CHAPTER

A motor control model of task-specific dystonia and its rehabilitation

21

Anna Sadnicka[a,b,*], **Jaume Rosset-Llobet**[c]

[a]*Motor Control and Movement Disorder Group, Institute of Molecular and Clinical Sciences, St George's University of London, London, United Kingdom*

[b]*Clinical and Movement Neurosciences, UCL Queen Square Institute of Neurology, London, United Kingdom*

[c]*Institut de l'Art. Medicina & Fisiologia, Terrassa, (Barcelona) Spain*

**Corresponding author: e-mail address: asadnick@sgul.ac.uk*

Abstract

Task-specific dystonia is a painless deficit of motor control specific to a particular motor skill. In this article we present a motor control model which integrates risk factors for the disorder with the neuroscientific literature of skill learning in health. We particularly focus on the idea that the amount and type of movement variability is critical and show how retraining therapies such as Differential Learning which reintroduces variability into practice can restore motor performance.

Keywords

Task-specific dystonia, Motor control, Model, Rehabilitation, Sensory Motor Retuning, Differential Learning

1 Introduction

Skilled movement represents one of the pinnacles of human development. Professional athletes, dancers and musicians hone their skills through hours of training and there is great beauty in the resulting performance. Worldwide, society has an arts and sports culture embedded in the celebration of such skill. However, the demands placed on the brain and body for such motor excellence are not without risks. In an unfortunate proportion of individuals, a painless deficit of motor control specific to a particular motor skill emerges called task-specific dystonia (Altenmuller, 2008). The most common subtypes are writing dystonia and musicians' dystonia but the disorder can affect the performance of any skill (from the foot of a flamenco dancer through to golfers' yips) (Dhungana and Jankovic, 2013;

Progress in Brain Research, Volume 249, ISSN 0079-6123, https://doi.org/10.1016/bs.pbr.2019.04.011

Garcia-Ruiz et al., 2011). Task-specific dystonia is predictably disabling due to its association with skilled tasks that are required for or which define the individuals' occupation. For example, within groups of professional musicians, the prevalence is approximately 1% and the disorder can mark the end of performing careers (Altenmuller et al., 2014).

For centuries, the pathophysiology of the disorder has remained obscure, with disease models oscillating between the domains of psychiatry and neurology (Lin et al., 2006). Task-specific dystonia is currently considered a subtype of dystonia, a neurological disorder characterized by an abnormality of posture (Albanese et al., 2013). However, traditional dystonic neurophysiological markers such as enhanced plasticity (Sadnicka et al., 2014) or reduced inhibition (Kassavetis et al., 2018) are unable to reliably identify patients with task-specific dystonia (ranges overlapping with controls, similar patterns of abnormalities are observed across a range of neurological disorders). Suggesting that subtle and non-specific shifts in excitability may be epiphenomena removed from the core pathophysiology. Neurophysiological abnormalities are also unable to explain why only an individual task is affected, as abnormalities are documented in circuits subserving unaffected body regions (Quartarone et al., 2008). Disease models built on such foundations such as the disordered sensory homunculus of the affected body part (Elbert et al., 1998) are also difficult to reproduce with updated and more reliable methodology (Ejaz et al., 2016) with inconsistent responses noted with retraining based on such principles (sensory re-education) (Butler et al., 2018).

So what does cause task-specific dystonia? We argue that once freed from traditional dystonia disease frameworks a motor control model can be built from existing clinical and experimental data (Sadnicka et al., 2018). The isolated and task-specific deficit clearly ties the problem to a specific motor skill, as at least at onset, the body region affected can be used normally for other tasks. Task-specific dystonia is therefore best understood by the integration of its clinical features with the neuroscientific literature of skill learning in health. In this article, we discuss the range of risk factors for task-specific dystonia and propose mechanisms by which such factors can translate into motor dysfunction. We particularly focus on the idea that the amount and type of movement variability is critical and show how retraining therapies such as Differential Learning which reintroduces variability into practice can restore motor performance.

2 Risk factors for task-specific dystonia

A range of environmental and genetic risk factors are associated with task-specific dystonia. Environmental risk factors can be subdivided according to the components required to perform any task (Fig. 1A); parameters that define the task and the tool; the central nervous system which includes the network that encodes skill performance, modulated by the individual's psychological state; and the periphery or characteristics of the body region that performs the task (Altenmuller and Jabusch, 2010; Sadnicka et al., 2018). As all components are required to work

