**Revised Selected Papers** 

Accademia Musicale Studio Musica Michele Della Ventura, *editor* 

# 2024

# Proceedings of the International Conference on New Music Concepts

Vol. 11



# Accademia Musicale Studio Musica

# International Conference on New Music Concepts

Proceedings Book Vol. 11

Accademia Musicale Studio Musica Michele Della Ventura Editor

Published in Italy First edition: April 2024

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### Musician's Dystonia: new Pevention and Mitigation Treatments

Joy Grifoni<sup>1</sup>, Valeria Crispiatico<sup>2</sup>, Anna Castagna<sup>3</sup>, Rosa Maria Converti<sup>4</sup>, Marina Ramella<sup>5</sup>, Angelo Quartarone<sup>6</sup>, Teresa L'Abbate<sup>7</sup>, Karolina Armonaite<sup>8</sup>, Luca Paulon<sup>9</sup>, Francescaroberta Panuccio<sup>10</sup>, Franca Tecchio<sup>11</sup>

<sup>1, 7, 8</sup> Uninettuno International University, Roma, Italy <sup>1, 7, 8, 9, 11</sup> Laboratory of Electrophysiology for Translational neuroScience LET'S, Institute of Cognitive Sciences and Technologies ISTC, Consiglio Nazionale delle Ricerche CNR, Roma, Italv <sup>2</sup> Milano Bicocca University, Milan, Italy <sup>3, 4, 5</sup> IRCCS Fondazione Don Carlo Gnocchi, Via Capecelatro 66, 20148 Milan, Italy <sup>6</sup> RCCS Centro Neurolesi "Bonino Pulejo", 98124 Messina, Italy. <sup>9</sup> Luca Paulon, Engineer Freelance, 00159 Rome, Italy <sup>10</sup> Department of Human Neurosciences, Sapienza University of Rome, Rome, Italy <sup>1</sup> j.grifoni@students.uninettunouniversity.net <sup>2</sup> valeriacrispiatico@gmail.com <sup>3</sup> acastagna@dongnocchi.it <sup>4</sup>rmconverti@dongnocchi.it <sup>5</sup>mramella@dongnocchi.it <sup>7</sup>t.labbate@students.uninettunouniversity.net <sup>8</sup> karolina.armonaite@uninettunouniversity.net 9 luca.paulon@gmail.com <sup>10</sup> francesca.panucciovg@gmail.com <sup>11</sup> francamatilde.tecchio@cnr.it

Abstract. This prospective article focuses on the most cited scientific evidence regarding the origin and therapeutic treatments for task-specific musician's dystonia (MD), on which new and increasingly effective therapeutic and prevention approaches can be designed for the future. To this end, we have selected data cited at least dozens of times in Scopus, regarding the characteristics and mechanisms underlying MD and the treatments currently available. Pathophysiological mechanisms demonstrate impaired sensorimotor integration at various levels of brain circuits, involving the limbic system and sensorimotor cortices. The alteration of sensorimotor feedback is accompanied by an impairment of intra-cerebral synchrony and long-term memory plasticity. Taskspecific focal dystonia occurs dozens of times more frequently in musicians than in other professions, highlighting that external environmental, social, and emotional factors are crucial dimensions in the origin of MD. This evidence opens the way to the development of multimodal psycho-physical therapeutic strategies to rebalance the alterations of the neural circuits typical of MD through an appropriate personalization of psychotherapy combined with physical rehabilitation activities.

**Keywords**: task-specific focal musician's dystonia (MD), sensory-motor integration, psychic trauma, multi-sensory multimodal rehabilitation.

#### 1 Definition, etiology, epidemiology

#### Definition

Task-specific dystonia is a movement disorder characterized by a painless loss of dexterity specific to a particular motor skill [1]. In musicians, the disorder emerges as painless loss of finger motor coordination or embouchure exclusively when playing the musical instrument [2], giving rise to hand or embouchure MD [3]. MD of the hand affects musicians who play strings and plucked strings such as violinists, cellists, guitarists, pianists and more rarely wind and brass players. MD of the embouchure affects brass and woodwind players, involving the perioral, lingual and facial musculature [4].

