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Inter-muscular coherence in speed skaters with skater's cramp

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ABSTRACT

Introduction: Skater's cramp is a career-ending movement disorder in expert speed skaters noted to be a likely task-specific dystonia. In other movement disorders, including task-specific dystonia, studies have found evidence of central dysregulation expressed as higher inter-muscular coherence. We looked at whether inter-muscular coherence was higher in affected skaters as a possible indicator that it is centrally driven, and by extension further evidence it is a task-specific dystonia.
Methods: In 14 affected and 14 control skaters we calculated inter-muscular coherence in the theta-band in a stationary task where tonic muscle activation was measured at 10%, 20% and 50% of maximum voluntary contraction. Additionally, we calculated wavelet coherence while skating at key moments in the stroke cycle.
Results: Coherence did not differ in the stationary activation task. While skating, coherence was higher in the impacted leg of affected skaters compared to their non-impacted leg, $p = .05$, $\eta^2 = 0.031$, and amplitude of electromyography correlated with coherence in the impacted leg, $p = .009$, $R^2_{\text{adjusted}} = 0.41$. A sub-group of severely affected skaters ($n = 6$) had higher coherence in the impacted leg compared to the left and right leg of controls, $p = .02$, Cohen's $d = 1.59$ and $p = .01$, Cohen's $d = 1.63$ respectively. Results were less clear across the entire affected cohort probably due to a diverse case-mix.
Conclusion: Our results of higher coherence in certain severe cases of skater's cramp is preliminary evidence of a central dysregulation, making the likelihood it is a task-specific dystonia higher.

1. Introduction

Task-specific dystonia (TSD) is considered a disorder of intermittent, sustained muscular over-activation resulting in repetitive movements and postures [1]. Its principally defining feature is task-specificity as it occurs only during the performance of a complex and highly practiced skill and rarely generalizes to affect daily life [2]. TSD is thought to originate from maladapted motor engrams that result from over-practicing a repeated movement while enduring a peripheral change (trigger factor), such as stress, equipment change, or injury [3]. These corruptions to the motor engrams are thought to result in dysfunction of central neural networks, such as dis-inhibition in cortical and subcortical brain areas that drive complex movement [4]. This results in stereotypical patterned jerking, over-activation and co-activation of muscles while performing a specific skill.

Skater's cramp is a mysterious movement disorder in speed skaters. It is referred to as skater's cramp (as opposed to speed skater's cramp) as an umbrella term accounting for the possibility it may also exist undiagnosed in hockey and figure skating. It was initially described as a sudden uncontrollable lateral jerking rotation of the ankle joint as a skater placed their skate on the ice, and was suggested to resemble a TSD [5]. Further investigations of 5 skaters in a case control study showed visual and kinematic evidence of a stereotypical patterned over-active jerking reminiscent of TSD [6]. Despite these suggestions, there is still currently no direct evidence linking skater's cramp to abnormal centrally generated muscle patterns in the brain. For this reason, to test whether skater's cramp is a centrally driven problem, we chose to perform an analysis of inter-muscular coherence (IMC).

IMC is a linear correlation-like coefficient that is determined by the consistency of phase differences between two oscillating signals from

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two muscles, and ranges from 0 to 1 (no coherence to perfect coherence). IMC is an important marker of dystonia, because without a direct measurement of the brain, such as fMRI, it can still provide evidence for central problems such as an abnormal synchronization which can be linked to cortical drivers of involuntary movements [7]. This has been shown in many related movement disorders such as DYT1 dystonia [8], acquired childhood dystonia/idiopathic/genetic dystonia [9], myoclonus [10], tremor and Parkinson's disease [11,12].