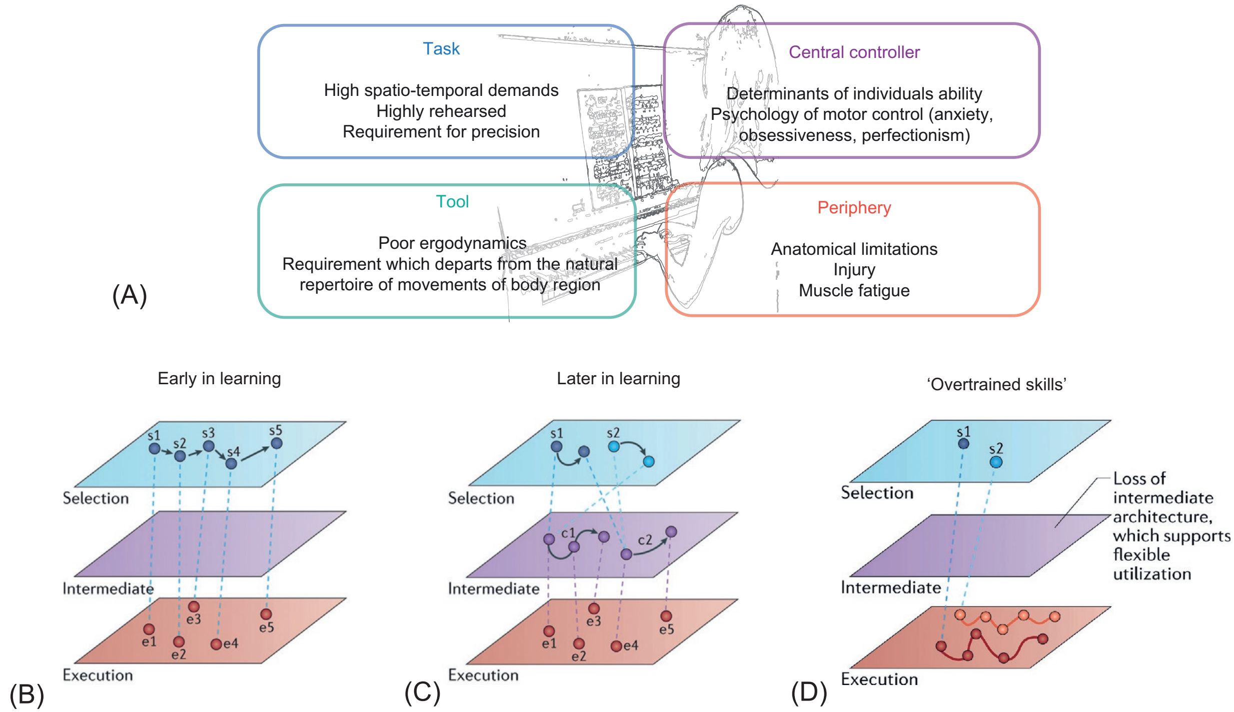

FIG. 1

(A) Risk factors for task-specific dystonia (B) Early skill learning is effortful and involves mapping individual task elements at the selection level (s1-s5) to execution elements (e1-e5) (C) Later in skill learning mechanisms such as chunking are used and chunks (c1, c2) can be flexibly combined in new sequences (s1 versus s2) (D) In "overtrained" skills which are highly optimized the representation at the intermediate level may be lost.

Panels (B)–(D) are taken from Sadnicka, A., Kornysheva, K., Rothwell, J.C., Edwards, M.J., 2018. A unifying motor control framework for task-specific dystonia. Nat. Rev. Neurol. 14, 116–124.

in concert to maintain task performance, a change in one component prompts a change or shift in others, each component a node within a dynamic network required to function in equilibrium for healthy performance. Subdividing risk factors in this manner reveals a risk profile specific to the individual which can be used to tailor management as patients with task-specific dystonia are heterogeneous.

Naturally, the task-specificity of this disorder focuses one on how *task* can confer risk. The highest relative prevalence is found in musicians; 1 in 100 in some studies, but the real figures are likely to be higher as many affected musicians are not diagnosed (Rosset-Llobet et al., 2009a). Within this group, dystonia preferentially affects the hand required to perform with the highest spatiotemporal demand (Altenmuller and Jabusch, 2009; Rosset-Llobet and Fàbregas-Molas, 2013); the right hand in keyboard players and the left hand in bowed instrument players. The increased incidence in classical musicians over jazz musicians also highlights that the invariant temporal and spatial parameters defined on the sheet music of a classical musician affords risk over the inherent flexibility of notes and timing in jazz. Task-specific dystonia typically affects highly rehearsed skills, most frequently presenting in the decade 30–40 (Rozanski et al., 2015). Therefore, dystonia has a predilection for tasks which push spatiotemporal control to its limits, require invariant accuracy and affects tasks which are highly rehearsed.

The influence of *tool* is beautifully exemplified by the shifting prevalence according to the tools used during different historical eras. For example, many telegraph operators developed motor problems of the finger used with the telegraph key which communicated Morse code (Ferguson, 1971; Suzuki et al., 2012). Here the need for individuated, forceful movements departed from the natural repertoire of finger movements with a precise rhythmical definition is salient. As technology advances, writing is less required and a decline or even extinction of writing dystonia is likely. Furthermore, with increasingly ergodynamic tools in the work place the hope is that this leads to a reduction in prevalence in most occupational domains outside of the arts sphere (for example, there are few reports of computer-related dystonia Suzuki et al., 2012).

In order to perform any skill the *periphery* must be capable of a complex set of task requirements and operate within its physiological constraints (Leijnse et al., 2015; Rosset-Llobet et al., 2009b). Correspondingly, peripheral risk factors for task-specific dystonia include inherent limitations (such as the individual's ability to independently move fingers) and acquired peripheral risk factors which temporarily change the operating parameters of the body region (such as local injury or muscle fatigue due to over practice) (Leijnse et al., 2015).