#### Aetiology

The causes of MD are still not yet completely clarified, but it is believed to arise from a combination of genetic predisposition and environmental and psychic factors, affecting the brain's motor control system. As for the genetic hypothesis, it is partly supported by evidence that up to 25% of patients with MD have another affected family member with dystonia. Furthermore, a recent genome analysis found an association with the arylsulfatase G gene (ARSG) in both musician's hand dystonia and writer's cramp, but a specific causal mutation within this gene has not yet been identified [3,14]. Possible predisposing risk factors for MD include a positive family history of dystonia, a history of musculoskeletal injury, nerve entrapment or overuse syndrome, and obsessive personality traits [15].

#### Epidemiology

MD is the most common form of focal task-specific dystonia, with a prevalence of 1:100 compared to 1:6600 for idiopathic dystonia [15]. The prevalence of MD varies depending on the instrument played, with musicians playing piano, violin, guitar, and brass instruments being about 85% [16], and embouchure MD accounting for 13-14% of MD [17,18], and cervical dystonia involving 1-2% of musicians with MD.

#### How to measure MD

It is essential to define standard scales to evaluate the severity of MD symptoms, as a basis for diagnosis and monitoring and to quantify the effects of changes induced through therapeutic approaches. As key reference we ground on the work [19], which

developed a wide overview of the rating scales used in MD. The vast majority of studies use subjective-reported or clinician-reported scales, while objective scales started to be introduced (see Table 1).

#### 2 Current treatments

Among approaches tested over the years to address MD, the most common is *botulinum neurotoxin (BoNT) injections* [20–22]. This commonly used treatment is effective for about 12-16 weeks with the effect dose dependent. Cautiousness is indicated, as BoNT is operator dependent, it depends on guides (electromyography/ultrasound), doses, expertise. It is not possible to consider BoNT as a recommended treatment, but some good results are published in fingers flexors dystonia [23]. However, in the case of embouchure dystonia, most authors report a worsening of musical performance with the use of botulinum toxin [24–26]. Furthermore, a recent investigation [27] revealed that, compared to the control group, the thickness and force strength of injected flexor digitorum superficialis (FDS) and profundus (FDP) muscles were decreased by about 10 -12 % with respect to the non-injected body side, with the extent of weakness and atrophy significantly predicted by the total amount of BoNT injected during the entire treatment period, and the time since the last injection not predicting the amount of strength and muscle mass recovery after cessation of treatment.

Non-pharmacological approaches were developed mainly involving various neuromuscular programs [22,28,29]. most studies were applied on a very small sample of musician with MD, with follow-up assessment absent or very short and often based on subjective nominal or ordinal scale; moreover, other study designs didn't provide any type of randomization to placebo or alternative treatments or blindness.

Sensory tricks are known to be able to provide temporary relief of dystonic symptoms [30–32] usually involving the alteration of tactile or proprioceptive feedback. Some musicians experience a marked improvement in fine-motor control playing with a latex glove or holding an object between their fingers, thus modifying the somatosensory input information [18]. Usually, the effects are very rapid in changing the motor pattern, but they last for a limited time.

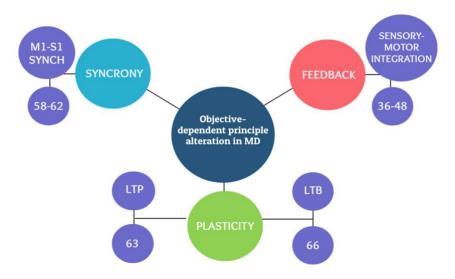
Sensory motor retuning (SMR) treatment has been introduced in a seminal study [9,10,33] introducing the immobilization with splints of one or more non-dystonic fingers. The dystonic finger/s then performed repetitive exercises in coordination with the non-splinted ones. The splint maintains the fingers in their characteristic rest position experienced on the instrument during normal performance.. In these studies, positive effects were seen in terms of dexterity, movement speed and accuracy. Sakai and colleagues [34] developed a motor control retraining technique in MD pianists, named "slow-down exercise" (SDE). During the exercise program, patients undergo

