HHS Public Access

Author manuscript

*Biol Cybern*. Author manuscript; available in PMC 2020 June 17.

Published in final edited form as:

*Biol Cybern*. 2015 February ; 109(1): 109–123. doi:10.1007/s00422-014-0631-5.

# A multifactorial conceptual model of peripheral neuromusculoskeletal predisposing factors in task-specific focal hand dystonia in musicians: etiologic and therapeutic implications

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## Abstract

A model is presented showing how peripheral factors may cause a process of movement adaptation that leads to task-specific focal hand dystonia in musicians (FHDM). To acquire a playing technique, the hand must find effective and physiologically sustainable movements within a complex set of functional demands and anatomic, ergonomic, and physiological constraints. In doing so, individually discriminating constraints may become effective, such as limited anatomic independence of finger muscles/tendons, limited joint ranges of motion, or (subclinical) neuromusculoskeletal defects. These factors may, depending on the instrument-specific playing requirements, compromise or exclude functional playing movements. The controller (i.e., the brain) then needs to develop alternative motions to execute the task, which is called *compensation*. We hypothesize that, if this compensation process does not converge to physiologically sustainable muscle activation patterns that satisfy all constraints, compensation could increase indefinitely under the pressure of practice. Dystonic symptoms would become manifest when overcompensation occurs, resulting in motor patterns that fail in proper task execution. The model presented in this paper only concerns the compensatory processes preceding such overcompensations and does not aim to explain the nature of the dystonic motions themselves. While the model considers normal learning processes in the development of compensations, neurological predispositions could facilitate developing overcompensations or further abnormal motor programs. The model predicts that if peripheral factors are involved, FHDM symptoms would be preceded by long-term gradual changes in playing movements, which could be validated by prospective studies. Furthermore, the model implies that treatment success might be enhanced

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by addressing the conflict between peripheral factors and playing tasks before decompensating/retraining the affected movements.

**Keywords**

Hand; Biomechanics; Control; Focal dystonia; Overuse; Musicians

## 1 Introduction

Task-specific focal hand dystonia in musicians (FHDM) is a painless, playing-specific hand/finger control problem that develops at a mean age of 33 but sometimes more than 10 years earlier. The function of the hand otherwise is generally normal. FHDM affects, depending on the instrument, approximately 1% of professional musicians, with a strong instrument–hand correlation and a significantly greater male incidence (up to 80%); see Table 1. The causes of FHDM are still unknown, no current treatment is predictably effective, and FHDM generally ends a performing career [1–16].

Musicians' hand dystonia is symptomatically grossly similar to focal hand dystonias (FHDs) with other repetitive activities, such as writer's cramp. Together these are often called *occupational cramps*. Sometimes patients have other focal dystonias as well, such as blepharospasm and cervical dystonia. However, this is quite atypical in FHDM. Associations have been investigated with central and peripheral neurological pathology. Peripheral nerve pathology in FHDM [17] was not systematic, although more present with certain affected fingers [7,18]. Epidemiological studies of focal dystonias suggest an autosomal dominant influence with reduced penetrance [19–22]. In more than 10% of cases, possibly up to 30%, FHDM musicians have a family history of FHDs or other focal dystonias [5,23,24].

A number of physiological abnormalities have been identified in patients with focal dystonias in general, with, however, some caution as to their presence in the subpopulation of FHDM. A fundamental abnormality seems to be decreased inhibition, which has been seen in spinal and brainstem reflexes, H reflex, blink reflex recovery in blepharospasm, and short and long intracortical inhibition [25–35]. There are also mild abnormalities of sensory function in FHDs, which might be due to a decreased inhibition in sensory processing [11,25,36–39]. Altered sensory representations can lead to abnormal motor behavior, and this has been mathematically modeled [40,41]. A third abnormality is enhanced sensorimotor cortex plasticity, including enhanced plasticity in some paradigms and a loss of homeostatic plasticity [5,25,42–49]. Interestingly, some of these phenomena are also found in nondystonic musicians [34,50,51], pointing to physiological changes in nondystonic musicians that are not seen in nonmusicians. Moreover, changes also take place in brain morphology in musicians, presumably based on their long hours of training [52].

In conclusion of the preceding discussion, predictive causative central nervous system (CNS) mechanisms in FHDM have not yet been clarified. Moreover, it is likely that environmental factors are also crucial since none of the occupational cramps happen without some repetitive behavior. Therefore, we present a model of how peripheral factors might help provoke FHDM; such factors are likely to be more important in FHDM than in other

FHDs because of the precise control needed and the much-increased repetitive behavior. The model generalizes evidence that enhanced muscle forces and finger trajectory limitations or trajectory interference due to finger muscle independence limitations, joint motion limitations, or neuromusculoskeletal defects may be causes of playing problems [53–61] that could predispose to or trigger FHDM [7,62–65].

The model's scope is limited to the peripheral circumstances and the peripherally driven processes that would precede the onset of symptoms. The model does not aim to explain what exactly happens at the onset of symptoms or the nature of dystonic motions themselves; in view of the paucity of current knowledge, this would be speculative. Rather, the model aims to determine what can be said with some certainty about a system of which, at present, very little is actually understood. The peripheral factors in the model can in principle be measured, and the processes considered involve normal movement learning processes, so no assumptions about pathological CNS processes need be made a priori. Therefore, many basic model assumptions can be validated in normal musicians, as discussed further.

The model has therapeutic implications in suggesting that treatment success might be enhanced by addressing the conflicts between individual peripheral constraints and tasks before considering treatment strategies such as movement decompensation and neurological retraining.

## 2 Peripheral factor FHDM model

### 2.1 Model parameters

*Individually normally feasible movements* Many musculoskeletal factors may differentiate individual hand function relative to instrumental playing requirements: hand/arm/finger dimensions, congenital limitations or lack of limitations (laxity) on joint ranges of motion (RoM), lack of finger independence due to interconnected finger tendons or insufficiently differentiated finger muscles, and others [53–56,61,62,66–78] (Fig. 1, block 1. Notation: the blocks in Fig. 1 will be further noted by their number only). There may also be clinical factors, for example, musculoskeletal defects such as ligament damage or peripheral nerve damage (Fig. 1, 6). These factors determine in each hand a set of *normally feasible movements* (NFMs) (Figs. 1 and 2). NFMs are defined as movements that can be performed by normal forces and motor activation patterns. While this definition is qualitative because the term *normal movement* may not be strictly definable; it expresses that certain factors or factor combinations may allow certain movements only by forcing. Even, constraints such as joint RoM limitations or intertendinous connections [57,58] may completely exclude certain movements. The definition includes the effects of factors such as peripheral nerve compressions that may diminish precise control or strength of certain muscles.

