Is Skater's Cramp a Task-Specific Dystonia?

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Discussion

An undiagnosed and mysterious movement disorder in speed skaters known in the Netherlands as *zwabbervoet* and in Japan as *bura bura*, has ended the careers of multiple Olympic level speedskaters over the last decades. (1,2). In 2014 it was suggested by neurologists that *zwabbervoet* – known in English as skater's cramp– was a task-specific dystonia (TSD) based on video analysis showing a consistent, and respective jerk, and clinical expertise that identified key suggestive features such as task specificity and a sudden onset (3). TSD is a sub-group of the more general Dystonia, which is a hyperkinetic movement disorder caused by abnormal brain activity that leads to "sustained and intermittent muscle contractions'" in affected individuals (4,5). In TSD, these contractions hinder the execution of a single skilled task, leaving others movements unaffected. Any task requiring skilled motor control can be affected, but its prevalence is highest where motor-demands are highest like in sports and music (6).

The studies presented in this thesis attempted to build on the strength of the initial clinical diagnosis of TSD in skater's cramp, with the use of quantitative measurements. Methodologies were selected based on prior research that identified features of TSD, with the caveat that they allowed us to collect data while speed skating. Features of movement, muscle activity, inter-muscular coherence and psychometrics were all chosen to answer our principal research question: *is skater's cramp a TSD?* Four chapters looked at 4 features of TSD: kinematics (**chapter 2-3**), electromyography (**chapter 3),** coherence analysis (**chapter 4**) and psychometrics (**chapter 5**). In all research chapters we also marked clinical features (**chapter 2-5**). A final aim was to investigate current treatments for TSD in sport (**chapter 6**) as a means of gauging their effectiveness in treating those with skater's cramp who have already been clinically diagnosed as having a TSD.

Outline of Discussion

In this discussion, the results of these investigations are presented in 3 major sections. In **section 1** the major quantitative analyses involving kinematics, EMG, coherence and psychometrics are summarized and discussed. Additionally the systematic review of treatments for TSD in sports is also summarized and discussed. **Section 2** answers our major thesis question: "*is skater's cramp a TSD?"* and discusses the pathophysiology of skater's cramp within the context of two major aspects of dystonia: maladaptive dis-inhibition and the motor control model of TSD. Subsequently, alternative explanations for skater's cramp are examined. Finally in **section 3** recommendations for future research and treatment are made.

Section One: Summary of Major Findings

In four experiments studying skaters affected with skater's cramp, clinical, movement, muscle, coherence and psychometric features were tested. Kinematic and EMG data was tested for various features of movement and muscle activity, respectively, related to TSD **(chapter 2 and 3).** Higher inter-muscular coherence was investigated as this would show evidence of dysregulation in the brain, likely related to subcortical areas such as the basal-ganglia previously implicated in TSD **(chapter 4)**. Psychometrics in the form of a specifically designed survey was used to measure for personality traits previously shown to be aberrant in TSD **(chapter 5)**. The results of these investigations are reported below.

Clinical Findings

Clinical features played a pivotal role as part of the goal to test whether skater's cramp was a TSD. In **chapter 2, 3 and 5** results were suggestive of this. Skater's cramp was task-specific, with only a few cases showing spread to adjacent related movements like inline skating. This spread was in line with previous studies of TSD (7). The condition presented without any pain, and the rate of onset, age of onset, lack of successful treatments and triggering factors like equipment change and injury all matched with profiles from other TSDs (6,8). Additionally, the ratio of men to women, a striking disparity that is biased towards men in TSD between 2:1 and 4:1 (9,10), was similar in skater's cramp at 3:1 **(chapter 5)**. All affected skaters showed a normal neurological examination, and no skaters reported any musculoskeletal issues. It is common in other forms of TSD that those affected have no other health complaints. Altogether, the clinical presentation of skater's cramp was in line with other TSDs.