In TSD, experiments have looked at IMC with mixed results. In two studies of writer's cramp IMC was not found to be higher than controls [8,13], and in one study IMC was only higher in patients with tremor [14]. Only in a single study of writer's cramp did researchers find higher IMC in the range of the theta-band (3–7Hz) [15]. Importantly, this study used a generic 'pinch grip' task unrelated to writing, measuring blocks of 30 s of tonic contraction and finding higher IMC at 10% of maximum voluntary contraction. A generic task was used to avoid undue muscle and movement activity, ensuring the basic assumptions of a stationary signal and no phasic contraction or relaxation (causing artifact low frequency coherence) [16]. To adhere to these assumptions, we opted for a similar generic stationary activation task in speed skaters. This task constituted the first of two experiments measuring IMC. In this experiment we aimed to answer the research question: is IMC higher in skaters with skater's cramp in a stationary task?

In addition to the generic stationary activation task, we investigated IMC task-specifically (while skating). At the time of writing, no TSD experiments in sports have measured IMC while performing the affected skill, possibly because dynamic movements may violate the assumption of static muscle activity and positioning required in traditional calculations of IMC. Therefore, to measure IMC while skating (where problems occur briefly during a dynamic skating stroke [6]), we used wavelet coherence. Unlike the generic stationary task, using wavelets can avoid longer measurements by detecting coherence over shorter time-blocks, improving detection in more dynamic movements where previous indications may have been lost to averaging [17]. Higher temporal resolution also avoids misinterpreting artifacts caused by spinal reflex loops [16]. We therefore employed wavelet coherence to avoid the traditional limitations of standard coherence analysis, and measured IMC while skaters skated. Our second research question was: is IMC higher in skaters with skater's cramp while they skate?

In keeping with the most comprehensive previous study [15], we hypothesized IMC would be higher in affected skaters in their impacted leg in a generic stationary activation task, and also while skating, when compared to a control group.

2. Methods

2.1. Participants

We recruited participants through a posted article on a popular Dutch skating website (www.schaatsen.nl). Of 50 possible candidates, 15 were randomly selected for an oral interview to determine whether they met the inclusion criteria. The inclusion criteria consisted of a minimum of ten years of skating experience with a minimum pre-onset practice-frequency of twice a week and 5 years of symptom-free skating prior to symptom-onset. Oral interview inclusion criteria required the condition to occur only while skating and present as an active patterned jerking. Finally, candidate participants underwent a physical exam by a neurologist (MT and AS). Covid-19 restrictions prevented 3 of 15 participants from being exanimated (no neurological issues were reported). Exclusion criteria: a neurological disorder recognized during examination. 14 participants were matched with controls based on skating proficiency as measured by dedication (years of practice and sessions per week). We obtained informed consent from all participants.

2.2. Task

All participants were fitted with surface electromyography (EMG) (Shimmer3 shimmersensing.com) that collected time synchronized measures of four muscles in both their lower legs at 512Hz. Sensors were placed on the peroneus longus (PERO), tibialis anterior (TA), gastrocnemius (medial side) (GAS), and soleus (SOL). We chose lower leg muscles as research in other TSDs showed the majority (75%) of cases presented distally [18]. Electrodes were Ag/AgCl 24 mm adhesives (covidien.com) placed 20 mm apart.

2.2.1. Experiment: Generic stationary activation task

Participants were seated with knee and ankle at 90° respectively and instructed to activate all lower leg muscles in both their lower limbs in a non-resisted task. Initially, they were instructed to co-activate their lower legs at maximum non-resisted voluntary contraction for 5 s. Subsequently, they were asked to activate both lower legs at their subjective assessment of what 20% and 50% of maximum non-resisted voluntary contraction would be for 30 s twice, respectively.