Centrally mediated risk factors encompass both neural skill control and its psychology. For athletes and musicians, the determinants of the "ceiling" of capacity of the nervous system is an important consideration. Those that reach professional levels of competition or performance usually require a combination of both inherent "talent" (such as their capacity for neural plasticity or processing) and intensive and structured nurture through exposure and training. Correspondingly, musicians who start practicing after the age of 10 have an increased incidence of task-specific dystonia, which is after the most sensitive/plastic periods of neural development have occurred (Altenmuller and Jabusch, 2009).

The psychology within which the individual rehearses and performs their motor skill is also critical. Compared to unaffected musicians those with task-specific dystonia are six times more likely to exhibit increased levels of anxiety, perfectionism or stress (Ioannou and Altenmuller, 2014). The influence of acute stress and its interaction over motor control is also apparent. For example, a cartographer for the National Guard had to participate in daily drills in which he had to make a dot on a map to show where bombing practice was to occur (Shamim et al., 2011). Although they were practice drills, they were tense situations and he started having difficulty making the dot, the onset of task-specific dystonia (Shamim et al., 2011).

Finally, genetic factors are also thought to be important and are suggested by the male preponderance and positive family history of movement disorders in a proportion of patients (Lohmann et al., 2014; Schmidt et al., 2006). Exactly how genetic and epigenetic factors contribute to the risk profile of an individual remains to be determined. Any process linked to performance of a skill will be influenced by genetics (ranging from the gating of synaptic plasticity through to the determination of personality traits or musical ability).

3 A motor control model of task-specific dystonia

In this section, we discuss how such epidemiological factors can be mapped to mechanism when profiled and interpreted within the rich neuroscientific literature which studies motor skill learning in health.

3.1 Motor skill learning in health

In order to initiate such a discussion we briefly outline core features of motor skill learning in health. A broad definition of motor skill learning is any neuronal changes that allow an organism to accomplish a motor task better, faster, or more accurately than before (Diedrichsen and Kornysheva, 2015). Skill learning is thought to involve various levels of a motor hierarchy. Within this motor hierarchy one fundamental division is between action selection and action execution (Diedrichsen and Kornysheva, 2015). The execution level causes muscle activity, the neurons that project to the spinal cord and synapse on motor neurones which ultimately cause the peripheral muscles to contract. Within the execution level, movement fragments are thought to be encoded within sub-networks of neurons that code for motor synergies which once activated reliably produce specific spatial-temporal patterns of coordinated muscle activity (Diedrichsen and Kornysheva, 2015). The selection level then activates the most appropriate set of motor synergies in a task-specific manner (dotted lines in Fig. 1B) (Diedrichsen and Kornysheva, 2015). With practice, refinement of reaction time-accuracy trade-offs are considered one of the hallmarks of skill learning (Telgen et al., 2014). This in part is mediated by the formation of intermediate representations which bind together execution elements. For example, experimentally, in addition to sequence completion becoming faster and more accurate, performance starts to show idiosyncratic temporal groups or chunks (Diedrichsen and Kornysheva, 2015). Elementary movements that are bound into one chunk are retrieved faster and more accurately than when the selection level triggers them individually. Such an organization also has the advantage that acquired chunks can be used in the context of novel sequences (Fig. 1C). For example the learning of one sequence (s1) consisting of two chunks (c1, c2) generalizes to the execution of another sequence (s2) which contains the same chunks in a different order (c2, c1) (Sadnicka et al., 2018).

Modular encoding of more abstract features of a task can also endow flexibility to the motor control system. Behaviorally, if an individual is trained on a sequence with temporal (rhythm) and spatial (finger press order) identifiers, a post-training advantage is seen if the temporal features are transferred to a new spatial sequence and vice versa (Kornysheva et al., 2013). Functional MRI data reveals that the temporal and spatial features of the sequence are independently represented in overlapping regions of the pre-motor cortex (Kornysheva and Diedrichsen, 2014). The primary motor cortex in contrast represents the two sequences features (temporal, spatial) in a non-separable fashion (Kornysheva and Diedrichsen, 2014).

Thus as learning progresses intermediate level representations of motor skill features (chunking, modular encoding) are thought to ensure both flexibility and efficiency in motor skill learning (Diedrichsen and Kornysheva, 2015; Sadnicka et al., 2018). Decreasing reliance on the selection level also frees the cognitive system to attend to other related or unrelated tasks whist the motor system is increasingly automatic in its operation.

3.2 Neural correlates of skill expertise

In high risk groups such as professional musicians and athletes the limitations of the neural networks supporting the skill are likely to be an important contributory factor (Sadnicka et al., 2018). Initially, as we have exemplified in healthy skill learning, chunking a sequence offers a behavioral gain in terms of reaction time and is thought to reduce the overall computational complexity associated with learning an entire sequence as a single horizon/sequence (Ramkumar et al., 2016). However, experimental data suggest that repetitive practice of stereotyped movements can lead to the formation of progressively longer motor chunks (Ramkumar et al., 2016). This shift is thought to reflect a trade-off between cost of computation versus efficiency (Ramkumar et al., 2016). Stereotyped practice is thought to iteratively reduce impediments to more complex computation (for example, the relative offset of cost if an individual is required to produce the same movement many times) and correspondingly the chunk structure appears to progressively elongate maximizing the efficiency of the movement (Ramkumar et al., 2016). Such changes are likely to be accentuated in groups whose profession it is to pursue performance perfection and efficiency through practice, an extreme version of the normal mode of operation. Furthermore, as chunks increase in length it is also thought that they are increasingly contextual and tied to the individual task or body region. Informatively, poor transfer of these performance gains to other tasks seems to be accentuated if a narrow training repertoire is applied, in contrast to more varied training approaches (Boutin et al., 2012). Practice predating the development of task-specific dystonia is often particularly intensive and stereotyped.