EMG/Kinematics

Chapter 2 and 3 found a consistent repetitive jerking of the foot that coincided with over-active muscles in the impacted leg in all affected skaters. The jerks were characterized as a higher absolute value of angular velocity in the endo- exo-rotational axis of the impacted foot, and coincided with higher muscle activity, measured with surface EMG, in the tibialis anterior, peroneus longus, soleus and gastrocnemius. The over-activity was consistent across stroke cycles, and confined to the moment of skate placement in all affected skaters. The results of video analysis of all affected skaters showed the same pattern of active, temporally and spatially stereotypical features across the whole cohort. Importantly, despite the moment-specificity observed in EMG activity, affected individuals appeared to have an individually-specific jerking pattern (some exo- or endo- rotational) and the severity of skater's cramp also differed greatly between individuals. No aberrant activity was noted in the non-impacted leg of affected skaters or in the legs of control participants. Over-activating muscles combined with aberrant movements occurring in a person-specific, consistent and patterned fashion are suggestive of TSD in general and lower limb TSD specifically (11,12).

Skating at different levels of intensity had no effect on the severity of muscular activity in the impacted limb of affected skaters. Aberrant activity remained consistent as skaters were asked to skate at 60% and 80% of their perceived maximum intensity (**chapter 3**). This finding relates skater's cramp to TSD, as it appears that the damage to motor programs in TSD is scalable to different weights and intensities (13). This is in contrast to if skater's cramp was resultant of a peripheral issue (such as muscle damage or neuropathy), where muscle and movement patterns would be more likely to change depending on the intensity of its execution (14,15). We interpret the consistency despite differing task-intensity in skater's cramp as another suggestive feature of TSD.

Inter-Muscular Coherence

There was no difference in inter-muscular coherence (IMC) in a generic stationary resistance/activation task (unrelated to skating), however, while skating, IMC in the theta frequency band (3-7Hz) was higher in the impacted leg of affected skaters compared to their non-impacted leg, and compared to the left and right leg of healthy controls. IMC is a coefficient determined by the consistency of phase differences between two signals from two different muscles (ranging from 0 to 1, i.e. no coherence to perfect coherence). IMC has been shown to be higher in dystonia generally (16–20), and in two studies of TSD in writer's cramp (21,22). Higher IMC is a marker of dystonia, because it provides evidence of abnormal synchronization in cortical and subcortical brain areas that have been linked to involuntary movements down the cortico-spinal tract (23). It should be noted that increased IMC in skaters with skater's cramp was only found in the more severely affected sub-group. Although tentative, these results using a novel approach involving wavelet coherence, suggest a central drive and by extension TSD.

Psychometrics

Skaters with skater's cramp had a different personality profile compared with age-, sex- and experience-matched controls. Sentimentality (a facet of emotionality) was higher, but only in males. Anxiety (also a facet of emotionality) also appeared higher in affected male skaters (but marginally p=.07). Higher emotionality correlates with TSD, and it is thought this is because maladaptive dis-inhibition of striatal pathways for both TSD and emotional regulation are partly shared (24). Identifying this exclusively in males is a complication to the findings, but as discussed in **chapter 5**, this may also be true for previous studies of TSD as it is a very understudied area of research. Unexpectedly, extraversion was lower in affected skaters compared to controls. One possible shared mechanism that may explain this finding is dysregulated striatal dopamine, that has been shown to play a role in both regulating higher neuroticism (25) and lower extraversion (26), as well as higher TSD prevalence (27).

Treatment of TSD in Sports - a Systematic Review

The systematic review in **chapter 6** revealed treatments of TSD in sports are poorly understood with very little quantitative evidence to support treatment strategies. We reviewed both non-invasive psychological and invasive and/or pharmacological interventions and found no evidence of effective treatment. There were some reports that both psychological and surgical interventions may be effective. In athletes, botulinum toxin injection was only partially effective (58%), similar to findings in musicians (28). Notably, emotional regulation techniques like cognitive behavioral therapy (CBT) and eye movement desensitization and reprocessing (EMDR) reported some improvement. It is important to recognize that these studies were limited to golfers, and significant differences in treatment efficacy for psychological interventions may be sport dependent. Another form of treatment attempted was invasive surgery. Ventro-oral thalamotomy (VoT) surgeries appeared to be mostly effective in a retrospective study of 171 cases of TSD (including 5 athletes) measuring the Task-Specific Focal Dystonia Scale (TFDS) (29). Importantly, the risks of such surgeries are considerable therefore the appropriateness of their use must be considered critically.