basic movement training at decreased speed, making sure that the dystonic patterns would not occur. The pianists would increase metronome speed every 2 weeks as long as they could maintain a normal movement pattern. Berque and colleagues [35] developed a standardized protocol combining SMR plus SDE intervention over a 12-month period. Butler and colleagues [36] introduced a variation including mirror box (six sessions plus daily exercises). Moreover, Ramella et al. [37] have developed the "modified graded motor imagery" (mMGI) treatment, a 4-weeks home – based comprehensive program designed on the three common steps of the GMI. Ackerman & Altenmuller [38] developed an off-instruments progressive retraining protocol for musicians (MusAARP) following an "add-on" approach, starting from individual intrinsic hand muscle actions to more functional hand and arm actions on the instrument.

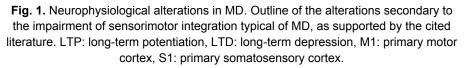
#### 3 MD mechanisms

#### Neurophysiological counterpart

Impairment in sensorimotor integration, crucial in the development of task-specific dystonia, is evidenced by findings from animal studies [39], behavioral research [40], and clinical reports utilizing neurophysiological and neuroimaging techniques in humans [41–50] (Quartarone & Hallett, 2013, Mov. Dis.). A seminal animal study [39] demonstrated that repetitive finger compression led to degradation of hand representation and impaired motor control, while a less repetitive strategy preserved sensory integrity and motor function. This suggests that the repetitiveness of motor patterns influences both somatosensory representation and motor execution. Following the SMR model by Byl and colleagues [36], treatment effects were studied on somatosensory hand representation. Pre-treatment, finger relations differed between affected and unaffected hands, but post-treatment, contralateral finger representations resembled those of the less affected side, and were ordered more according to homuncular principles. These physiological changes correlated with behavioral outcomes [2]. Building on extensive research in non-invasive studies of neural network communication in humans [52–58], we have developed a conceptual framework suggesting that neural networks, comprised of nodes and connectors, are governed by a unified functional principle known as the objective-dependent feedback-synchronyplasticity circuit [59,60]. According to this model, alterations in feedback observed in task-specific dystonia are likely to induce changes in synchronization and plasticity. synchronization, investigations employing transcranial Regarding magnetic stimulation (TMS) protocols revealed the absence of local inhibitory mechanisms during movement initiation and maintenance phases in individuals with task-specific hand dystonia, leading to disrupted cortical inhibition-excitation dynamics [61]. This disturbance affects cortical surround inhibition critical for skill acquisition and relies on inter-hemispheric balances [62,63]. Notably, patients with task-specific hand dystonia exhibit disrupted cortical surround inhibition even during motor preparation [61] (Quartarone et al 2005, Mov. Dis.).



## **NEUROPHYSIOLOGICAL ALTERATIONS IN MD**



Altered excitation and inhibition balances in local motor cortex circuits, reflecting disrupted basal ganglia input, were evidenced by reduced excitability of cortical inhibitory circuits in patients with task-specific dystonia [64]. While no differences were observed in spectral properties between patients and controls in primary motor or somatosensory hand representations, these areas demonstrated reduced functional coupling during movement, along with excessive synchronization in patients compared to controls in the ongoing alpha frequency around 10 Hz [65]. Concerning plasticity, utilizing established non-invasive models of plasticity in humans [66], it has been observed that the motor system exhibits abnormal increases in cortico-spinal excitability and attenuated intracortical inhibitory reinforcement following central associative and peripheral stimulation [67]. Moreover, evidence confirms alterations in long-term potentiation and depression in MD [68–70]. Aligned with the feedback-

synchrony-plasticity model, exemplary cases illustrate the intricate relationship between instrument, action, central nervous system, and periphery in MD [1]. For instance, significant modifications in required actions for an illustrator transitioning to a demanding new job had profound psychological effects, whereas a classical pianist experiencing relevant changes in sensorimotor feedback and slight adjustments in motor command faced comparatively less challenging psychological changes when accepting a new role in a musical.