Therefore, in such highly rehearsed tasks, intermediate-level representations that previously conferred flexibility for related tasks could become redundant. If highly stereotyped sequences begin to dominate the movement repertoire, the original transferrable chunk structure could disappear, as the concatenation into long execution bound mega-synergies effectively replaces such intermediate elements (Fig. 1D). Such an architecture within the motor hierarchy could reliably encode an extreme optimization of performance parameters with little variability across movement repetitions. However, the likely cost of such optimization is that the skill representation retains little capacity for flexibility and generalization to other contexts.

3.3 Psychology of motor control

The impact of psychological influences is worth emphasizing as such factors can have an important effect on skill learning. For example, across animal species in stressful situations a reduction in movement variability and exploration is seen, repetition of movements with rigid movement patterns thought to help one regain a feeling of control (Lang et al., 2015). Experimentally, inducing anxiety in healthy controls during baseline practice undermines later sequence learning and at the behavioral level the mechanism appears to be via a reduction in trial-by-trial variability (Sporn et al., 2018). An unnatural reduction of physiologically informative variability secondary to factors such as anxiety or a personality type which favors rigidity of practice are likely to be detrimental. It is well established that in health subsets of variability are informative for skill learning and are dynamically regulated by the motor control system in response to task requirements (Wu et al., 2014).

Another important psychological factor is the influence of attention. For example, the negative effects of self-focus are commonly discussed within the sports science literature (for example, in relation to "the yips" in golfers) and may be equally relevant in forms of motor impairment that share phenomenology in musicians and writers such as motor block or choking under pressure (Edwards and Rothwell, 2011). Triggering factors for task-specific dystonia such as injury, pain and explicit attempts to alter technique or performance will naturally focus attention on the body region. Misdirected attention can then focus on the mechanics of movement rather than on the external consequences or goals of movement and such a strategy has been shown to worsen skill performance (Lewthwaite and Wulf, 2017). Such mechanisms are paramount when considering the cartographer that struggled to mark a dot on the map (Shamim et al., 2011).

3.4 Onset of dystonia

In many individuals the onset of task-specific dystonia is then commonly precipitated by a triggering event (Rosset-Llobet and Fàbregas-Molas, 2013). Such events include a change in playing technique, injury, a new instrument with a slightly different action/dimension. Conceptually these triggers are best described simply as mismatches between the capacity of the individual's motor control system and the required movement defined by the task and the tool. Some mismatches between capacity and requirement are biomechanical in nature. For example, a task with a high force requirement will limit the capacity to make individuated finger movements leading to greater unintentional and undesired movements of neighboring fingers. Alternatively, changes in capacity due to fatigue or injury of the body can also result in an effector system that responds more erratically to a given motor command. A change in task requirements might also result from external factors, such as changes in the size of a tool or an attempt by the performer to change their instrumental technique. As discussed, in high risk groups, many of their associated risk factors can be interpreted as mechanisms via which the central representation of skill can become particularly narrow in its remit and the required trigger can be a very minor event indeed.

If the individual can accommodate this change in the task requirement by adjusting and scaling its motor commands or finding an alternative combination of neural elements to maintain performance peripherally, an effective compensation has been found. If, however, the new task requirement cannot be accommodated, the performer is pushed outside the boundaries of their learned skill. Task performance then breaks down as no effective compensation is available.

Once a critical mismatch has occurred, novel motor control strategies alien to the existing neural representation of skill must be employed to maintain task performance. However, de novo solutions are unlikely to be able to match or maintain the level of skill performance that was formerly encoded by a hierarchy of neuronal elements optimized over many years of practice (Sadnicka et al., 2018). The skills that are usually affected in task-specific dystonia are characterized by automaticity with little conscious control of movement. By contrast, during de novo learning, task requirements are explicitly mapped to basic execution elements, a time-consuming process that conflicts with the demand for rapid task reproduction within a millisecond timescale. Access to subcomponents of more-abstract movement elements, which previously underpinned some features of expert task performance, is limited. Thus, once task performance has broken down, alternative motor control options are ill-equipped to immediately reinstate motor performance using new elements. Inappropriate and dysfunctional movements are likely to be produced, which are unable to match required task performance levels, marking the onset of task-specific dystonia (Fig. 2A) (Sadnicka et al., 2018).