Section 2 Overall Discussion

What is Skater's Cramp?

In totality, the constellation of quantitative features described in this thesis: clinical characteristics, kinematic/EMG patterns, higher intermuscular coherence and distinct personality characteristics, were all supportive of our hypothesis that skater's cramp is a TSD.

Although it is impossible to state that these findings are confirmatory, this thesis found features in skater's cramp that implicate involvement of the central nervous system. Evidence for this was found in the overflow of muscular activity observed in **chapter 3** and higher inter-muscular coherence observed in **chapter 4**. Both these findings may be related to the brain through an aspect of the pathophysiology of TSD known as *maladaptive dis-inhibition*. Evidence was also found that skater's cramp was a consistent and repetitive jerking occurring irrespective of differences in intensity (**chapters 2 and 3**). These findings likely relate to another aspect of TSD pathology: *the motor control model of TSD* (30).

Maladaptive Dis-inhibition

Maladaptive dis-inhibition is hypothesized to be a major component in the pathophysiology of dystonia. Performing any movement requires a finely regulated balance between inhibitory and excitatory circuits that allow for appropriate movements and suppresses competing undesirable ones. Failure to properly suppress unwanted movement, i.e. failure

to properly inhibit, appears to be an important factor in dystonia (31,32). Protocols of short- and long- intercortical inhibition (SICI and LICI) measure aberrant modulation of cortical excitability, and have found a failure of inhibition in dystonia (33,34) and TSD in particular (31). In combination with neuroimaging data (35), these findings suggest a loss of inhibition in dystonia that is likely regulated by gamma-amino butyric acid type A (GABA-A) receptors in substructures of the basal ganglia (5,31).

This proposed dis-inhibition in cortical and subcortical motor networks is thought to produce flaws in motor signaling down the cortico-spinal tract. These flaws result in agonist- antagonist muscle pairs of the impacted limb exhibiting co-contraction and maladaptive muscular overflow (36), as well as higher IMC (21,23). These features have been seen in different forms of dystonia, such as DYT1 dystonia (16), acquired childhood dystonia (17) and myoclonus dystonia (18). In TSD specifically, a similar pattern of symptoms has emerged with both over- and co-activation of muscles uninvolved in skill execution (37,38) and higher IMC in writer's cramp (21,22).

Connecting these findings to this thesis, affected skaters both exhibited muscular overflow in **chapter 3**, and higher inter-muscular coherence in **chapter 4**. In skater's cramp, muscular overflow was maladaptive to the affected skill, as it was not compensatory to the skating movement, suggesting it is an overflow-related dis-inhibition similar to other forms of TSD (31). Perhaps more convincing, though, were findings from **chapter 4** showing higher IMC in the most severely affected skaters. Coherence was higher in the key theta frequency band (3-7Hz), found in other forms of dystonia and TSD (17,21). **Chapter 4** is the first study of IMC in TSD to collect data during task-execution. As there are no other studies of TSD validating the idea of higher IMC in a task-specific setting, results from **chapter 4** are preliminary. In sum, findings from **chapters 3 and 4** are task-specific examples suggestive of maladaptive dis-inhibition along the cortico-spinal tract, implicating TSD.

Finally, in addition to **chapters 3 and 4**, higher emotionality observed in affected skaters in **chapter 5** may also relate to TSD through dis-inhibition. In both cases of TSD and high anxiety sensitivity, a shared neuro-circuitry involving the basal-ganglia and motor cortex has been found to be over-active (39–43). This has prompted researchers to posit that a major cause of both problems may be related to maladaptive dis-inhibition (24). Therefore, in **chapter 5**, results showing higher emotionality in affected skaters may correlate with a higher prevalence for TSD through a shared cortico-striatal dis-inhibitory mechanism.