*Required instrumental playing movements (RM)* To be played, an instrument requires the realization of all trajectories of its keys or impressions of strings at appropriate speeds with appropriate forces to overcome the key or string resistances and inertia forces (Fig. 1, 3 and 4).

*Occupational physiological load* General occupational factors (e.g., playing load, number of concerts), individual performance factors (e.g., playing technique, practice methods, repertoire), and exceptional factors (e.g., extra concert practice) result in an individual occupational physiological load (Fig. 1, 5a and b).

*Individual neurogenetic FHDM predispositions* are included as an unknown quantity (black box) that may range from total absence to severe (Fig. 1, 8).

## 2.2 Fundamental test: are the required playing motions part of the normally feasible motions in view of the physiological load?

A feasible instrumental playing technique requires that the RM can be realized by the hands at the level of the occupational physiological load. In the model, this feasibility is broken up into subtests (Fig. 1, 10 and 20), which are explained using the following scenarios in which, for the sake of argument, neurogenetic predispositions are not considered.

**2.2.1 Unconstrained hand—**When RM is a subset of NFM (Fig. 1, 10), the hand can meet all playing requirements by normal forces and motor patterns. By the model hypothesis, such a hand will have no playing problems and may develop a playing technique without FHDM risk. This hand then remains for life in a *safe state* event loop (Fig. 1, 11–13), unless by factor changes RM ceases to be a subset of NFM and the hand reenters the model at Fig. 1, 0.

**2.2.2 Constrained hand—**When RM is not a complete subset of NFM (Fig. 1, 10), a hand–instrument interface manipulation incompatibility exists, resulting in instrument-technical playing problems with those RMs that are not in NFM (Fig. 1, 14). These incompatibilities do not necessarily exclude acquiring a playing technique, but they necessitate the use of greater muscle forces and/or alternative trajectories involving more muscles and joints, which will further be called *compensation* (Fig. 1, 15).

**2.2.3 Compensation: effectiveness and physiological feasibility—**Compensation is defined as the recruitment of function from other joints and muscles or any significant motor activity (force, timing) other than would normally be used in carrying out the playing task. This definition remains intuitive since *normal* playing movements are generally not well defined. Nevertheless, comparative quantification may establish greater forces in more muscles or (phasic) actions of muscles controlling more proximal joints to enhance the trajectories of more distal segments. Examples are wrist or pronation–supination movements supporting finger actions. These compensations result from motor pattern adaptations that by the model hypothesis in first order are driven by normal movement optimization and learning behaviour (Fig. 1, 16). To be stable, compensations must satisfy two conditions, which are at the core of the FHDM model, together with test 10 of Fig. 1.

1. *Effectiveness* The RM must be realized, if not perfectly, to the satisfaction of the player (Fig. 1, 17).
2. *Physiological feasibility* Even when effective, compensated movements will only be stable if the effort to perform them is within the muscles' short- and long-term

physiological capacities (Fig. 1, 20). Physiological feasibility means in this model not primarily that the movement can be physically executed (this is implied, as otherwise the movement in test 17 of Fig. 1 cannot be performed), but rather that it is compatible with the body's movement optimization feedback loops. For example, a movement that leads to chronic exhaustion of certain muscles may remain physically feasible at all times but may yet be rejected (modified) in the long term as the body seeks to minimize the movement's cost.

**2.2.4 Nondystonic overuse—**Physiological overload may cause inflammatory overuse conditions, such as tendonitis (Fig. 1, 21). This is generally clinically evident and treated as such. Only when loads remain subinflammatory – and therefore generally painless – do individuals carry on in the model.

**2.2.5 Overconstrained hand (Fig. 1, 22, loop A)—**When no effective compensations exist, which may happen when too many RMs require compensations that may even be mutually incompatible or negatively interact, the subject will not be able to achieve an acceptable playing technique and will eventually quit playing (Fig. 1, 18).

**2.2.6 Feasibly constrained hand—**By hypothesis, effective and physiologically feasible compensations are safe for FHDM (Fig. 1, 20, exit a). The hand will then use these compensated movements for life, unless factors change sufficiently to bring the hand back into the model at Fig. 1, 0. However, the more compensated the playing technique, the more vulnerable the hand will be to factor changes, possibly to the point where mere occupational changes (trigger events) suffice for muscle actions exceeding physiological feasibility, at which point the hand continues in the model at Fig. 1, 20, exit b (loop B).

**2.2.7 Hands at risk of FHDM: open loop for movement compensation—**When, at subinflammatory levels, effective compensations in the long term exceed physiological limits (Fig. 1, 20, exit b), subconscious muscle feedback will induce the controller (brain) to change muscle motor patterns (Fig. 1, 16) in an attempt to redistribute muscle load so that the physiological limitations can be satisfied. This means that compensations will increase (Fig. 1, 15). These changed motor patterns and ensuing playing motions must again pass the effectiveness and physiological feasibility tests (Fig. 1, 17 and 20). Metablock 22 in Fig. 1 thus allows for open positive feedback loops of increasing compensations, which occur when (i) no compensations are effective but the subject persists in practicing (Fig. 1, 22, loop A) or (ii) when effective compensations never satisfy the physiological feasibility condition (Fig. 1, 22, loop B). The likelihood of (ii) should be far greater than (i) because subjects who do not reach an acceptable level of performance will likely quit playing. Loops A and B also include voluntary movement changes (e.g., by teaching), as the effectivity and feasibility tests (Fig. 1, 17 and 20) only pertain to the resulting movements and not the causes that produced them.

**2.2.8 Occurrence of FHDM symptoms (Fig. 1, 19 and 23)—**The positive feedback loop B of compensation allows for the possibility of movement control collapse due to overcompensation, resulting in dystonic symptoms. Such control problems would arise when, because of a lack of motor activation patterns producing movements that satisfy both

effectivity or physiological feasibility, the controller (brain) ultimately develops motor patterns that are too complex to perform accurately in real time, or that corecruit antagonists to the intended movements, resulting in cramping or dyscontrol, or that even involve complex multifinger task rearrangements, leading to finger function overload, as was, by hypothesis, the case in [63]. Then the movements become ineffective (test 17 in Fig. 1 fails).

**2.2.9 Dystonic versus ineffective movements—**The effectivity test (Fig. 1, 17) can fail for at least two reasons.

1. An instrument-technical playing problem, i.e., the inability to accurately execute a series of notes at the required playing speed because of anatomical/biomechanical constraints.
2. Movement dyscontrol, i.e., the inability to properly control the fingers or body part in playing a series of notes because of ineffective motor patterns.

These two cases must be distinguished; therefore the model contains a *diagnostic test* (Fig. 1, 19). This test, however, has an irreducibly ambiguous discriminatory range because especially in the early stages of onset of symptoms an instrument-technical playing problem (with possible associated fatigue in repetitive execution) and a finger control problem may not be clearly distinguishable.