If dysfunctional or dystonic movements are repeatedly practiced they will become encoded in a similar manner to any other sequence of movements. Conscious access to dystonic movement elements declines, causing frustration for individuals

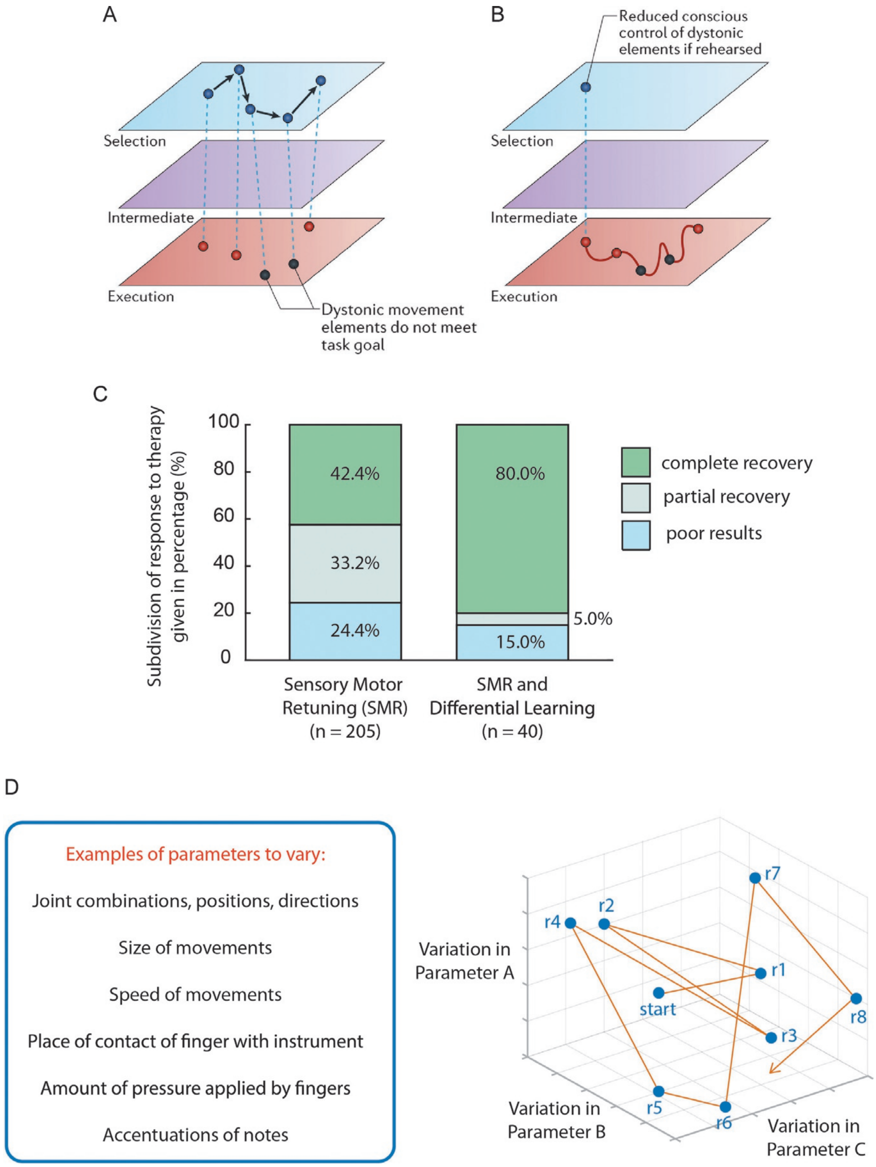

FIG. 2

Neural encoding of task-specific dystonia and its rehabilitation (A) Once a critical mismatch has occurred de novo learning mechanisms are unable to meet task goals. This results in dysfunctional/dystonic movements starting to be encoded within skill network. (B) If dysfunctional/dystonic movements are repeated these will become encoded within lower levels of the hierarchy and there will be less conscious access to dystonic elements. (C) Therapy results before and after adding Differential Learning to Sensory Motor Retuning (SMR). 245 consecutive patients treated at Institut de l'Art, Spain, 2011–2017. Data from a proportion of these patients has been published in preliminary format

(Continued)

with undiagnosed task-specific dystonia as they attempt to implement strategies to address their movement difficulties. Subsequently, skill representations that are activated for a particular context or performance goal will become corrupted, with dystonic movements incorporated into their neural skill network (Fig. 2B) (Sadnicka et al., 2018).

This model for task-specific dystonia is representational and has purposefully resisted providing a direct mapping between the different levels of motor skill learning and specific neural regions as this relationship is likely to be complex. For example, the cerebellar and basal ganglia circuitry form partially parallel loops with multiple cortical regions and may therefore play a role in each of the hierarchical levels of skill learning. Furthermore, emerging treatments for task-specific dystonia such as Differential Learning detailed below target theoretical and behavioral correlates which are thought to involve a broad brain network.

4 Management of task-specific dystonia

4.1 Prevention

One of the important implications of this model is that a proportion of cases of task-specific dystonia could be prevented. As discussed, many occupational forms of task-specific dystonia are characterized by mismatches between the tool or task requirements and the capability of the individual. Improving the ergonomics of tools and limiting task parameters that stress the motor system is likely to be beneficial. However, professional musicians and athletes cannot modify their tool or task requirements to any great extent. Moreover, in response to developing a problem the individual often themselves submits the body to greater and greater demands with the intention of improving aspects of technique or performance. Prevention strategies therefore should focus on modifiable factors; maximizing the "resilience" of relevant representations in the brain, maintaining efficient physiological movements of the periphery, and nurturing a healthy psychological profile.