Motor Control Model of TSD

Another way in which our findings in skaters implicate the brain is related to the *motor control model of TSD* (30). In this model, plasticity that drives the learning of specialized

motor-skills is thought to malfunction and become maladaptive, increasing the risk of developing TSD.

In dystonia generally, researchers have found evidence of maladaptive neuro-plasticity (5). In healthy individuals neuro-plasticity is defined as "the ability of the nervous system to respond to intrinsic or extrinsic stimuli by reorganizing its structure, function and connection" (44). Within a motor control context it drives the adaptation of movement to increase precision and efficiency (45). However, in the context of movement disorders it is hypothesized that plasticity can also be maladaptive, where reorganization of the brain results in a loss of motor control (5). Evidence for this in dystonia has been found in abnormal organization of the motor homunculus (46), as well as clear excesses of plasticity and hyperexcitability in affected individuals (47,48).

In TSD, studies have also found evidence of maladaptive plasticity reorganizing both structure and function in the brain (8,37). However, these structural changes to the brain do not result in broader movement disturbances as in other forms of dystonia. It has been proposed this is because TSD is driven not only by maladaptive plasticity (49), but also errors in motor control (8). Together these two processes form the basis for the *motor control model of TSD.*

In this model, problems begin due to errors developing in highly specific motor programs that are part of highly trained skills. Researchers hypothesize that an over-practiced skill increases the length of a motor program beyond a certain limit (50). Once this limit is reached, maladaptive plasticity can be triggered by behaviors that negatively influence the motor program (49). Such behaviors are called "trigger factors" and are thought to cause a "mismatch between the capacity of the individual's motor control system, and the required movement" (30,50). Often these mismatches are caused when something in the peripheral environment of a movement changes, triggering the rigid motor program to try, but fail, to adapt. Trigger factors can include a sudden change in biomechanics, for example caused by injury or equipment change (13). A fascinating history of trigger factors involving equipment has been noted, including accounts describing "epidemics" of TSD following the mass adoption of steel-nibbed pens replacing quills, and electric pianos replacing analogue ones (13). Therefore, in the motor control model of TSD, trigger factors cause motor control error resulting in a sudden re-organisation of highly specific motor engrams (maladaptive plasticity) that leads to typical features of TSD.

In skater's cramp, our clinical findings in **chapters 2,3 and 4** found many examples of trigger factors that appeared to fit within the motor control model. In the majority of participants we noted many examples of trigger factors including changes in technique, and equipment; as well as biomechanical changes caused by a fall preceding the

development of skater's cramp. Importantly, these observations are cursory and a more robust epidemiological investigation is warranted.

Within the context of the motor control model of TSD, researchers have theorized that errors are limited to certain sections of the broader motor program. It is speculated that this may explain why TSD symptoms often present with high spatiotemporal consistency, and at a consistent moment and position (30). Furthermore, it is proposed that symptoms will occur with greater frequency when demands on motor control are highest – positing narrower degrees of freedom in a movement increase susceptibility to motor programmatic error (30). Evidence of spatiotemporal consistency, task/moment-specificity, and relation to task-difficulty are apparent in a host of sports such as golf, baseball, billiards, rowing, archery etc. where jerking appears to occur in the same way and at the same time in affected individuals during moments of greatest motoric challenge (51–60). In classical music, many TSDs are moment-specific as well, with some forms even presenting only during a particular piece of music that corresponds with the highest level of difficulty (61). In further support of a correlation with task-difficulty, in music, TSD preferentially affects the limb with the highest motor control demands, this is the right hand of pianists and the left hand of bowed instrument players (62).