**2.2.10 Central nervous system factors—**In the model, FHDM symptoms can result from dysfunctional motor patterns acquired by normal movement learning processes (Fig. 1, 16), which would imply that persons without any CNS predispositions might develop FHDM. Two CNS factors, possibly but not necessarily fully associated, are envisioned that might enhance the instability of compensatory motor pattern development, with sliding scales symbolizing an individually variable degree of presence: (1) neurogenetic predispositions (Fig. 1, 8) and (2) abnormal cortical adaptability (plasticity) (Fig. 1, 16). In terms of neurogenetic predispositions, any factors that reduce fine motor control resolution will be aggravating, in the same way as, for example, peripheral nerve compressions will be aggravating factors.

**2.2.11 Model limitations and predictions—**The model hypothesizes a generic mechanism of how normal motor learning processes can cause FHDM development in a favorable factorial environment. In this, the model horizon is limited to the point where dysfunctional motor patterns, i.e., symptoms, develop (test 19 in Fig. 1). The model does not speculate on what happens thereafter in the CNS with further practice, nor does it aim to account for specific motor learning processes or mechanisms if predisposing CNS factors were present. However, the following statements would be consistent with the model hypothesis: the more task-specific the symptoms, the less likely that significant neurogenetic predispositions exist and, therapeutically, the more likely that appropriate factorial changes, if these are possible, would allow reversing the symptoms.

**2.2.12 Trigger events (Fig. 1, 5b)—**Trigger events preceding FHDM symptoms are frequently reported. Such events include increased practice of a difficult repertoire for a concert, a new instrument with (slightly) different action/dimensions, changes in playing

technique (new teacher), or minor trauma [79]. By hypothesis, trigger events have in common that they acutely increase physiological load by increasing occupational activity or hand-interface incompatibilities. The increased physiological muscle load would accelerate replacement of borderline feasible movements by overcompensated dysfunctional motor patterns. Note that instrument-technical playing problems (Fig. 1, 14) by themselves are also triggers to the degree that they invite predisposing behavior by becoming the object of excessive practice.

**2.2.13 FHDM development dynamics: musicians' aggravating responses—**In the model, FHDM symptoms should start gradually because in the movement compensation loop, movements may balance on the borderline between functional and slightly dysfunctional motor patterns, modulated by (periods of) greater or lesser physiological playing load. However, at slight initial symptoms of dyscontrol, musicians tend to practice the problematic movements excessively, assuming that practice would improve control. In doing so, by hypothesis, they acutely induce further muscle exhaustion and drive dysfunctional motor pattern development, aggravating symptoms in a short period of time. Once the motor patterns are dysfunctional, further practicing these movements will likely worsen matters, even after rest. Learning requires proportionality between inputs (muscle activations) and intended outputs (functional movements). This proportionality is disrupted when dysfunctional movements are produced, meaning that control cannot likely be relearned by practicing dystonic movements. Rather, in the attempt to regain control in the dysfunctional input–output relationship, the controller will likely involve more muscles with increased motor activations, increasing symptoms.

**2.2.14 Peripheral clinical control problems—**Clinical factors such as nerve compressions or ligament damage may lead to control problems by themselves. When severe, such factors affect other activities besides playing and will likely be clinically diagnosed (Fig. 1, 6, 7, and 26). Subclinical factors can be considered as individual hand factors (Fig. 1, 1) and may cause dysfunctional overcompensations by the same processes as in the compensation loops (Fig. 1, 22). Nerve lesions in FHDM would also fall into this category [7,18,80]. The effects of a suspected neuromuscular defect were analyzed in [63].

**2.2.15 Specific cases of factors—**In neurological studies, FHDM cases are generally treated as a homogenous group. However, according to the proposed model, symptoms should relate to compensations for specific individual factors, which should make it possible to discriminate individual symptomatology. As an example, in what follows, two classes of factors are juxtaposed: antagonistic couplings and underconstrained hands.

*Antagonistic couplings* in multifinger systems result from interconnections between tendons to different fingers or insufficiently individuated finger muscles. Through these connections or common forces a muscle in one finger may pull tendons of other fingers with it. If the ensuing movements in the coupled fingers are unwanted, antagonists of the pulled tendons must be activated to suppress these motions. Muscles from different fingers may thus antagonistically load each other as a function of the required movements. These mutual muscle loads remain internal and cannot be appraised from the external forces [65], although the increased muscle activity should be reflected in EMG. Moreover, tendinous couplings

may become taut (active) or slack (inactive) during finger trajectories, perturbing the intended trajectories. In playing, it might then be necessary to compensate (correct) for these perturbations, resulting in temporally highly complex motor activation patterns or generally increased muscle activations in anticipation of such disturbances, even if this merely results in increased mutual antagonistic multifinger muscle loading. Such internal muscle force and activation timing adaptations should also be interpreted as compensation, even if the ensuing trajectories would not appreciably change by the mutually increased muscle forces. With antagonist couplings, the symptomatic dystonic motions would most likely be the faster motions because the execution of highly complex compensations would pose more problems in fast than in slow motions.

*Underconstrained hands* At the other end of the factor spectrum are underconstrained systems, in which defects introduce degrees of freedom (DoFs) that are not fully covered by the available muscles. As stated earlier, gross defects will also affect daily functions and will be clinically detected, but small defects may go unnoticed while still introducing instabilities in the joint chain. Muscles then obtain dual functions: (1) to generate task-accomplishing motion and (2) to provide additional compensatory chain stability by controlling the malcontrolled DoFs. This might lead to overstimulation of muscles (cramping) driven by control feedback if the instability cannot be adequately compensated and to conflicts in task priorities because the objective function of the task is no longer unique. Playing problems may then result because the stability requirement takes precedence over task execution. Such a case was analyzed in [63], where it was hypothesized that because stability compensations became dominant, the hand no longer met certain playing targets. In that case, slow motions were dystonic, while the fast playing motions were unaffected, even after 7 years of symptoms.

## 2.3 FHDM treatment in the model

**2.3.1 Diagnostics of peripheral factors—**The model hypothesizes that peripheral factors leading to hand–task conflicts or to diminished movement control (in the case of subclinical defects) would underlie FHDM. Diagnostics in the model should therefore include checking all possible (hand) factors in Fig. 1, 1, such as anatomical constraints on joint RoM, finger independence, hand/finger size, and others; as well as clinical factors (Fig. 1, 6), such as nerve compressions, ligament damage, or joint laxity that might require enhanced muscle activity for joint stabilization, old scars that might increase stiffness, smooth tendon gliding or limit tendon excursions, and any other factor that might compromise peripheral function (Fig. 1, 27). In this, the effects of adverse hand factors are cumulative: each additional factor might further constrain normally feasible movements and increase physiological load in movement execution.