FIG. 2—Cont'd

(Rosset-Llobet and Fabregas-Molas, 2018). Patients have been classified into three groups depending on their self-reported functional outcome: complete recovery (those who returned to instrumental activity with full recovery, with no dystonia symptoms at all), partial recovery (those with improvement of dystonia symptoms making possible the return to instrumental activity, but still feeling some functional limitation) and poor result (patients who dropped out of treatment or having had certain improvement but not compatible with their musical career). (D) Differential Learning reintroduces noise into training. Parameters are varied in a stochastic manner as shown across repetitions of three theoretical parameters in the three-dimensional scatter and line plot (repetition (r) 1 to 8 demonstrated). Examples of the many parameters that can be varied are given in the box.

Panel (A) is taken from Sadnicka, A., Kornysheva, K., Rothwell, J.C., Edwards, M.J., 2018. A unifying motor control framework for task-specific dystonia. Nat. Rev. Neurol. 14, 116–124.

4.2 Traditional management

Once task-specific dystonia has developed conventional dystonia treatments are often tried. Medications such as trihexyphenidyl have inconsistent effects and are often limited by their side effects (Jabusch et al., 2005; Termsarasab et al., 2016; van Vugt et al., 2014). Botulinum toxin injections, which block the transmission of the nerve to dystonic muscles are often helpful in specialist settings (although some query their long term efficacy) (Kruisdijk et al., 2007; Lungu et al., 2011). However, botulinum toxin injections appear to be acting by suppressing the endpoint of the disease, the inappropriate muscle contractions, rather than addressing the underlying mechanism. Recently a series of patients have received thalamotomy with good outcomes reported (Taira et al., 2006). However, its mechanism of action is entirely unknown, the profile of motor deficits largely uncharacterized and clinical trials are needed before such treatment should be considered mainstream.

Rehabilitation or retraining methods continue to offer a promising treatment option for task-specific dystonia. One of the main form of rehabilitation for task-specific dystonia is Sensory Motor Retuning which was developed by Victor Candia in the 1990s (Candia et al., 1999). Sensory Motor Retuning introduces a novel sensory input by the use of orthotic devices, changing the manner in which the brain perceives the task and facilitating the learning of a healthy performance pattern. For example, splints (to place one or more fingers in a particular position) or rubber bands (that introduce a forced finger flexion or extension in one or more fingers) have commonly been used with the patient performing exercises on the instrument (Rosset-Llobet and Fàbregas-Molas, 2013). Its efficacy is mixed. For example, in a cohort of 205 consecutive patients treated with Sensory Motor Retuning, self-reported functional outcome yields 24% with poor results, 33% with partial recovery with 42% with complete recovery (Fig. 2C). Multifaceted retraining approaches that draw on multiple specific strategies for task-specific dystonia and generic therapy approaches have also been tried (Butler et al., 2018). However when analyzed systemically it remains difficult to clearly identify which elements are the effective ones (Butler et al., 2018; Rosset-Llobet and Fàbregas-Molas, 2013).

4.3 Emerging treatments

The hierarchical motor control model outlined in this paper, invites one to speculate that making a musician's nervous system more flexible could be used in the prevention and treatment of task-specific dystonia. Here, the work of Schöllhorn, a major proponent of Differential Training/Learning, is particularly relevant (Schollhorn et al., 2012). Differential Learning has the primary aim of enlarging fluctuations or stochastic perturbations that occur over movement repetitions in order to provide additional information to the learner (Schollhorn et al., 2012). Variation in normally "invariant" parameters are pursued (for example, the joints used, movement geometry, tool used, and environment Fig. 2D) (Schollhorn et al., 2012).

In task-specific dystonia, if a musician, due to risk factors such as psychological behavior or training workload, shapes a rigid and inflexible representation of their skill, he or she will not easily be able to adapt to internal or external changes. This can lead to dystonia if he or she faces a critical mismatch between an inflexible nervous system and a new task parameter or requirement. Conceptually, Differential Learning therefore offers a targeted manner by which to retrain individuals with task-specific dystonia.

Introducing high doses of variation during retraining is thought to achieve a number of aims. For example, skill networks and neural representations are thought to be "destabilized" facilitating the formation of new functional skill networks (Schollhorn et al., 2010). The use of Differential Learning is also thought to encourage the neural control system to become more flexible and able to accommodate changes the functional parameters of a task (Lungu et al., 2011).

One method of applying this in clinical rehabilitation of task specific dystonia is to perform Sensory Motor Retuning exercises with a change of task parameters every 5 to 10 s (Table 1). The aim is not to look for solutions (a way to play where dystonia symptoms improve or where the patient feels more comfortable) but to introduce

Table 1 Example of a technical exercise on the piano playing C Major scales with both hands over 3 min.

Parameter	Change
Finger joint angle	Metacarpophalangeal joint flexion and interphalangeal complete extension
	Metacarpophalangeal joint extension and moderate interphalangeal flexion
	All joins in extension
	All joins in slight flexion
	All joints in flexion
	Metacarpophalangeal join in extension and maximal interphalangeal join flexion
Wrist position	Neutral position
	Slightly bended
	Moderately extended
	Right wrist slightly bended, left slightly extended
	Right wrist slightly extended, left slightly bended
	Moving from moderate extension to flexion each scale
Finger force	All fingers pressing keys very softly
	Right hand pressing keys very softly and left strongly
	All fingers softly but right and left thumbs
	Right hand softly but index and left hand strongly but middle
	Odd fingers softly and even strongly
	Each three notes change from softly to strongly and vice versa

Each change should be rehearsed for a duration of 10 s.

noise (different ways to play, can be more or less functional than the original one). Movements do not have to make any biomechanical or technical sense and outcomes such as the final sound are also considered unimportant as this will otherwise limit the repertoire of movements that can be incorporated into training, hindering motor system fluctuations and reorganization. Recently, 40 patients have finished a protocol that combined Sensory Motor Retuning with Differential Learning (preliminary data published Rosset-Llobet and Fabregas-Molas, 2018). Notably 80% had complete recovery with only 15% reporting poor results and 5% reporting partial recovery (Fig. 2C).