Relating these insights to our thesis, a very similar pattern appeared in skater's cramp of highly consistent repetitive jerking that was moment- and position specific, as well as a being correlated to task-difficulty. It was shown visually, kinematically and muscularly that problems arose consistently over many skating strides in individuals, and that they were both moment- and position-specific to skate placement in **chapters 2 and 3**. We can infer that skater's cramp correlated with the highest moment of task-difficulty in speedskating, because skate placement is the most challenging moment in the skating stroke (the skate must land on the ice at high speed in the correct direction and any small deviation can cause a fall). Also, as in other TSDs, a greater impact on the limb with highest motor control demands was apparent. In skating this is the left leg, and over **chapters 2, 3 and 4** clinical data showed a skewed ratio of 35/45 L/R (75%).

Although it seems that TSD symptoms correlate with task-difficulty (13), they appear to correlate less with task-intensity. For example, in golf, it was found that symptoms remained consistent despite differences in the intensity of the golf swing and distance from the hole (63). Similar qualitative observations have been made in other lower limb TSD (64,65) This aspect can also be explained within the motor control model of TSD (13), where research has proposed that the complexity of affected motor-programs likely makes them scalable to different intensities (8,66,67). In **chapter 3** we hypothesized that this would also be observable in affected skaters as consistencies in muscle intensity and movement patterns irrespective of changes in skating, and this was confirmed, as jerking

remained highly consistent in spite of changes in skating intensity between 60% and 80%. These findings appear to be in agreement with the concept of a corrupted central driver, which is fundamental to the motor control model of TSD.

In sum, the aberrant muscular overflow and IMC described in **chapters 3 and 4,** and the higher emotionality observed in **chapter 5** suggest that those with skater's cramp exhibit maladaptive dis-inhibition. In its consistency in space, time and, intensity, and its correlation with task-difficulty observed in **chapters 2 and 3**, skater's cramp also fits within the motor control model of TSD. Taken together, both maladaptive dis-inhibition and the motor control model are key facets of TSD, making our quantitative findings from **chapters 2-5** broadly supportive of our major hypothesis.

Alternative Explanations

Prior to this thesis, clinicians, coaches and experts in movement have proposed other explanations for skater's cramp such as a peripheral neurological/musculo-skeletal, or mechanical problems (1). The results of this thesis broadly contradict these assumptions in favor of TSD.

Results in **chapters 2-5** made alternative movement disorders an unlikely cause. For example, skater's cramp was found to be painless in all skaters **(chapters 2, 3 and 5)** reducing the likelihood it is a functional dystonia, peripherally induced dystonia, or myotonia. The consistent and persistent patterns **(chapters 2 and 3)** in skaters reduce the chances that skater's cramp is a functional dystonia (which would fluctuate while skating) or paroxysmal dyskinesia which presents intermittently in shorter "attacks" and is not task-specific (68).

Our findings also make it less likely that skater's cramp is a peripheral nerve or muscle problem (1). A chronic musculoskeletal problem, i.e. damage to a joint or muscle causing instability (or co-contraction caused by fear as a result of instability), would likely not present as a uniform muscle and movement pattern as seen in **chapters 2 and 3**. Another proposed explanation, peripheral neuropathy, would create a weakening of the affected leg, while results from **chapter 3** showed clear universal over-activity. Also differences in coherence **(chapter 4)**, and personality **(chapter 5)** in skaters do not support a peripheral explanation, as they are inherently of a neurological origin. Finally, participants reported no actual peripheral ailments in any of our clinical assessments **(chapters 2, 3 and 4)**.

Another suggestion was a mechanical or technical flaw in speedskating. An incorrect placing of the skate upon the ice was proposed to result from flawed positioning and timing of back, hip and knee joints. From this perspective, a successful placing of the skate may not be pro-active, but a muscularly passive action dependent on precise positioning

at skate placement (applying a torsion spring model). Findings from this thesis, and from previous research, contradict this hypothesis. Firstly, it was clearly shown in **chapters 2 and 3** that over-activation of the affected foot occurred at skate placement. Higher muscular activation in the affected leg would not be expected if skate placement were modeled as a torsion spring, nor would higher IMC be expected. Furthermore, skater's cramp proved persistent regardless of changes to the weight of the foot and the intensity of skating, indicating that a mechanical issue is unlikely (69). As a replacement theory for TSD, peripheral neurological/musculo-skeletal, or mechanical problems are not supported by the observations of this thesis.