**2.3.2 Treatment—**If (severe) factors causing hand-task conflicts can be identified, treatment would consist of two steps.

1. Elimination of sufficient factors to reduce the hand– task conflict to the degree that effective and physiologically feasible movements for the underlying instrument-technical playing difficulties would likely become possible (Fig. 1, 28, exit a).

2. Decompensating/retraining of normal muscle synergies (Fig. 1, 29), after which the hand reenters the model at Fig. 1, 0.

Apart from resolving possible clinical defects, the model provides three therapeutic parameter categories to diminish the hand–task conflict:

*Occupational load* changes in, for example, performance demands (repertoire), practice intensity, and instrumental technique (Fig. 1, 5).

*Ergonomic adaptations* of the instrument interface to the hand limitations (Fig. 1, 3).

*Adapting the hand to the instrument interface* by modifying hand factors, including surgical removal of hand constraints [64,78,81](Fig.1, 1). Note that this parameter analysis only considers the possibilities of principle, irrespective of the risks involved in surgical procedures on the musician's hand.

**2.3.3 Treatment prognosis—**The model predicts that without changing *any* model parameters, movement retraining of whatever nature would not likely be successful because at full playing load the hand would simply reenter the unsafe compensation loops, likely with the same outcome, i.e., recurrence of symptoms. In borderline cases, changes in occupational factors (Fig. 1, 5: repertoire, workload, including playing technique) or ergonomic factors (instrument interface) might suffice. Feasible solutions might exist that have not been discovered by the controller (brain). To learn these movements – if they are identified – with the hand in a dystonic state would require an exceedingly specialized and controlled practice environment. With severe hand–task incompatibilities, conservative factor changes might not sufficiently resolve the underlying hand–task conflicts, meaning that no retraining therapy might be successful. In selected cases, elimination of hand-interface incompatibilities by surgical removal of anatomical constraints might reduce or eliminate the necessity of compensations in the first place. In that case, further therapy would consist of learning a new playing technique based on the expanded set of normally feasible movements.

In the presence of CNS neurogenetic factors, treated in the model as black boxes but possibly evidenced by less task-specific or focally progressive symptoms, and while it would not be excluded that peripheral factors could also be aggravating, the outcome prognosis would be worse.

## 2.4 Complementary model views

**2.4.1 Practice-invariant playing movements—**A complementary model view is provided by considering playing technique development with practice over time (Fig. 2). When starting to learn playing an instrument, great movement changes occur with practice. However, with further practice, these changes decrease. In the fully practiced hand, movements will be repeated with great accuracy, becoming entities that can be analyzed. The model predicts the following. (i) In an unconstrained hand, playing movements become practice-invariant (Fig. 2, curve 1). At a certain point, they will not fundamentally change anymore with further practice unless (hand) factors change (Fig. 1, 11–13). (ii) In a constrained hand with effective and physiologically feasible movement compensations,

acquiring a playing technique requires more practice. This is because the necessary compensations, which may be highly context sensitive (i.e., dependent on previous and subsequently playing fingers), must be developed and optimized. However, these compensated playing movements also eventually become practice-invariant, unless factors sufficiently change (Fig. 2, curve 2). (iii) In a hand at risk of FHDM, playing movements not satisfying physiological feasibility (Fig. 1, 20) do not become practice-invariant. Muscle motor patterns and ensuing movements keep changing – gradually – with practice over time (months, years) as they cycle the positive feedback loop of compensation (Fig. 1, 22, loop B). However, when viewed within short time periods, these movements are still stably reproducible entities because they are, at any time, well practiced. With insufficient exposure to practice, the compensated movements may never reach a dystonic state (Fig. 2, curve 3). With sufficient practice, a dystonic state may ultimately be reached, at which point further practice will strongly accelerate movement changes (Fig. 2, curve 4). Trigger events (Fig. 1, 5b) may hasten, or even provoke the process because it remains possible that without the trigger event and the usually ensuing excessive practice at onset of symptoms a dystonic state would not be reached (Fig. 2, curve 5). Physiological capacities may increase with practice (Fig. 2, shifting line 6 up), but they could also decrease (shifting line 6 down) with too much practice, age, nerve compressions, systemic conditions, or other clinical events (Fig. 1, 6). Such factors may turn safe movements into unsafe movements, even when the practice load remains constant (e.g., one FHDM patient in Table 1 was in the initial stages of multiple sclerosis). Anecdotal evidence of practice variance in FHDM is regularly presented by patients bringing videos in which gradual playing movement changes can be observed over years before the onset of symptoms.

**2.4.2 Movement control feedback loops—**The main movement feedback loops relevant to acquiring a playing technique are sketched in Fig. 3. At any level in these loops, pathologies may produce more or less generalized control problems. Forward control stops with muscle activations. Tactile feedback extends up to the interface contact. Auditory feedback in musicians is of obvious task-performance relevance, but visual feedback of posture and playing trajectories is also crucial in playing technique development and its teaching. Musculoskeletal factors or defects are beyond voluntary control. The controller can only mediate conflicts between feasible and required movements by creating muscle activations to adequately manipulate the body’s joints. If effective and physiologically feasible movements cannot be realized, the brain keeps modifying muscle activation patterns, while system feedback from chronic muscle exhaustion, intrinsic control problems due to defects, or others keeps rejecting these over time. Instrument mechanics, fed back through required finger trajectories and key or string resistance forces, may also increase physiological hand load, e.g., in badly maintained instruments. Changes of instrument, such as a new piano with different (heavier, deeper) key action, have been reported as FHDM trigger events (Fig. 1, 5b). Visual feedback may play an aggravating role when playing trajectories or postures are imposed by the teacher or the musician himself that suppress necessary functional compensations. Auditory feedback also has pathogenic potential, e.g., when leading to excessive practice to play problematic note series right.

# 3 Discussion

## 3.1 Peripheral and neurological factors in FHDM: model limitations

It should be reemphasized that the model only describes processes *before* symptom development. The model makes no suppositions as to what happens in the CNS at the moment of symptom development or thereafter. In the model, FHDM arises as the end stage of a positive feedback loop of compensation for peripheral constraints or defects relative to the task. The inherent instability of a positive feedback loop implies that in the model, FHDM might develop in neurologically normal subjects. However, human movement control functions from a biological substrate that may individually enhance movement control/learning instability. FHDM may thus result from an individually variable weighting of neurogenetic factors and normal physiological learning mechanisms and movement adaptions under peripheral pressure, possibly leading to maladaptive cortical changes. This would agree with partially positive genetic evidence – although peripheral factors may also be genetic and should be verified; neurological case–control differences as may be present in nonaffected body parts; and enhanced plasticity, which, however, was also enhanced in asymptomatic musicians [34,50]. Sanger and Merzenich [41] proposed that FHD might result from motor control loop gain increases due to increases in sensorimotor cortical mapping of muscles in movements, which might occur by normal plasticity processes with predisposing peripheral factors, such as fused muscles or couplings between movement antagonists. Such factors were also considered as predisposing on biomechanical considerations [65].

