Leon Fleisher who continued to play with only one hand and who claimed 'dystonic once, dystonic forever' [47].

We believe that the development of different options of treatment and rehabilitation in musician's cramp demonstrate, in some way, that there is no established and certainly effective therapy that restores complete professional functionality to the musician. Anyhow, a correct strategy could permit to improve motor performance and the quality of life of musicians. We would like to have created a guide for choosing a rehabilitation protocol combining more treatments and personalising the rehabilitation intervention.

Further researches in this area are required, such as large-scale, controlled trials to compare the different rehabilitation strategies and find a better protocol for this disorder.

References

1. Berque P, Gray H, McFadyen A. A combination of constraint-induced therapy and motor control retraining in the treatment of focal hand dystonia in musicians. Med Probl Perform Art 2010; 25: 149-61.
2. Altenmüller E, Jabusch HC. Focal dystonia in musicians: phenomenology, pathophysiology, triggering factors, and treatment. Med Probl Perform Art 2010; 25: 3-9.
3. Lin PT, Hallett M. The pathophysiology of focal hand dystonia. J Hand Ther 2009; 22: 109-14.
4. Lederman J R. Robert Schumann. Semin Neurol 1999; 19 (Suppl 1): S17-24.
5. Wilson FR. Teaching hands, treating hands. Piano Q 1988; 141: 34-41.
6. Wilson FR. Acquisition and loss of skilled movement in musicians. Semin Neurol 1989; 9: 146-51.
7. Wilson FR, Wagner C, Hömberg V. Biomechanical abnormalities in musicians with occupational cramp/focal dystonia. J Hand Ther 1993; 6: 298-307.
8. Candia V, Rosset-Llobet J, Elbert T, Pascual-Leone A. Changing the brain through therapy for musicians' hand dystonia. Ann N Y Acad Sci 2005; 1060: 335-42.
9. Shamseer L, Moher D, Clarke M, Ghersi D, Liberati A, Petticrew M, et al. Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. BMJ 2015; 350: 7647.
10. Higgins JPT, Altman DG, Gøtzsche PC, Jüni P, Moher D, Oxman AD, et al. The Cochrane Collaboration's tool for assessing risk of bias in randomised trials. BMJ 2011; 343: 889-93.
11. Chen R, Karp BI, Hallett M. Botulinum toxin type F for treatment of dystonia: long-term experience. Neurology 1998; 51: 1494-6.
12. Cohen LG, Hallett M, Geller BD, Hochberg F. Treatment of focal dystonias of the hand with botulinum toxin injections. J Neurol Neurosurg Psychiatry 1989; 52: 355-63.
13. Cole R, Hallett M, Cohen LG. Double-blind trial of botulinum toxin for treatment of focal hand dystonia. Mov Disord 1995: 4, 445-71.
14. Jabusch HC, Zschucke D, Schmidt A, Schuele S, Altenmüller E. Focal dystonia in musicians: treatment strategies and long-term outcome in 144 patients. Mov Disord 2005; 20: 1623-6.
15. Karp BI, Cole RA, Cohen LG, Grill S, Lou JS, Hallett M. Long-term botulinum toxin treatment of focal hand dystonia. Neurology 1994; 44: 70-6.
16. Lungu C, Larp BI, Alter K, Zolbrod R, Hallett M. Long term follow-up of botulinum toxin therapy for focal hand dystonia: outcome at 10 or more years. Mov Disord 2011; 26: 750-3.
17. Ross MH, Charness ME, Sudarsky L, Logigian EL. Treatment of occupational cramp with botulinum toxin: diffusion of toxin to adjacent noninjected muscles. Muscle Nerve 1997; 20: 593-8.
18. Schuele S, Jabusch HC, Lederman RJ, Altenmüller E. Botulinum toxin injections in the treatment of musician's dystonia. Neurology 2005; 64: 341-3.
19. Van Vugt FT, Boullet L, Jabusch HC, Altenmüller E. Musician's dystonia in pianists: long-term evaluation of retraining and other therapies. Parkinsonism Relat Disord 2014; 20: 8-12.
20. Vecchio M, Malaguarnera G, Giordano M, Malaguarnera M, Li Volti G, Galvano F, et al. A Musician's dystonia. Lancet 2012; 379: 2116.
21. Byl N, McKenzie A. Treatment effectiveness for patients with a history of repetitive hand use and focal hand dystonia. J Hand Ther 2000; 13: 289-301.
22. Byl NN, Nagajaran S, McKenzie AL. Effect of sensory discrimination training on structure and function in patients with focal hand dystonia: a case series. Arch Phys Med Rehabil 2003; 84: 1505-14.
23. Byl NN, Archer ES, McKenzie A. Focal hand dystonia: effectiveness of a home program of fitness and learning-based sensorimotor and memory training. J Hand Ther 2009; 22: 183-97.
24. McKenzie AL, Goldman S, Barrango C, Shrime M, Wong T, Byl N. Differences in physical characteristics and response to rehabilitation for patients with hand dystonia: musicians' cramp compared to writers' cramp. J Hand Ther 2009; 22: 172-81.
25. Zeuner KE, Shill HA, Sohn YH, Molloy FM, Thornton BC, Dambrosia JM, et al. Motor training as treatment in focal hand dystonia. Mov Disord 2005; 20: 335-41.
26. Buttkus F, Baur V, Jabusch HC, Paulus W, Nitsche MA, Altenmüller E. Retraining and transcranial direct current stimulation in musician's dystonia –a case report. Mov Disord 2010; 25: 1758-60.
27. Furuya S, Nitsche MA, Paulus W, Altenmüller E. Surmounting retraining limits in musicians' dystonia by transcranial stimulation. Ann Neurol 2014; 75: 700-7.
28. Jabusch HC, Alpers H, Kopiez R, Vauth H, Altenmuller E. The influence of practice on the development of motor skills in pianists: a longitudinal study in a selected motor task. Hum Mov Sci 2009; 28: 74-84.
29. Candia V, Elbert T, Altenmüller E, Rau H, Schäfer T, Taub E. Constraint-induced movement therapy for focal hand dystonia in musicians. Lancet 1999; 353: 42.
30. Candia V, Schäfer T, Taub E, Rau H, Altenmüller E, Rockstroh B, et al. Sensory motor retuning: a behavioral treatment for focal hand dystonia of pianists and guitarists. Arch Phys Med Rehabil 2002; 83: 1342-8.

