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Review article

The curse of motor expertise: Use-dependent focal dystonia as a manifestation of maladaptive changes in body representation

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ABSTRACT

Focal task-specific dystonia (FTSD) impairs not only motor dexterity, but also somatosensory perception involved in well-trained behavioral tasks. Occupations that carry a risk of developing FTSD include musician, writer, painter, surgeon, and golfer, which are characterized by repetitive and precise motor actions over a prolonged period. Behavioral studies have uncovered various undesirable effects of FTSD on sensorimotor functions, such as a loss of independent movement control, unintended muscular co-activation, awkward limb posture, and impairment of fine discrimination of tactile and proprioceptive sensations. Studies using neuroimaging and noninvasive brain stimulation techniques have related such sensorimotor malfunctions to maladaptive neuroplastic changes in the sensorimotor system, including the primary motor and somatosensory areas, premotor area, cerebellum, and basal ganglia. In this review, we summarize recent empirical findings regarding phenomenological and pathophysiological abnormalities associated with the development of FTSD. We particularly focused on maladaptive alterations of body representations underlying the degradation of fine motor control and somatosensory perception in FTSD patients.

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"Practice makes perfect" is a common principle in many cultures. Indeed, the development of expertise is associated with deliberate and intensive practice, often initiated at an early age (Krampe and Ericsson, 1996). In support of this concept, neurophysiological evidence has demonstrated that extensive training can lead to neuroplastic adaptations in the cortical and subcortical regions subserving sensory, motor, and cognitive skills (Jancke, 2009; Münte et al., 2002; Zatorre et al., 2007). Unfortunately, excessive practice, in combination with innate biological vulnerability, can yield maladaptive changes in certain neural networks, leading to compromised motor dexterity (Furuya and Altenmüller, 2015). A representative example is focal task-specific dystonia (FTSD), a sporadic movement disorder that can interfere with movements involved in highly-trained tasks such as writing, playing an instrument, tool use, and sports. FTSD often

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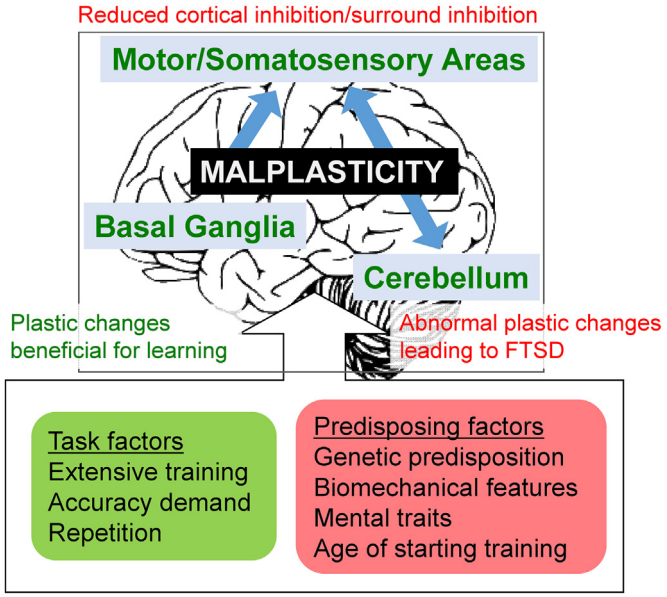

Fig. 1. Hypothetical model of the pathogenesis of FTSD. An interaction between task factors (e.g., extensive training, accuracy demand, and repetitiveness) and predisposing factors (e.g., genetic predisposition, biomechanical features, mental traits, and age of training onset) triggers maladaptive changes in cortical regions (e.g., motor, somatosensory, premotor), subcortical regions (e.g., basal ganglia and cerebellum), and their connections, which eventually produces dystonic movements.

develops as an occupational disorder through repetitive and precise use of a specific body part over a prolonged period of time (occupational dystonia). Typical symptoms include involuntary movements and muscular cramping as well as sensory dysfunctions. The incidence of FTSD depends on the type of tasks involved, and ranges from 0.008% for writer's cramp to 2% for musician's dystonia (Altenmüller, 2003; Altenmüller et al., 2012). A recent study reported that the frequency of dystonic problems, i.e., embouchure dystonia, is even higher in brass musicians (i.e., trumpet players) (Steinmetz et al., 2014). As few effective treatments are available, FTSD is a potential threat to the professional careers of those affected.