For the isometric resistance task, participants were seated with knee and ankle at 90° in a bespoke lower limb dynamometer designed to mimic the activation pattern of skater's cramp mimicking protocols for IMC measurement in writer's cramp [15]. Participants' feet were fixed to foot-plates and positioned so that during plantarflexion the foot would experience resistance in the direction of endo-rotation. Participants were asked to plantarflex while resisting endorotation (keeping the foot straight). To create a baseline, dynamometer readings were recorded while participants exerted maximum voluntary contraction (MVC) while keeping their foot straight for 5 s. Subsequently, participants underwent 1 block of 30 s at 10% of MVC, and 2 blocks at 20% and 50% of MVC respectively per leg. There was a minute-long rest between trials. These percentages of MVC were chosen based on previous studies of IMC in TSD [15]. Participants were given oral cues ('higher', 'lower' and 'hold') at regular intervals (approximately every 5 s) to maintain the appropriate resistance level throughout each trial. We refrained from showing real-time force output to avoid the known issue of 'chasing', where participants' motivation to maintain exact force-output results in steady oscillation around the target value (increasing total variance) [19]. Giving an oral cue maintained a more stable force output (more important than a specific force level).

2.2.2. Experiment: In-skating task

Participants skated two sets of four laps at 60% of maximum speed and two sets of two laps at 80%. Inter-trial rest periods were 2 gliding laps (approximately 4 min).

2.3. Signal processing

Unlike previous studies, we limited our investigation to theta-band coherence analysis (excluding beta-band) due to the confounding role that factors like age [20,21], exertion [22] and coordination [23] might play in interpreting results (Appendix A.1).

2.3.1. Experiment: Generic stationary activation task

We located blocks of sustained muscle activity using an example signal of rectified EMG as a visual indicator, and manually selected and cut out trial-blocks of 30 s. Correct labeling of each trial-block was accomplished by locating MVC. For each participant, for each leg, every trial-series began with a 5-s MVC. We located MVC for each participant visually and by consulting our activity log and we labeled subsequent trials as either 10%, 20% or 50% of MVC.

IMC was calculated for the agonist-antagonist muscle combinations TA/GAS and PERO/SOL using the Welch method with the coherence significance-level calculated using the Halliday et al. method [24] and based on the number of segments using the formula: $\text{signlevel} = 1-(0.05)^{1/(n-1)}$ (Appendix A.3). We assumed within the constraints of our static task that TA/GAS and PERO/SOL were the clearest agonist-antagonist pairs, and did not assess combinations PERO/TA and GAS/SOL due to possible crosstalk [25]. This led to a coherence significance-level of 0.0658 over 45 segments. We chose 45 segments of 1 s with an overlap of 50%, resulting in 23 s of continuous activation corresponding with trial length.

2.3.2. *Experiment: In-skating task*

Skating strokes were classified with a bespoke software package called SkateView [26], that detected ‘skate placement’ (a skate landing on the ice after swing phase) and ‘take off’ (when a skate lifts off after a stroke) from inertial mass units (IMU) data from sensors on skaters’ skates. SkateView also classified straightaway skating from corner skating. Stroke and corner information was synced to all EMG signals.

We calculated IMC from 10-s blocks of straightaway skating as defined by SkateView for agonist-antagonist muscle combinations: PERO/SOL, PERO/GAS, TA/SOL and TA/GAS. We created spectrographs depicting wavelet IMC patterns for individual muscle pairs over 10-s blocks of straightaway skating, where indicators of skate placement were marked to aid in interpretation.

Within these 10 s-blocks, we investigated IMC per stroke quantitatively, at two specific time windows:

1) Skate placement: the period .3 s preceding the skate landing on the ice (as defined by SkateView).
2) Entire Stroke: the entire skating stroke as defined from take-off to take-off (similar to onset of swing phase in a walking gait-cycle).

At these two time windows for each participant, we calculated average coherence for the following four agonist-antagonist muscle combinations: PERO/SOL, PERO/GAS, TA/SOL and TA/GAS. Subsequently, we created a summary IMC score, which was the average of these muscle combinations as one IMC score per leg, per participant for skate placement and over the entire stroke cycle.

Wavelet coherence was calculated with a Morlet Wavelet (FS ratio 4) using a bespoke software module (LabView 2018, www.ni.com) based on previous methods [27]. Coefficients were calculated every 25 ms over 50 bins (upper limit: 50Hz). Boxcar filter and Monte Carlo simulations were used for smoothing and significance limit respectively resulting in sig. threshold of 0.425 [27]. For complete details consult Appendix A.4.