31. Candia V, Wienbruch C, Elbert T, Rockstroh B, Ray W. Effective behavioral treatment of focal hand dystonia in musicians alters somatosensory cortical organization. Proc Natl Acad Sci U S A 2003; 100: 7942-6.
32. Priori A, Pesenti A, Cappellari A, Scarlato G, Barbieri S. Limb immobilization for the treatment of focal occupational dystonia. Neurology 2001; 57: 405-9.
33. Betti S, Spoto A, Castiello U, Sartori L. Testing rTMS-induced neuroplasticity: a single case study of focal hand dystonia. Neural Plasticity 2018; 2018: 6464896.
34. Rosset-Llobet J, Fàbregas-Molas S, Pascual-Leone A. Effect of transcranial direct current stimulation on neurorehabilitation of task-specific dystonia: a double-blind, randomized clinical trial. Med Probl Perform Art 2015; 30: 178-84.
35. Berque P, Gray H, McFadyen A. A combination of constraint-induced therapy and motor control retraining in the treatment of focal hand dystonia in musicians. A long-term follow-up study. Med Probl Perform Art 2013; 28: 33-46.
36. Rosset J, Fabregas-Molas S. Long-term treatment effects of sensory motor retuning in a pianist with focal dystonia. Med Probl Perform Art 2011; 26: 106-7.
37. Jabusch HC, Schneider U, Altenmüller E. D9-tetrahydrocannabinol improves motor control in a patient with musician's dystonia. Mov Disord 2004; 19: 990-1.
38. Butler K, Sadnicka A, Freeman J, Meppelink AM, Parees I, Marden J, et al. Sensory-motor rehabilitation therapy for task-specific focal hand dystonia: a feasibility study. Hand Therapy 2018; 0: 1-11.
39. Rosenkranz K, Butler K, Williamon A, Rotwell J. Behavioural and neurophysiological effects of proprioceptive training in musician's dystonia. Brain Stimulation 2008; 1: 315. doi 10.1016/j.brs.2008.06.179.
40. Bravi R, Ioannou CI, Minciacchi D, Altenmüller E. Assessment of the effects of Kinesiotaping on musical motor performance in musicians sufferingfrom focal hand dystonia: a pilot study. Clin Rehabil 2019; 1636-48.
41. Pesenti A, Priori A, Scarlato G, Barbieri S. Transient improvement induced by motor fatigue in focal occupational dystonia: the handgrip test. Mov Disord 2001; 16: 1143-7.
42. Horisawa S, Taira T, Goto S, Ochiai T, Nakajima T. Long-term improvement of musician's dystonia after stereotactic ventro-oral thalamotomy. Ann Neurol 2013; 74: 648-54.
43. Byl NN, McKenzie A, Nagarajan SS. Differences in somatosensory hand organization in a healthy flutist and a flutist with focal hand dystonia: a case report. J Hand Ther 2000; 13: 302-9.
44. Yoshie M, Sakai N, Ohtsuki T, Kudo K. Slow-down exercise reverses sensorimotor reorganization in focal hand dystonia: a case study of a pianist. Int J Neurorehabil 2015; 2: 157.
45. Rosenkranz K, Butler K, Williamon A, Cordivari C, Lees AJ, Rothwell JC. Sensorimotor reorganization by proprioceptive training in musician's dystonia and writer's cramp. Neurology 2008; 70: 304-15.
46. Pelosin E, Avanzino L, Marchese R, Stramesi P, Bilanci M, Trompetto C, et al. Kinesiotaping reduces pain and modulates sensory function in patients with focal dystonia: a randomized crossover pilot study. Neurorehabil Neural Repair 2013; 27: 722-31.
47. Oliver S. Musicophilia. Tales of music and the brain. New York: Knop; 2007.