5 Conclusions

The proposed motor control model has its roots in observations made by patients and clinicians throughout the centuries. We are now additionally aided by a neuroscientific literature which studies skill learning in health which allows one to elaborate on mechanism and infer potential neurobiological correlates. A motor control model for task-specific dystonia is most able to describe its features and provides a conceptual background to emerging rehabilitative therapies. Promisingly for those in the performance arts such as musicians, Differential Learning shows an encouraging ability to facilitate motor system functional reorganization and flexibility, both a possible preventive tool and therapeutic aid.

References

Albanese, A., Bhatia, K., Bressman, S.B., et al., 2013. Phenomenology and classification of dystonia: a consensus update. Mov. Disord. 28, 863–873.

Altenmuller, E., 2008. Neurology of musical performance. Clin. Med. (Lond.) 8, 410–413.

Altenmuller, E., Jabusch, H.C., 2009. Focal hand dystonia in musicians: phenomenology, etiology, and psychological trigger factors. J. Hand Ther. 22, 144–154. quiz 55.

Altenmuller, E., Jabusch, H.C., 2010. Focal dystonia in musicians: phenomenology, pathophysiology, triggering factors, and treatment. Med. Probl. Perform. Art. 25, 3–9.

Altenmuller, E., Ioannou, C.I., Raab, M., Lobinger, B., 2014. Apollo's curse: causes and cures of motor failures in musicians: a proposal for a new classification. Adv. Exp. Med. Biol. 826, 161–178.

Boutin, A., Badets, A., Salesse, R.N., Fries, U., Panzer, S., Blandin, Y., 2012. Practice makes transfer of motor skills imperfect. Psychol. Res. 76, 611–625.

Butler, K., Sadnicka, A., Freeman, J., et al., 2018. Sensory-motor rehabilitation therapy for task-specific focal hand dystonia: a feasibility study. Hand Ther. 23, 53–63.

Candia, V., Elbert, T., Altenmuller, E., Rau, H., Schafer, T., Taub, E., 1999. Constraint-induced movement therapy for focal hand dystonia in musicians. Lancet 353, 42.

Dhungana, S., Jankovic, J., 2013. Yips and other movement disorders in golfers. Mov. Disord. 28, 576–581.

Diedrichsen, J., Kornysheva, K., 2015. Motor skill learning between selection and execution. Trends Cogn. Sci. 19, 227–233.

Edwards, M.J., Rothwell, J.C., 2011. Losing focus: how paying attention can be bad for movement. Mov. Disord. 26, 1969–1970.

Ejaz, N., Sadnicka, A., Wiestler, T., Butler, K., Edwards, M., Diedrichsen, J., 2016. Finger representations in sensorimotor cortex are not disrupted in musician's dystonia. In: Society for Neuroscience Annual Meeting 2016. https://doi.org/10.1038/srep37632.

Elbert, T., Candia, V., Altenmuller, E., et al., 1998. Alteration of digital representations in somatosensory cortex in focal hand dystonia. Neuroreport 9, 3571–3575.

Ferguson, D., 1971. An Australian study of telegraphists' cramp. Br. J. Ind. Med. 28, 280–285.

Garcia-Ruiz, P.J., del Val, J., Losada, M., Campos, J.M., 2011. Task-specific dystonia of the lower limb in a flamenco dancer. Parkinsonism Relat. Disord. 17, 221–222.

Ioannou, C.I., Altenmuller, E., 2014. Psychological characteristics in musicians dystonia: a new diagnostic classification. Neuropsychologia 61, 80–88.

Jabusch, H.C., Zschucke, D., Schmidt, A., Schuele, S., Altenmuller, E., 2005. Focal dystonia in musicians: treatment strategies and long-term outcome in 144 patients. Mov. Disord. 20, 1623–1626.

Kassavetis, P., Sadnicka, A., Saifee, T.A., et al., 2018. Reappraising the role of motor surround inhibition in dystonia. J. Neurol. Sci. 390, 178–183.

Kornysheva, K., Diedrichsen, J., 2014. Human premotor areas parse sequences into their spatial and temporal features. Elife 3, e03043.

Kornysheva, K., Sierk, A., Diedrichsen, J., 2013. Interaction of temporal and ordinal representations in movement sequences. J. Neurophysiol. 109, 1416–1424.

Kruisdijk, J.J., Koelman, J.H., Ongerboer de Visser, B.W., de Haan, R.J., Speelman, J.D., 2007. Botulinum toxin for writer's cramp: a randomised, placebo-controlled trial and 1-year follow-up. J. Neurol. Neurosurg. Psychiatry 78, 264–270.

Lang, M., Kratky, J., Shaver, J.H., Jerotijevic, D., Xygalatas, D., 2015. Effects of anxiety on spontaneous ritualized behavior. Curr. Biol. 25, 1892–1897.