The model incorporates CNS predispositions as *black boxes* because exact predisposing CNS mechanisms have not yet been clarified and because CNS predispositions, whatever their nature, would not diminish the core message that in the individual playing hand peripherally driven positive feedback loops for movement compensation may exist.

## 3.2 Experimental evidence supporting the model

A sizeable body of evidence exists supporting basic model propositions.

*Individually discriminating hand factors (*Fig. 1, 1*)* Individual anatomic finger muscle independence differences, as well as many other individual hand factors, including clinical factors, have been amply documented (see Sect. 2.1).

*Instrumental playing difficulties caused by anatomical constraints (*Fig. 1, 10 and 14*)* Anatomic limitations as causes of playing problems were theoretically analyzed [53,55,57,58,64,65], experimentally established [82], and demonstrated by instrument-technical playing improvements after surgical release (e.g., [78,81]).

*Case–control studies of predisposing peripheral factors* Wilson et al. [62] reported a weak correlation of FHDM with metacarpophalangeal joint abduction limitations. Note that these are only some of the many factors that can cause a hand to fail test 10 of Fig. 1.

*Epidemiological evidence* of peripheral factor involvement in FHDM is strong and includes, among other evidence, the correlation between affected hand and instrument type (Table 1),

including the remarkably high FHDM prevalence in classical guitarists' right hands (see subsequent discussion), and trigger events involving instrument ergonomics (new instrument with different action) or minor trauma.

*Task specificity of symptoms* Analysis of individual symptoms provides strong evidence that at least in some cases CNS predispositions must be slight. Some FHDM musicians, for example pianists, can play quite well while avoiding the use of one or more fingers, while the hand becomes immediately dysfunctional if the excluded fingers are also used. It is difficult to imagine a CNS predisposition for a single finger, but biomechanically disturbing motor couplings that become active with one finger while remaining inactive with disuse of this finger can be readily imagined. [63] presented a 7 year history of dystonic slow motions, while fast motions remained unaffected and extremely accurately controlled. Clearly, in this case CNS predispositions were unlikely, as was the notion that repetitive motions by themselves were causal, since fast motions are repeated orders of magnitude more frequently than slow motions.

### 3.3 Experimental feasibility of model validation

The model relates measurable quantities and therefore can be validated. Hand factors can be measured and biomechanically modeled. Instrument-technical playing problems can be quantified, for example, by reproducible irregularities in note series executions. Compensations can be detected by trajectory analysis, EMG patterns, and hand-interface contact forces. Therefore, the fundamental relationship between hand factors, playing problems and compensations can be validated, which would validate the model up to Fig. 1, 15. Note that this relationship must be established in unsymptomatic musician controls because in FHDM, patients' previous instrumental playing problems can no longer be relevantly quantified because of the dysfunction. However, in patients, hand factors *can* be measured and correlated with hand factors that cause playing problems in controls. This would make it possible to establish that FHDM patients would have technical playing problems prior to symptoms. The entire model could be validated by a prospective study of a large group of musicians, preferably of instruments with high FHDM incidence, in which hand factors and playing technique would be quantified at the start of, and periodically during, a professional instrumental study and thereafter, so that in those that become dystonic, the full performance history would be available. Clearly, in studies of such scope, neurogenetic factors in the subjects should likewise be investigated.

### 3.4 FHDM characteristics consistent with the model hypothesis

**3.4.1 Task specificity—**In the model, dystonic symptoms result from dysfunctional motor patterns, not from dysfunctional muscles or their elemental controllers. Therefore, when controlled by normal motor patterns in other tasks, the same muscles would function normally. Control would likely be more stable to the degree that movements differ from affected movements. Some degree of flow-over of affected FHDM movements to similar movements in other activities has been reported [83].

**3.4.2 Painlessness—**FHDM symptoms result from acquired motor patterns, not inflammatory overuse or acute (painful) muscle fatigue, even if chronic muscle exhaustion

drove FHDM development. The model does not exclude the possibility that FHDM and inflammatory overuse interact. Pain-induced compensations may stimulate development of overcompensations. Conversely, workload reduction caused by inflammatory overuse might in some cases prevent FHDM by diminishing the occupational exposure (Fig. 1, 5).

**3.4.3 Late onset—**The optimization and rejection of effective but physiologically unsustainable motor patterns would require intensive practice for a long period of time (metablock 22, Fig. 1). Not mutually exclusive, with CNS predispositions, the compensatory processes of repetitive activity with abnormal biomechanics might take time to produce the plastic motor cortex changes that could lead to maladaptive change. In Leijnse and Hallett [63], FHDM occurred only 2years after a traumatic hand muscle defect, suggesting that with clinical peripheral control problems less time may be required to develop symptoms.

**3.4.4 Brain plasticity—**Two plasticity aspects may be distinguished: (1) facility in forming synaptic connections and (2) overlapping (smearing) of sensorimotor cortical finger presentations. Concerning the second aspect, it may be observed that the cortex can only map what peripheral control can differentiate. A lack of anatomic finger muscle/tendon individuation will be reflected in overlapping sensorimotor cortex functions at many levels: in primary common motor mappings, in motor patterns with common motors active in control of different fingers, and by muscle coactivations correcting for unwanted force transfers from connected motors to other fingers in specific movements [65]. With highly connected finger muscles, seemingly simple single finger movements may thus require exceedingly complex multifinger muscle activations. Finger movement maps may therefore be highly complex and overloaded for purely peripheral anatomical/mechanical reasons. Overlapping motor cortex areas in FHDM may thus reflect (over)compensatory muscle synergies superimposed on a substrate of already poorly diversified primary motor cortex maps. Superimposed on these complex but normal movement control cortex reflections may be maladaptive changes.