We calculated EMG muscle activity in PERO, TA, SOL and GAS while skating. EMG was filtered with a Journée filter [28] using a low/high band-pass filter at 10 and 50Hz. This envelope filter (high-pass at 10Hz) corrected for dynamic acceleration and deceleration of speed skating movements. Stroke cycles were time normalized with skate placement at the center (50%) of every completed stroke cycle (100%). For complete details consult Appendix A.5. We calculated average EMG at two time windows: 1) Skate placement, 35% and 50%; and 2) Entire Stroke, 0%–100% using the time normalized stroke cycles. Subsequently, we created a summary EMG score which was the average of PERO, TA, SOL and GAS scores as one EMG score per leg, per participant for skate placement and over the entire stroke cycle.

2.4. *Statistical analysis*

Because both stationary and skating tasks were bilaterally symmetrical, we compared the impacted and non-impacted leg of skaters with skater’s cramp to the left and right leg of controls i.e. 14 left and 14 right legs of controls vs the pooled results of the impacted (11 left 3 right) and non-impacted (3 left 11 right) legs of affected skaters (Appendix A.2).

2.4.1. *Experiment: Generic stationary activation task*

A 2x12 mixed design analysis of variance (ANOVA) was performed to compare IMC in the stationary experiment. The dependent variable was the average standard IMC over 30-s time-blocks. The between subjects factor was *group*: affected participants vs control participants. The within subjects factors were *leg*: impacted vs non-impacted leg (left vs right for controls); *muscle combination*: PERO/SOL and TA/GAS; and *intensity*: trials performed at 10%, 20% or 50% of MVC.

2.4.2. *Experiment: In-skating task*

All analyses in the in-skating task were performed using dependent variables: summary EMG (average: TA, PERO, SOL, and GAS) and summary IMC (average: PERO/SOL, PERO/GAS, TA/SOL, and TA/GAS). We performed two 2x2 mixed design ANOVAs with dependent variable summary IMC, between subjects factor: *group* (affected vs control), and the within subject factor: *leg* (impacted vs non-impacted leg [left vs right for controls]). One ANOVA was conducted on the .3 s time-window representing skate placement, and one was over the entire stroke cycle.

For muscle activity we used the same 2x2 mixed design ANOVA but, with dependent variable: magnitude of EMG. Two ANOVAs were performed at skate placement (35%–50% of normalized stroke cycle), and one across the entire stroke (0%–100%).

We used simple linear regression to predict summary IMC scores, based on summary EMG scores. We conducted eight regression analyses on the impacted and non-impacted legs of affected skaters and left/right of controls at skate placement and across the entire stroke cycle.

We performed a sub-group analysis based results of regression (EMG/IMC) and EMG variance suggesting a diverse case-mix. The subgroup comprised of affected participants with EMG scores of their impacted leg higher than the 95th percentile of median EMG scores of the control group (average across both legs). We designated this group as having higher disease severity. Muscle activity has been a proxy for disease severity in other TSDs in runners and golfers [29,30]. We compared IMC of this sub-group to the control group using the same 2x2 Mixed Design ANOVA as in the initial IMC and EMG analyses. For all ANOVAs, we tested for normality with the Shapiro Wilk test and equality of variance with Levene’s test, where violated, Mann Whitney and Friedman’s test were used.

3. Results

3.1. *Participants*

Fourteen otherwise healthy participants (3 Females and 11 Males) with a mean age of 47 (STD17) were included. Skaters had 23 (STD11) years of skating experience. Onset of skater’s cramp was 40 (STD17) and symptom-duration was 7 (STD7) years. Eleven were impacted in their left leg and 3 in their right, with 62% quitting due to skater’s cramp. Fourteen controls were selected based on matching skating proficiency and dedication. Controls’ mean age was 36 (STD 16) with 22 (STD 10) years of skating experience (Appendix B.1).