An increasing body of literature focuses on phenomenology, pathophysiology, etiology, and clinical interventions associated with FTSD. In this review, we aim to summarize recent empirical data on the pathophysiological mechanisms of FTSD, with a particular focus on maladaptive changes in the neural representations of the body resulting from over-practicing. As the alleviation of dystonic symptoms may be possible via intervention techniques targeting the altered neural structures, a causal relationship may exist between use-dependent maladaptive changes and movement abnormalities in people with FTSD (Fig. 1).

1. Phenomenology of task-specific focal dystonia

FTSD typically involves a single body part used repetitively under precise control. For example, writers and painters are more likely to develop FTSD in the dominant hand, whereas string musicians such as violinists and cellists more frequently suffer from FTSD in the nondominant left hand. Dystonic symptoms, including involuntary movements and muscular cramping, primarily emerge while a patient is performing specific over-trained motor tasks. For instance, musicians with FTSD may present symptoms during a musical performance, but not during writing. Indeed, a study using a variety of motor tasks demonstrated that musicians with FTSD were able to perform highly precise nonmusical fine motor tasks without any strong signs of a deficit (Hofmann et al., 2015). Similarly, patients with writer's cramp may exhibit an increase in finger force during handwriting, but not during other fine motor tasks (Schneider et al., 2014). However, in the advanced stages of FTSD, dystonic symptoms may 'overflow' to other parts of the body, such as the adjacent digit and/or contralateral hand, resulting in segmental and/or bilateral dystonia.

FTSD impairs motor performance. For instance, the dystonic symptoms of FTSD can degrade movement accuracy during musical performance in pianists (Furuya and Altenmüller, 2013a,b; Jabusch et al., 2004), tone production in brass players (Lee et al., 2014), and voice production in singers (Halstead et al., 2014). More specifically, pianists with FTSD demonstrate larger variability in both the timing and force of successive piano keystrokes (Furuya and Altenmüller, 2013b). Brass instrumentalists with embouchure dystonia and singers with laryngeal FTSD display instability of tone production (Halstead et al., 2014; Lee et al., 2014).

To further investigate the phenomenology of FTSD, several studies have conducted kinematic measurements by means of a motion-capture system or data gloves (Adler et al., 2011; Curra et al., 2004; Furuya et al., 2015; Schneider et al., 2010). One such study examined the characteristics of "yips", a type of FTSD characterized by a transient tremor, jerk, or spasm that occurs while a golfer tries to chip or make a putt. Patients with yips show abnormally pronounced pronation and supination at the right wrist, which is dominantly used for putting in right-handed individuals. This abnormal wrist movement disrupts the directional control of movements during putting (Adler et al., 2011). This symptom may originate from impaired control across different muscles, for instance, the activation of task-relevant muscles and inhibition of surrounding muscles (e.g., surround inhibition). A recent study used a data glove to demonstrate an association between the loss of fine motor control and kinematically abnormal finger movements during musical performance in pianists with FTSD (Furuya et al., 2015). In this study, the authors employed principal component analysis to decompose hand kinematics into three distinct patterns of movement coordination across fingers. They found that, compared with healthy pianists, those with FTSD displayed lower independent control of movements between the affected finger and a finger used to generate a keystroke. In addition, with a regression analysis an association of the altered coordination was found with impaired finger motor control. Similarly, a study also reported less individuated finger movements in patients with writer's cramp (Curra et al., 2004). Taken together, it is likely that FTSD impairs independent force control across muscles, thereby degrading fine motor control.

Patients with FTSD typically display an abnormal increase in the amount of simultaneous contractions between agonist and antagonist muscles, termed muscular co-contraction (Berardelli et al., 1998; Cohen and Hallett, 1988). An excessive level of muscular co-contraction has been reported in patients with writer's cramp and musician's dystonia (Cohen and Hallett, 1988) as well as in patients with golfer's yips (Adler et al., 2005, 2011). This phenomenon is thought to emerge as a symptom of FTSD. However, muscular co-contraction is typically elevated in proportion to the accuracy demand of movements, even in healthy populations (Gribble et al., 2003; Osu et al., 2004). This finding enables an alternative interpretation regarding increased co-contraction in FTSD patients: affected individuals may attempt to compensate for decreased movement accuracy, resulting in excessive muscular co-contraction. Therefore, whether muscular co-contraction reflects a compensatory mechanism or is directly associated with the loss of fine motor control has yet to be established.

In addition to movement abnormalities, FTSD is also characterized by dysfunctional sensory processing (Avanzino et al., 2015; Quartarone et al., 2014). FTSD impairs both spatial and temporal discrimination capabilities, suggesting defective tactile perception

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