Leijnse, J.N., Hallett, M., Sonneveld, G.J., 2015. A multifactorial conceptual model of peripheral neuromusculoskeletal predisposing factors in task-specific focal hand dystonia in musicians: etiologic and therapeutic implications. Biol. Cybern. 109, 109–123.

Lewthwaite, R., Wulf, G., 2017. Optimizing motivation and attention for motor performance and learning. Curr. Opin. Psychol. 16, 38–42.

Lin, T., Shamim, E., Hallett, M., 2006. Focal hand dystonia. Pract. Neurol. 6, 278–287.

Lohmann, K., Schmidt, A., Schillert, A., et al., 2014. Genome-wide association study in musician's dystonia: a risk variant at the arylsulfatase G locus? Mov. Disord. 29, 921–927.

Lungu, C., Karp, B.I., Alter, K., Zolbrod, R., Hallett, M., 2011. Long-term follow-up of botulinum toxin therapy for focal hand dystonia: outcome at 10 years or more. Mov. Disord. 26, 750–753.

Quartarone, A., Morgante, F., Sant'angelo, A., et al., 2008. Abnormal plasticity of sensorimotor circuits extends beyond the affected body part in focal dystonia. J. Neurol. Neurosurg. Psychiatry 79, 985–990.

Ramkumar, P., Acuna, D.E., Berniker, M., Grafton, S.T., Turner, R.S., Kording, K.P., 2016. Chunking as the result of an efficiency computation trade-off. Nat. Commun. 7, 12176.

Rosset-Llobet, J., Fàbregas-Molas, S., 2013. La Dystonie du Musicien. Alexitere Editions, Montauvan.

Rosset-Llobet, J., Fabregas-Molas, S., 2018. Chapter 51: Rehabilitation and Plasticity of Task-Specific Focal Hand Dystonia, first ed. Cambridge University Press.

Rosset-Llobet, J., Candia, V., Fabregas i Molas, S., Dolors Rosines i Cubells, D., Pascual-Leone, A., 2009a. The challenge of diagnosing focal hand dystonia in musicians. Eur. J. Neurol. 16, 864–869.

Rosset-Llobet, J., Garcia-Elias, M., Montero, J., Valls-Sole, J., Pascual-Leone, A., 2009b. Linburg's syndrome, can it cause focal dystonia? Mov. Disord. 24, 1704–1706.

Rozanski, V.E., Rehfuess, E., Botzel, K., Nowak, D., 2015. Task-specific dystonia in professional musicians. A systematic review of the importance of intensive playing as a risk factor. Dtsch. Arztebl. Int. 112, 871–877.

Sadnicka, A., Hamada, M., Bhatia, K.P., Rothwell, J.C., Edwards, M.J., 2014. A reflection on plasticity research in writing dystonia. Mov. Disord. 29, 980–987.

Sadnicka, A., Kornysheva, K., Rothwell, J.C., Edwards, M.J., 2018. A unifying motor control framework for task-specific dystonia. Nat. Rev. Neurol. 14, 116–124.

Schmidt, A., Jabusch, H.C., Altenmuller, E., et al., 2006. Dominantly transmitted focal dystonia in families of patients with musician's cramp. Neurology 67, 691–693.

Schollhorn, W.I., Beckmann, H., Davids, K., 2010. Exploiting system fluctuations. Differential training in physical prevention and rehabilitation programs for health and exercise. Medicina (Kaunas) 46, 365–373.

Schollhorn, W., Hegen, P., Davids, K., 2012. The non linear nature of learning—a differential learning approach. Open Sports Sci. J. 5, 100–112.

Shamim, E.A., Chu, J., Scheider, L.H., Savitt, J., Jinnah, H.A., Hallett, M., 2011. Extreme task specificity in writer's cramp. Mov. Disord. 26, 2107–2109.

Sporn, S., Hein, T., Herrojo, R.M., 2018. Bursts and variability of beta oscillations mediate the effect of anxiety on motor exploration and motor learning. bioRxiv, https://doi.org/10.1101/442772.

Suzuki, K., Takano, M., Hashimoto, K., et al., 2012. Computer mouse-related dystonia: a novel presentation of task-specific dystonia. J. Neurol. 259, 2221–2222.

Taira, T., Ochiai, T., Goto, S., Hori, T., 2006. Multimodal neurosurgical strategies for the management of dystonias. Acta Neurochir. Suppl. 99, 29–31.

Telgen, S., Parvin, D., Diedrichsen, J., 2014. Mirror reversal and visual rotation are learned and consolidated via separate mechanisms: recalibrating or learning de novo? J. Neurosci. 34, 13768–13779.

Termsarasab, P., Thammongkolchai, T., Frucht, S.J., 2016. Medical treatment of dystonia. J. Clin. Mov. Disord. 3, 19.

van Vugt, F.T., Boullet, L., Jabusch, H.C., Altenmuller, E., 2014. Musician's dystonia in pianists: long-term evaluation of retraining and other therapies. Parkinsonism Relat. Disord. 20, 8–12.

Wu, H.G., Miyamoto, Y.R., Gonzalez Castro, L.N., Olveczky, B.P., Smith, M.A., 2014. Temporal structure of motor variability is dynamically regulated and predicts motor learning ability. Nat. Neurosci. 17, 312–321.