3.1.1. *Experiment: Generic stationary activation task*

There was no difference in IMC between the impacted and non-impacted leg of affected skaters, and with the left and right legs of controls in the generic stationary activation task. Shapiro Wilk tests confirmed within subjects factors were not normally distributed in the stationary coherence task, therefore a non-parametric approach was employed. Mean theta-band IMC during stationary contraction in resisted and non-resisted conditions was the same for the control group vs the affected group (Table 1a), and the same between the impacted vs non-impacted leg of skaters with skater’s cramp, and the left vs the right leg of controls (Table 1b). The complete Mann Whitney and Friedman test output for all analysis can be seen in Table 1a, b.

3.1.2. *Experiment: In-skating task*

3.1.2.1. *Coherence while skating.*

There was a visual indicator of higher wavelet IMC while skating. Fig. 1 shows EMG activity and IMC in the

Table 1
Experiment: Generic stationary activation task.

a. Between Subjects:		
Comparing Groups:		
Affected vs. Control	Resisted	Non-Resisted
Impacted/Left, 10%MVC, PERO/SOL	$p = .18$	N/T
Impacted/Left, 10%MVC, TIB/GAS	$p = .93$	N/T
Impacted/Left, 20%MVC, PERO/SOL	$p = .98$	$p = .21$
Impacted/Left, 20%MVC TIB/GAS	$p = .84$	$p = .36$
Impacted/Left, 50%MVC, PERO/SOL	$p = .48$	$p = 1$
Impacted/Left, 50%MVC TIB/GAS	$p = .67$	$p = .96$
Non-Impacted/Right, 10%MVC PERO/SOL	$p = .63$	N/T
Non-Impacted/Right, 10%MVC, TIB/GAS	$p = .59$	N/T
Non-Impacted/Right, 20%MVC, PERO/SOL	$p = .13$	$p = .19$
Non-Impacted/Right, 20%MVC TIB/GAS	$p = .71$	$p = .23$
Non-Impacted/Right, 50%MVC, PERO/SOL	$p = .2$	$p = .36$
Non-Impacted/Right, 50%MVC TIB/GAS	$p = .75$	$p = .87$
b. Within Subjects:		
Comparing Legs:		
Impacted vs Non-Impacted and Left vs Right (controls)	Resisted	Non-Resisted
Impacted vs Non-Impacted Leg (affected participants at 10,20, 50%MVC)	$p = .68$	$p = .84$
Left vs Right leg (controls at 10,20, 50%MVC)	$p = .39$	$p = .81$

a: All results are p-values for a between subject Mann Whitney U analysis, testing the null-hypothesis that IMC is different in affected skaters. N/T: not tested. Impacted: impacted leg of affected group. Non-impacted: non-impacted leg of affected group. Left and Right: Left and right leg of control group. %MVC: Percentage of Maximum Voluntary Contraction.

b: All results are p-values for the within subjects non-parametric repeated measures Friedman analysis comparing impacted/non-impacted and left/right legs for the affected and control participants respectively.

impacted leg for TA and GAS in one affected participant and one control group participant. It illustrates an increase in IMC immediately prior to skate placement in the affected participant, corresponding with higher muscle activity at the moment of dystonic jerking.

Average wavelet IMC at skate placement was the same for the affected compared to control group (between subjects), F(1,22) = 0.14, p = .71, $\eta^2 = 0.001$, however IMC differed for the within subjects factor 'leg' where the impacted (left in controls) leg was higher compared to the non-impacted (right in controls) leg, F(1,26) = 4.23, p = .05, $\eta^2 = 0.031$. There was a marginal non-significant interaction effect between factor leg and subject group, F(1,26) = 3.29, p = .08, $\eta^2 = 0.024$, and in post hoc comparisons IMC in affected skaters' impacted leg appeared to be higher compared to their non-impacted leg, t(1,26) = 2.74, p_{tukeyi}=.05, Cohen's d = 0.66 (Table 2a). For null results: Appendix B.2.