Section Three Future Directions

The findings of this thesis characterized skater's cramp as a TSD by measuring movement and muscle activity. In this section we will focus on suggestions for future research that may assess the major question of this thesis by measuring the brain directly. Specifically, the possible use of fMRI, fNIRS and EEG is discussed. We further examine which of these quantitative measures could best evaluate treatment effectiveness in future interventions studies (outcome variables). Additionally, we elaborate on possible future treatments.

Future Studies

The next step in investigating if skater's cramp is a TSD is obtaining measures of brain activity in affected skaters to gain insight into the specific brain areas involved and find possible further evidence of a centrally driven form of dystonia.

fMRI is a possible choice for future investigations. Studies in other forms of TSD in writing and music observed: 1) abnormal inhibition both in the motor and somatosensory networks, 2) abnormal somatotopic representations of affected limbs and 3) subcortical abnormalities (7). While deep penetration allowing for a subcortical analysis is an advantage of fMRI compared to other brain measures, disadvantages include common spatial and temporal artifacts that result from movement, physiological noise (breathing and heartbeat), and other measurement distortions. Specifically, movement artifacts are a problem when measuring skater's cramp, obviously negating the use of the stationary fMRI while skating. One alternative may be visualization, i.e. imagining, speed skating while in the scanner. Studies of TSD in writers registered abnormal surround inhibition in dystonic patients triggered simply by imagining the movement of their dystonic limb, supporting this approach (70). Nevertheless, remaining motionless while visualizing skating may still prove challenging, as prior studies have shown it can be challenging for participants to remain inactive during movement imagery, specifically in sports (71).

Measuring brain activity through fMRI in skater's cramp not only offers insight into underlying pathophysiology, but may also give further insights into brain changes that may occur after a treatment intervention. fMRI has been used in multiple dystonia studies to show evidence of intervention-based changes. Invasively treating general dystonia using DBS found functional network state changes in patients before compared to after the treatment(72). Conversely, a non-invasive study investigated the effects of a sensory trick on cervical dystonia and showed decreased sensorimotor network connectivity and increased cerebellum activation after the intervention (73). Similar possible analyses may be conducted in skaters, looking at generalized and task-based (using visualization) changes in participants' brain activity.

fNIRS is an alternative to fMRI for measuring the blood oxygenation dependent response in TSD. Compared to fMRI, fNIRS is far less susceptible to movement artifacts and is available in a portable form that could possibly be worn while speedskating. Unfortunately, its use of light to infer blood oxygenation results in a very low degree of cranial penetration. This limits assessment of the regions representing the leg and foot, which are located away from the outwardly exposed motor and somatotopic cortices (deeper within the longitudinal fissure, unlike the externally exposed hand or mouth areas). Nevertheless, fNIRS may still prove useful as inter-hemispheric imbalance is a feature of TSD that was located with this approach (74). As the temporal resolution is superior to fMRI (100Hz), these imbalances could be visible per-stroke while skating.

EEG may also prove useful. Although movement artifacts are a concern in EEG, they are less serious than fMRI and can be partially overcome by new filtering techniques (75). Unfortunately EEG has low spatial resolution, and low penetrative capacity making it comparably ineffective in measuring somatotopic abnormalities to fNIRS. However, its very high temporal resolution allows us to observe subcortical frequency oscillations, thought to drive the inter-muscular coherence observed in this thesis (76). It can also measure abnormal inter-hemispheric coherence, as well as bilateral event related de- and re- synchronization (ERD and ERS) that have been shown to be abnormal in other forms of TSD in sports (40).