**3.4.5 Trigger events—**Trigger events were discussed in sect. 2.2.12

**3.4.6 FHDM prevalence per hand and per instrument type—**Gross instrument statistics provide strong evidence of causal peripheral factor involvement beyond mere repetitive activity. According to the model, even with equal constraints, left and right hands may have significantly different instrument-dependent physiological playing loads and therefore different susceptibilities to FHDM. For example, in keyboard players, the more active right hand is affected in 90% of cases and in bowed string instruments the left hand is more affected (Table 1). This is consistent with the model, but also with a causal hypothesis based on mere exposure. However, the statistics of other instruments violate the mere exposure pattern. For instance, the number of classical guitarist FHDM cases, almost exclusively in the right hand, equals that of all keyboard instruments combined (Table 1). However, there are far fewer classical guitarists than keyboard players.[1] Therefore, the guitarists' FHDM prevalence should be several times that of keyboardists and is likely the

[1]For instance, as an indicator, we found the ratios of piano teachers to classical guitar teachers in six major schools of music worldwide ranging between 5:1 and 12:1.

highest of any instrument, possibly 3 or 4%, meaning it can no longer be considered a rare occupational disorder. Predisposing CNS factors cannot explain this high prevalence because it is unlikely that predisposed subjects would select classical guitar with such a bias. Nor can repetitive finger motion by itself explain the prevalence because the pianists' right hand in the advanced repertoire plays far more notes per time. Nor is it likely that guitarists have a statistically higher exposure to practice than pianists: piano is one of the most competitive instruments, as attested by the abundance of piano competitions worldwide, which has no comparable equivalent for the guitar.

Another odd FHDM prevalence is with the flute, with a 5:1 left-to-right-hand FHDM ratio. Mere exposure cannot explain this pattern either since it is unlikely that the left and right hands play such a different ratio of notes. Hypotheses for peripheral factors in the FHDM prevalence in guitar and flute are discussed further.

**3.4.7 FHDM in musicians with excellent playing technique—**The model prediction that hand-interface manipulation conflicts underlie FHDM may seem in contradiction with musicians who, while having apparently excellent playing technique, acquire FHDM. However, even virtuoso hands are not necessarily free of adverse factors. The effectiveness condition (Fig. 1, 17) merely requires the ability to keep the effects of hand factors within acceptable auditory perception limits. While hand factors will likely be less prominent in virtuosos, lesser factors may also cause physiologically predisposing playing loads violating the physiological feasibility condition because of greater performance demands (Fig. 1, 20).

**3.4.8 Why more musicians do not get FHDM—**Anatomical constraints on finger independence are systematically present in the population, though they are highly individually variable. Therefore, a key question is why only some musicians get FHDM. While this certainly requires more study, an important reason may be that musicians generally eventually adapt their repertoire to their technical possibilities, e.g., many predominantly teach. Conversely, but not mutually exclusive, neurological predispositions maynot be that common, but, again, neither exposure nor predispositions by themselves explain the odd prevalence ratios in, for example, guitarists and flutists.

**3.4.9 Playing technique—**While many movement learning processes are subconscious, there is a large voluntary component in instrumental playing, which allows for teaching a playing technique. The risk to FHDM may increase when movements are imposed in conflict with hand constraints, enhancing physiological load in the physiological feasibility test (Fig. 1, 20). Conversely, teaching playing techniques that are optimally tailored to individual hand factors might decrease FHDM risk. Important is the realization that a universally optimal playing technique cannot exist because each hand will have a different optimum depending on its individual constraints.

**3.4.10 Why rest generally does not help—**Rest may benefit physiological factors such as chronic muscle fatigue, but it does not change hand–task conflicts nor acquired dysfunctional motor patterns. Task-specific load will therefore likely reproduce prerest dystonic motor patterns at some point. However, in mild FHDM cases, some recuperative

effect of rest does exist, with symptoms returning only after practicing the provoking movements for some time. By hypothesis, after partial recovery by rest, predystonic movements are executable until underlying muscle fatigue drives the system back to dysfunctional synergies. In severe cases, dysfunctional motor patterns completely replace predystonic motor patterns, so that no normal playing movements can be performed even after rest.

## 3.5 Analysis of FHDM prevalence in specific instruments

### 3.5.1 Model hypothesis of the high left–right FHDM prevalence ratio in flutists—

To explain the 5:1 left–right FHDM prevalence ratio in flutists, two differentiating factors are proposed here.

1. *Different left–right finger postures differently offset anatomical constraints* Tendinous connections between finger tendons are generally least stretched (i.e., active as limitations on finger independence) with extended fingers. This correlates with the fact that the basic hand anatomy is embryologically laid down in a hand plate where fingers have equal positions. This neutral position is more or less the right-hand flute position. In contrast, the left-hand position is with the index finger strongly flexed and the middle finger flexed more than on the right hand. This statistically offsets possible anatomical constraints against the neutral anatomical position, enhancing the likelihood of a manipulation conflict.
2. *The left thumb plays keys and the right thumb does not*, which means that left-hand manipulation is inherently more complex than right-hand manipulation. For example, the left-hand fingertip forces must be otherwise balanced when the thumb is lifted and its force opposing the other fingertip forces becomes zero.

For these reasons, a flutist's left hand is, in terms of constraints and control, more vulnerable to hand-interface manipulation conflicts, which is consistent with its higher FHDM prevalence. This does not mean that the right hand could not become dystonic instead of the left hand. The individual variability in possible factors may be such that the factors in the right hand outweigh the factors in the left hand, even against the biomechanically predisposing left-hand tasks.

### 3.5.2 Model hypothesis of high FHDM prevalence in guitarists' right hand—

As stated earlier, a guitarist's right-hand FHDM prevalence is likely higher than that of any other instrument. By the model hypothesis, this is because the classical guitarist's right hand presents a worst-case scenario predisposing it, anatomically and biomechanically, to FHDM.

1. *Absence of contact forces* Other instruments have keys or fingerboards that stop keystroke motion. The resulting contact forces constitute additional variables that become integrated into the playing technique and that can be used in compensations. With the help of contact forces and the ensuing frictional fixation of the fingertips, motions in nonpressing fingers may become possible that otherwise could not be realized. In guitar, the string in string plucking provides no equivalent set of contact force variables. Finger motions are predominantly ballistic, string stroke forces are small and motions light and, therefore,

vulnerable to perturbations from interdigital anatomical couplings. All effects of such couplings must be resolved by muscle activations of fast, free-moving fingers without the help of contact forces.

2. *Compulsory use of deep flexors* String plucking requires active use of the deep flexors, which are anatomically the most interconnected of the finger flexors. Moreover, with no fingerboard to stop the fingerstroke, finger flexion depth is deeper than with other instruments. Both factors enhance the likelihood of adverse effects of interdigital anatomical couplings, especially in the deep flexors, in playing.

3. *String layout limits the compensatory solution space* Guitar strings are not widely spaced and require rather precise trajectories for individual string plucking. These trajectory demands fix the hand position and limit the solution space for movement compensations that could avoid or diminish the adverse effects of interdigital couplings.

In conclusion, by the model hypothesis, the accumulation of these adverse factors should make a guitarist's right hand significantly more vulnerable to FHDM than other instruments.