3.1.2.2. Muscle activity while skating. Muscle activity was higher in the impacted vs non-impacted leg and compared to controls (Fig. 2b). Levene's test was violated for the impacted leg, F(1,26) = 6, p = .02, $\eta^2 = 0.14$, therefore to restore normality we transformed samples using the square-root. EMG was higher for the affected group at skate placement compared to controls (between subjects), F(1,26) = 5.8, p = .02, $\eta^2 = 0.14$. EMG was higher for factor leg, F(1,26) = 9, p = .006, $\eta^2 = 0.051$, with a significant interaction effect between factor leg and subject group, F(1,26) = 7.54, p = .043, $\eta^2 = 0.043$. Post hoc tests revealed EMG in the impacted leg was higher than the non-impacted leg and the left and right leg of controls, t(1,26) = 4.01, p_{tukey}=.002, Cohen's d = 0.96; t (1,26) = 3.39, p_{tukey}=.007, Cohen's d = 1.28; t(1,26) = 3.5 p_{tukey}=.006, Cohen's d = 1.32 (Table 2a).

3.1.2.3. Correlation between muscle activity and inter-muscular coherence while skating. Higher muscle activity predicted higher IMC in the impacted leg. Linear regression was used to predict the IMC from EMG scores. For affected skaters' impacted leg, EMG activity explained a significant amount of the variance in IMC at skate placement, F(1,13) = 9.86, $p = .009$, $R^2 = 0.45$, $R^2_{\text{adjusted}} = 0.41$. The regression coefficient (B = 1.6, 95% CI [0.49,2.71]) indicated a 0.1 increase in millivolts of EMG corresponded to an IMC increase of 0.16. There was no correlation in the non-impacted leg or the left/right of controls (Table 2c), or across the entire stroke cycle (Appendix B.2, B.3).

3.1.2.4. Subgroup analysis. Six participants showed EMG scores in their impacted leg above the 95th percentile of the median EMG scores in the control group (averaged across both legs) and were defined as a severely affected sub-group. IMC was higher in the affected sub-group generally, F(1,18) = 6.24, p = .02, $\eta^2 = 0.18$. For the within subjects factor 'leg', IMC was higher in the impacted (left in controls) leg compared to the non-impacted (right in controls) leg, F(1,18) = 5.49, p = .03, $\eta^2 = 0.06$. There was a significant interaction effect between factor leg and subject group, F(1,18) = 4.67, p = .04, $\eta^2 = 0.051$, and in post hoc comparisons, IMC in the impacted leg was higher compared to the controls' left t (1,18) = 3.26, p_{tukey}=.01, Cohen's d = 1.59, and right t(1,18) = 3.36, p_{tukey} = .01, Cohen's d = 1.64 leg, and marginally but not significantly higher in the impacted leg compared to the non-impacted leg t(1,18) = 2.69, p_{tukey} = .07, Cohen's d = 1.16. There was no difference between