Future Treatments

Based on the current standard that TSD is diagnosed clinically by neurologists, and the findings from this thesis suggesting skater's cramp is a TSD **(chapters 2-5)**, we recommend that the most successful treatments noted in the systematic review of treatments for TSD in sports **(chapter 6)** be trialed in skater's cramp. These treatments fall into two major categories: 1) the use of psychological interventions involving either the regulation of emotions, or sensory-motor retraining strategies; and 2) invasive and pharmacological

strategies including the use of anxiolytic drugs, botulinum toxin, and in some exceptional cases, invasive surgery.

Treatment protocols for TSD often involve both invasive and non-invasive interventions employed at the same time. Such a multifaceted approach has shown promise in the treatment of musician's and writer's dystonia. For example, invasive botulinum toxin was more effective when combined with non-invasive occupational therapy in writer's cramp (77) and sensory-motor retraining in musician's dystonia (78). TSD is considered a network disorder, therefore researchers posit the greater effectiveness of these multimodal approaches is likely because they target multiple mechanisms that underlie TSD simultaneously. For instance, psychological interventions are thought to aid in the reprograming of damaged motor engrams fitting with the motor control model of TSD (30), while simultaneously the effects of botulinum toxin may both influence maladaptive neuro-plasticity and dis-inhibition. This is because, next to its role as an acetylcholine release inhibitor resulting in local muscle weakening, botulinum toxin also influences the central nervous system by inhibiting gamma motor neurons and by extension reducing afferent sensory input from muscles to the central nervous system (79).

In light of these findings, a psychological intervention such as emotional regulation or sensory motor retraining and/or botulinum toxin is recommended in skater's cramp. Importantly, some limitations must be recognized in the use of botulinum toxin as a treatment. In cases of TSD it has been less successful in treating musicians and athletes compared with writer's cramp where the motor control challenges are proposed to be less extreme (80). In addition to coordination decrements, possible losses of strength due to botulinum toxin injection is of special importance in speed skating, due to safety concerns around falling. Despite these risks, with the appropriate safety protocols in place, botulinum toxin injections are likely feasible. This would likely include an incremental buildup in dosages in affected muscles starting at a very low dose, while conducting continuous testing of a skater's ability to maintain coordination and control between dosage increases. Importantly, the application of an outcome measure during skating would be a useful addition to the self-reported response of skaters and coaches. Therefore, applying the methods pioneered in this thesis, including measuring movement, muscle activity, and possibly IMC **(chapters 2-4)** throughout the administering of the botulinum toxin regimen is recommended.

Impact

The efforts in this thesis to understand skater's cramp as a possible TSD, and the efforts to review possible treatments, were helpful steps towards improved medical outcomes for people suffering from this affliction. This is essential due to the traumatic emotional and physical repercussions of developing the disorder. During the course of this research we

noted many first-hand accounts of affected skaters reporting that the loss of their skating ability was synonymous with the loss of an intimate relationship. Adding to this emotional trauma is the anxiety and confusion that results from the medical community not knowing what skater's cramp is or how to treat it. Beyond the emotional toll, some skaters reported undergoing failed invasive medical procedures such as surgery for compartment syndrome. Therefore both on an emotional and physical level, a better understanding of skater's cramp could offer relief to affected skaters in the present and future.

Conclusion

This thesis showed evidence in multiple domains to support our hypothesis that skater's cramp is a TSD. The results fit well within the cognitive and physiological models proposed to explain TSD, specifically maladaptive dis-inhibition and motor control. These findings also made less likely many alternative explanations for skater's cramp, such as mechanical issues, peripheral neuropathies, and functional dystonia. In the future, fMRI or EEG studies may help gain insight into the neurological pathophysiology of skater's cramp. Moreover, well conducted intervention studies are needed to find treatments options in skater's cramp. Techniques pioneered in this thesis including movement, muscle and coherence analysis could help in this pursuit. Conducting these future studies will constitute the next step in the further investigation of the major research question of this thesis: *is skater's cramp a TSD?* Based on our clinical and experimental data, the current answer to this question is: yes.

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