Rehabilitación de la distonía focal de mano en músicos: una revisión sistemática de los estudios

Introducción. La distonía focal de la mano en los músicos es un trastorno del movimiento relacionado con una tarea específica, que se caracteriza por una pérdida involuntaria del control y la coordinación de los movimientos de los dedos al tocar un instrumento.

Materiales y métodos. Se llevaron a cabo búsquedas bibliográficas con las palabras clave *'dystonia'* (distonía) Y ***'musician'*** (músico) Y *'finger'* (dedo) O *'treatment'* (tratamiento) O *'therapy'* (terapia) O *'rehabilitation'* (rehabilitación) en PubMed, EMBASE, Cochrane Library y Web of Science para realizar la revisión sistemática sobre las diversas estrategias usadas para tratar la distonía en los músicos. La búsqueda se realizó de forma independiente por dos autores (R.C. y M.V.) entre el 6 de abril de 2020 y el 6 de junio del mismo año. La investigación identificó un total de 423 artículos. Los encargados de la revisión analizaron 77 artículos que fueron previamente seleccionados. Treinta y seis publicaciones cumplieron con los criterios de inclusión y se incluyeron en la revisión sistemática.

Resultados. La revisión sistemática se realizó para identificar los principales tratamientos utilizados para la distonía en músicos. Se definieron las diversas técnicas existentes para orientar mejor a los médicos a la hora de diseñar un protocolo de rehabilitación que adopte las mejores estrategias descritas en la bibliografía actual.

Conclusión. Esta revisión sistemática intenta proporcionar al lector una mirada completa sobre todos los posibles tratamientos diferentes para la distonía en los músicos. Un protocolo correcto podría permitir mejorar el rendimiento motor y la calidad de vida de los músicos.

Palabras clave. Calidad de vida. Dedos. Distonía. Músicos. Rehabilitación. Revisión sistemática.