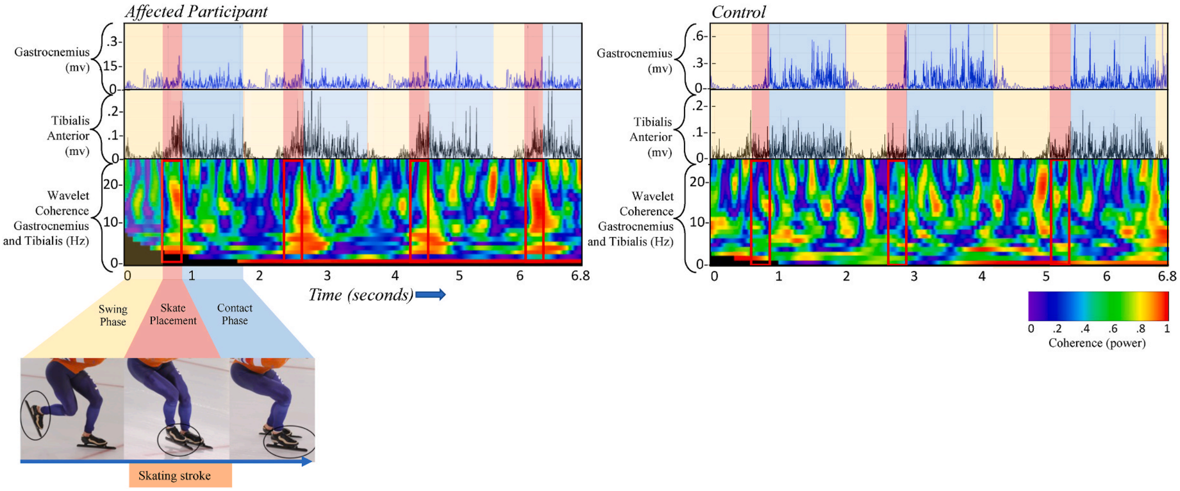

Fig. 1. Depicted is a graphical indication of higher IMC at skate placement in one affected skater. The two uppermost rows depict rectified EMG activity in two muscles during one straightaway section, and below it the resultant wavelet IMC for those two muscles. The vertical red color bands and corresponding red box represent the period of skate placement for repeated strokes.

Table 2
Experiment: In-skating task.

a. Mixed Design ANOVA n = 14 Affected/Controls	Coherence		EMG
Between Subjects: Group	$p = .71, \eta^2 = .001$		**$p = .02, \eta^2 = .14$**
Within Subjects: Leg	**$p = .05, \eta^2 = .031$**		**$p = .006, \eta^2 = .051$**
Interaction Effect: Leg/Group	$p = .08, \eta^2 = .024$		**$p = .04, \eta^2 = .043$**
Post-Hoc Tests	**Impacted vs Non-Impacted**	**$P_{tukey} = .05$, Cohen's d $= .66$**	**$P_{tukey} = .002$, Cohen's d $= .96$**
	Impacted vs Left	$P_{tukey} = .68$, Cohen's d $= .43$	**$P_{tukey} = .007$, Cohen's d $= 1.28$**
	Impacted vs Right	$P_{tukey} = .61$, Cohen's d $= .47$	**$P_{tukey} = .006$, Cohen's d $= 1.32$**
	Non-Impacted vs Right	$P_{tukey} = .95$, Cohen's d $= -.2$	$P_{tukey} = .78$, Cohen's d $= .36$
	Non-Impacted vs Left	$P_{tukey} = .92$, Cohen's d $= .24$	$P_{tukey} = .79$, Cohen's d $= -.35$
	Left vs Right	$P_{tukey} = .1$, Cohen's d $= .04$	$P_{tukey} = .1$, Cohen's d $= .002$

b. Mixed Design ANOVA Subgroup Analysis n = 6 Affected vs n = 14 Controls	Coherence	
Between Subjects: Group	**$p = .02, \eta^2 = .18$**	
Within Subjects: Leg	**$p = .03, \eta^2 = .06$**	
Interaction Effect: Leg/Group	**$p = .04, \eta^2 = .05$**	
Post-Hoc Tests	Impacted vs Non-Impacted	$P_{tukey} = .07$, Cohen's d $= 1.16$
	Impacted vs Left	**$P_{tukey} = .01$, Cohen's d $= 1.59$**
	Impacted vs Right	**$P_{tukey} = .01$, Cohen's d $= 1.64$**
	Non-Impacted vs Right	$P_{tukey} = .76$, Cohen's d $= .48$
	Non-Impacted vs Left	$P_{tukey} = .81$, Cohen's d $= -.43$
	Left vs Right	$P_{tukey} = .1$, Cohen's d $= .05$

c. Linear Regression	Coherence vs EMG
Impacted Leg	**$p = .009, R^2 = .45, R^2_{adjusted} = .41$**
Non-Impacted Leg	$p = .16, R^2 = .16, R^2_{adjusted} = .08$
Control Left Leg	$p = .80, R^2 = .006, R^2_{adjusted} = -.08$
Control Right Leg	$p = .61, R^2 = .023, R^2_{adjusted} = -.06$