### 3.6 Current FHDM treatments, outcome unpredictability

Many FHDM treatment strategies have been proposed, for example, symptom reduction by botulinum toxin, movement and sensory retraining to increase sensorimotor cortical hand representation resolution, and others [84,85]. Outcomes have been unpredictable and full playing return rare. Clinically, outcome unpredictability is a major problem because any FHDM treatment represents a considerable time/effort investment for both patient and therapist, excluding other treatments possibly more appropriate for the given case.

The common basis of retraining strategies is limiting movements to subsets that avoid provoking symptoms, in the hope that the motor patterns thus obtained could be expanded to encompass all playing motions. The model allows that such procedures make it possible to develop asymptomatic motor patterns for the therapeutic movement subset because this subset likely satisfies conditions 10, 17, and 20 of Fig. 1. However, the model also predicts that, independently of the treatment strategy, a return to presymptomatic occupational levels without changes in any factors in Fig. 1, 1, 3, 5, and 6 would have very high FHDM recidivism. Indeed, the hand would simply reenter the compensation feedback loop in similar circumstances with a likely similar dystonic outcome. This suggests that reported treatment successes would have included permanent changes in – usually occupational – factors: playing load, playing technique, repertoire, overpractice avoidance (trigger events), and others. Reporting all occupational changes together with the treatment successes would thus be relevant. Occupational or instrument-ergonomic changes may not suffice to sufficiently resolve hand–task conflicts. In selected cases, surgical release of anatomical constraints may achieve this [64,78,81] but carries risks not incurred in conservative treatments and should be the subject of a comprehensive risk–benefit analysis, including long-term follow-up. When hand–task conflicts cannot be sufficiently resolved, according to the model, no permanent success could likely be obtained, regardless of treatment strategy. Analysis of individual hand–task conflicts would allow tailored treatments and systematic

development of clinical experience, with the prospect of improved prognostics and outcomes. Note that the present model makes no statements concerning optimal decompensation/retraining strategies themselves but merely considers the boundary conditions within which such strategies could be successful.

## 4 Conclusion: future FHDM research

A model has been presented showing how peripheral hand/ instrument manipulation conflicts with sufficient repetitive physiological load could be important or necessary factors in FHDM development. To validate this model and to harmonize it with neurogenetic findings and neuromotor control models such as [41], peripheral and neurogenetic factors should be *jointly* investigated in case–control, prospective, and treatment-outcome studies.

## Acknowledgments

The authors thank J. E. Bonte, MD, J. C. Van der Meulen, MD, PhD, J. M. F. Landsmeer, MD, PhD, C. W. Spoor, PhD, J. C. Snijders, PhD, A. B. M. Rietveld, MD, A. Gupta, MD, N. Campbell-Kyureghyan, PhD, P. M. Quesada, PhD, W. P. Hnat, PhD, G. Prater, PhD, P. Mathys, PhD, A. Delchambre, PhD, and F. Schuind, MD, PhD, for their support. This paper is part of the study ‘Anatomical variations predisposing to focal dystonia in the musicians’ hand’, supported by the Dystonia Medical Research Foundation, Chicago, IL (http://www.dystonia-foundation.org), the University of Louisville (IRIG-MRG grant), Louisville, KY, USA, and the Université Libre de Bruxelles, Brussels, Belgium. Dr. Hallett is supported by the NINDS Intramural Program.

## Abbreviations

| | |
|---|---|
| **FHD** | Focal hand dystonia (not specific to musicians) |
| **FHDM** | Focal hand dystonia in musicians |
| **NFM** | Normally feasible movement |
| **MC** | Movement compensation |
| **RM** | Required instrumental movement |
| **RoM** | Range of motion |
| **DoF** | Degree of freedom |

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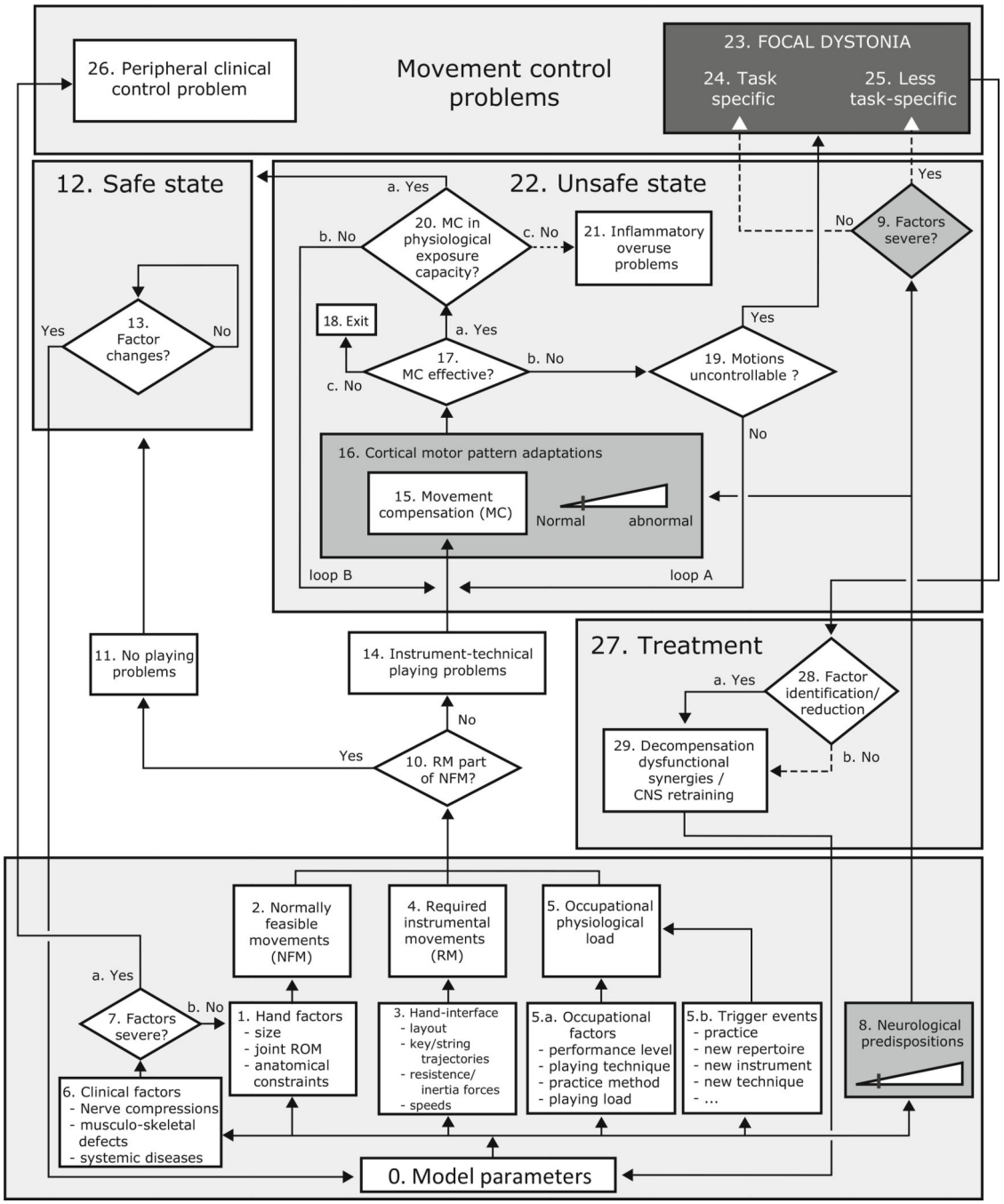