#### Psycho-social counterpart

While the neurophysiological profile described above is typical of task specific dystonia of the hand, in musicians the occupational dimension decisively modifies the impact of the symptomatology [71] consistent with epidemiological data that indicate MD as the most incident task-specific dystonia [15].

It is immediate to consider that for a musician whose center of professional life is making music, the body is even more immediately at the center of the relationship with the rest of the world. For musicians, health tends to acquire two key dimensions in a dynamic balance, one that concerns the parts of the body directly involved in playing the instrument (especially the hand), and another that focuses on maintaining the health of the whole person [72]. These two dimensions must be integrated in daily attention and care, deepening the awareness that learning through experience, aimed at mutual help and communication, is the key to protect and recover the well-being of the whole body [73].

In the context of the increasingly clear nature of the human being determined within the experience of lifelong learning or trauma (and its profound impact on personal identity), it is possible to speculate that psychological support therapy with EMDR technique (elective for the treatment of trauma) may have particularly encouraging results, especially in its innovation protocol enriched by music and sound stimulation EMDR+ **(**[74, 76] where one of the three main dimensions that index the development of a population is the length of the course of study, the learning approaches for musicians become critical in the training years and all along life. In fact, musicians typically start their training at a young age, with the need to develop movements automated through extensive repetitions, gradually increasing complexity, with the need to balance stereotyped production and emotional participation to the story they like to tell. Given the psycho-emotional pressure intrinsic with the public engagement of a musician's activity, affecting the respiratory, circulatory, sensorimotor, and cognitive systems [77–78], the organization of individual and orchestral training is a critical element determining future evolution managing challenges in adapting to the demands of their profession. Indeed, trauma can have far-reaching consequences, extending beyond psychological distress to impact sensorimotor functionality in

musicians. These repercussions are evident in the disruption of motor coordination, manifested as hesitations, involuntary movements, or complete motor blocks, which impede the expression of artistic intentions.

Furthermore, trauma-induced negations can exert influence on cognitive processes, including memory, attention, and decision-making, exacerbating the challenges faced by musicians during performance. Such cognitive impairments may lead to errors or inconsistencies in musical execution, undermining the quality of their performances. In light of these challenges, addressing trauma and its associated negations is paramount for preserving both physical and mental well-being in musicians. By acknowledging and confronting these negations, musicians can embark on a psychotherapeutic path towards reclaiming agency over their bodies and artistic expression. This process fosters resilience and facilitates a more comprehensive approach to musical performance and self-care for MD.

#### 4 Conclusions

We reported about most stable achievements in MD, where the epidemiology underlines how much professional experience implies the highest occurrence of task specific dystonia in musicians with respect to other workers.

Modern rehabilitation approaches aim to restore a physiological position that supports musicians' natural gestures, mainly via botulinum neurotoxin (BoNT) injections and sensory-motor retuning and its variants, emphasizing body awareness and adjusting positions for relaxation in a dynamical training programming.

The consolidated neurophysiological knowledge on the alteration of sensorimotor integration in MD, which coherently with the *feedback-syncrony-plasticity* functioning principle that governs neuronal networks, is accompanied by altered intra-cortical synchronizations and plasticity, has recently been complemented by the growing awareness of the impact on symptoms of socio-emotional-cognitive experience.

In agreement, new therapeutic strategies are being proposed, which aim to alleviate symptoms that profoundly affect the quality of life of musicians and which often interrupt their career through multidisciplinary approaches with the involvement of interdisciplinary teams including neurology, physiotherapy, occupational therapy, psychology competences integrated into personalized precision approaches.

Author Contributions: Conceptualization, J.G., F.T; methodology, J.G., F.T., V.C.; software, n.a.; formal analysis, n.a.; investigation, n.a.; resources, F.T., L.P.; data curation, n.a.; writing—original draft preparation, J.G., V.C., F.T. A.Q.; writing—review and editing, A.C., RM.C., M.R, T.L. and K.A.; visualization, F.T.; supervision, F.T.; project administration, F.T.,

L.P.; funding acquisition, F.T. All authors have read and agreed to the published version of the manuscript.

#### Declaration of interests: none

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