**Fig. 1.**

Multifactorial model of peripheral factors in focal hand dystonia in musicians. For explanation, see text. RoM (block 1): range of motion

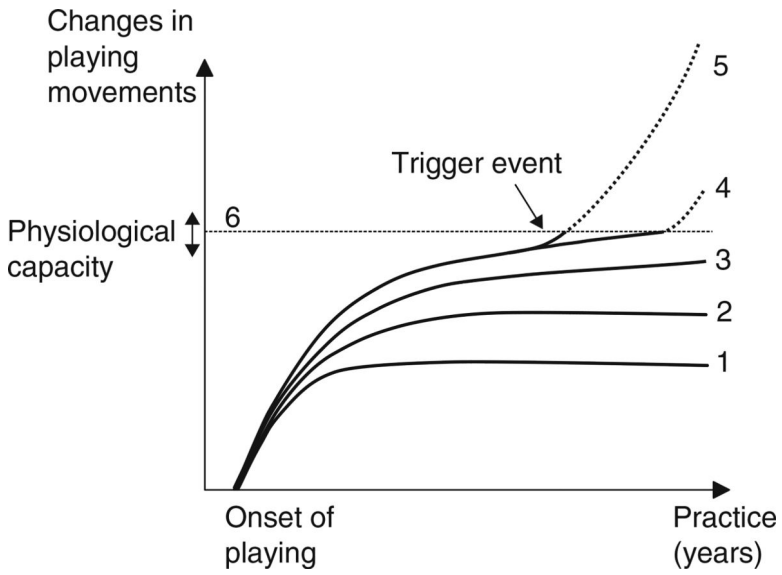


**Fig. 2.**

Playing technique changes with practice over time–hypothesis. (*1*) Unconstrained hand: playing movements become practice-invariant. (*2*) Constrained hand with effective and physiologically feasible compensated movements: longer learning process, but playing movements also become practice-invariant. (*3–5*) Hands at FHDM risk. (*3*) Playing technique does not become practice-invariant, but exposure (amount of practice) remains too small for FHDM development. (*4*) Greater practice intensity leads to FHDM, with resulting run-away changes in movements.(*5*) Trigger event provoking FHDM development. (*6*) Physiological muscle capacities may increase/decrease over time, increasing or reducing the critical physiological load for FHDM development

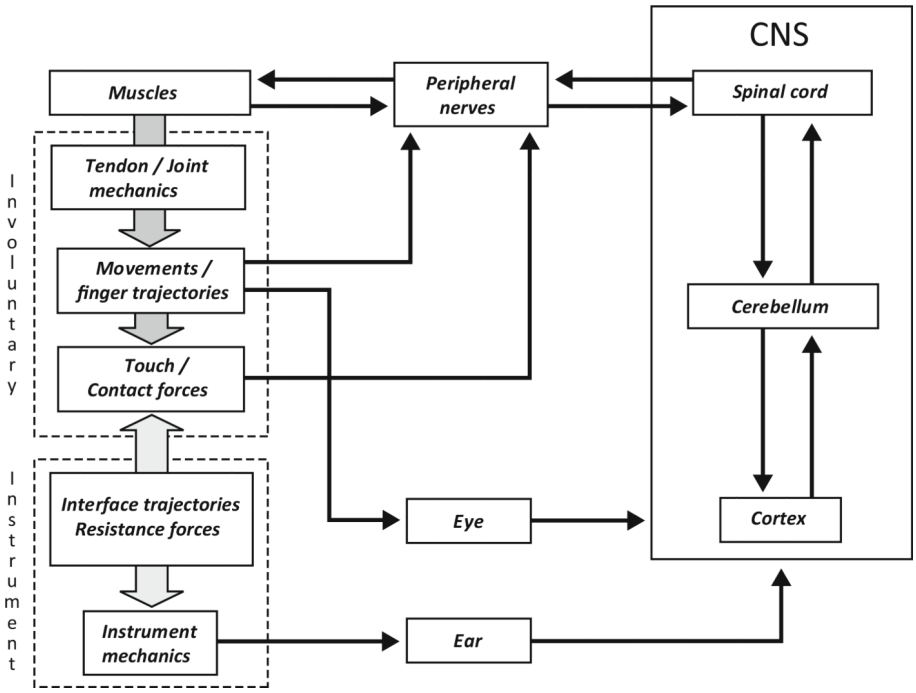


**Fig. 3.**

Schematized are the control and overarching feedback loops that determine the development of a playing technique. Musculoskeletal hand factors or defects, musculoskeletal mechanics, and instrument manipulation requirements are beyond voluntary control (*dashed boxes*). The limit of control is muscle activation. The brain's sole means of solving conflicts between factors in the dashed box is the production of motor patterns leading to effective movements. Depending on the factors, effective and physiologically feasible motor patterns to solve the instrument-technical playing problems may or may not exist. Damage to peripheral nerves (compressions) may diminish the responsiveness of muscles to fine control and thus make the hand more vulnerable to developing inadequate motor patterns. Voluntary components in developing playing motions relate to auditory (task achievement) and visual (playing posture) feedback

**Table 1**

Instrument type, affected hand, and gender of FHDM patients of Hand Clinic for Musicians, Rotterdam, The Netherlands, 1991–2000

| Instrument type | Instrument | L | R | % R | Male | Total |
|---|---|---|---|---|---|---|
| Keyboard | Piano, organ, clavichord, MIDI keyboard, accordion | 2 | 17 | 88 | 12 | 19 |
| String | Guitar | 2 | 18 | 90 | 20 | 20 |
| | Violin | 2 | 0 | 0 | 1 | 2 |
| | Cello | 2 | 0 | | 0 | 2 |
| Wind | Flute | 5 | 1 | 17 | 3 | 6 |
| | Sax | 1 | 0 | − | 1 | 1 |
| Total | | 14 | 36 | 72 | 74% | 50 |

Hand surgeon: G. J. Sonneveld, MD. Biomechanics consultant, research and development: J. N. Leijnse, PhD. These distributions are similar to larger series. For example, the ratio of 5:1 left hand prevalence in flutists is maintained in larger series