a: Results are p-values and effect-size for 2x2 Mixed ANOVA for summary scores of IMC and EMG comparing between subjects factor group: affected skaters vs controls skaters, and within subject factor leg: impacted (left for controls) and non-impacted (right for controls) legs, and interaction effect: group*leg. Post-hoc observations: Impacted and Non-impacted are the legs of skaters with skater's cramp. Left and Right are the legs of controls.
b: We performed subgroup analysis of IMC selecting affected participants whose EMG scores in their impacted leg were higher than the 95th percentile of median EMG scores of the control group (averaging across both legs).
c: Depicted are results of a simple linear regression where we predicted IMC of skaters from their EMG scores. EMG activity explained a significant amount of the variance in IMC only in the impacted leg.

affected skaters' non-impacted leg and controls. For complete results consult Table 2b.

4. Discussion

4.1. Main findings

Our study aimed to measure if inter-muscular coherence was abnormally high in those affected with skater's cramp, a movement disorder previously proposed to be a TSD. Two experiments were performed: a stationary condition measuring muscle activation unrelated to speed skating, and a task-specific condition measuring IMC while speed skating. IMC was not higher in the generic stationary activation condition. We observed tentative evidence that while skating IMC was higher in the impacted leg of affected skaters compared to their non-impacted leg. Also, in more severe cases (measured by EMG), IMC was higher in the impacted leg compared to the right and left leg of controls. These results are tentative evidence that while skating, skater's cramp may have a centrally driven component which makes TSD a more likely diagnosis.

4.1.1. Experiment: In-skating task

Spectrographs qualitatively showed higher IMC in affected skaters in their impacted leg at the moment of skate placement (where symptoms of skater's cramp occur) [6] (Fig. 1). Quantitatively, IMC appeared higher in the impacted leg of skaters compared to their non-impacted leg (Table 2a). Contrary to our hypothesis, IMC was not higher in the affected group compared to the control group (Fig. 2a). Based on our findings that higher coherence correlated with disease severity (only in the impacted limb) (Table 2c), and the observed high variance in disease severity (as measured by EMG) (Fig. 2b), we posited a possible mild and diverse case-mix among affected skaters which may have influenced our results. Based on this, we performed a sub-group analysis of the most severely affected skaters that showed clear higher IMC compared to controls (Table 2b). We speculate disease severity was generally mild in our cohort because skater's cramp was observed to be quite dangerous and falls were common, leading us to postulate that severely affected skaters may be less common among the pool of potential candidates that took part in this study. Despite the ambiguity in the severity of skater's cramp in our sample, we interpret our results to be tentative evidence of higher IMC in the impacted leg of affected skaters compared to their non-impacted leg, and in more severely affected skaters compared to controls.

Research has shown IMC is higher in TSD due to subcortical structures that drive disinhibition via the primary motor cortex down the corticospinal-tract resulting in higher coherence inter-muscularly [15]. This process is independent of exertion, therefore IMC does not increase with higher muscle activity naturally [31,32], but does increase with maladaptive central drivers in dystonia in general [8], and in TSD in particular [15,33]. Therefore, the instances of higher IMC we observed, all of which occurred only at skate placement (where skater's cramp occurs) and only in the impacted leg, constitute the first tentative evidence of a centrally driven problem and possibly a TSD in skaters with skater's cramp. Importantly, this evidence remains tentative because IMC may not necessarily arise centrally but could also originate due to maladaptive oscillations within spinal reflex loops as observed in essential tremor [16]. Future studies distinguishing cortical and peripheral coherence in skater's cramp could help clarify this. Although our findings are preliminary, they suggest future research could use wavelet coherence to study inter-muscular, cortico-muscular and cortical coherence in skater's cramp and other TSDs.

4.1.2. Experiment: Generic stationary activation task

We found IMC was normal in skaters with skater's cramp in a generic stationary activation task, implying no centrally driven problems outside of their affected skating. Although not in line with our

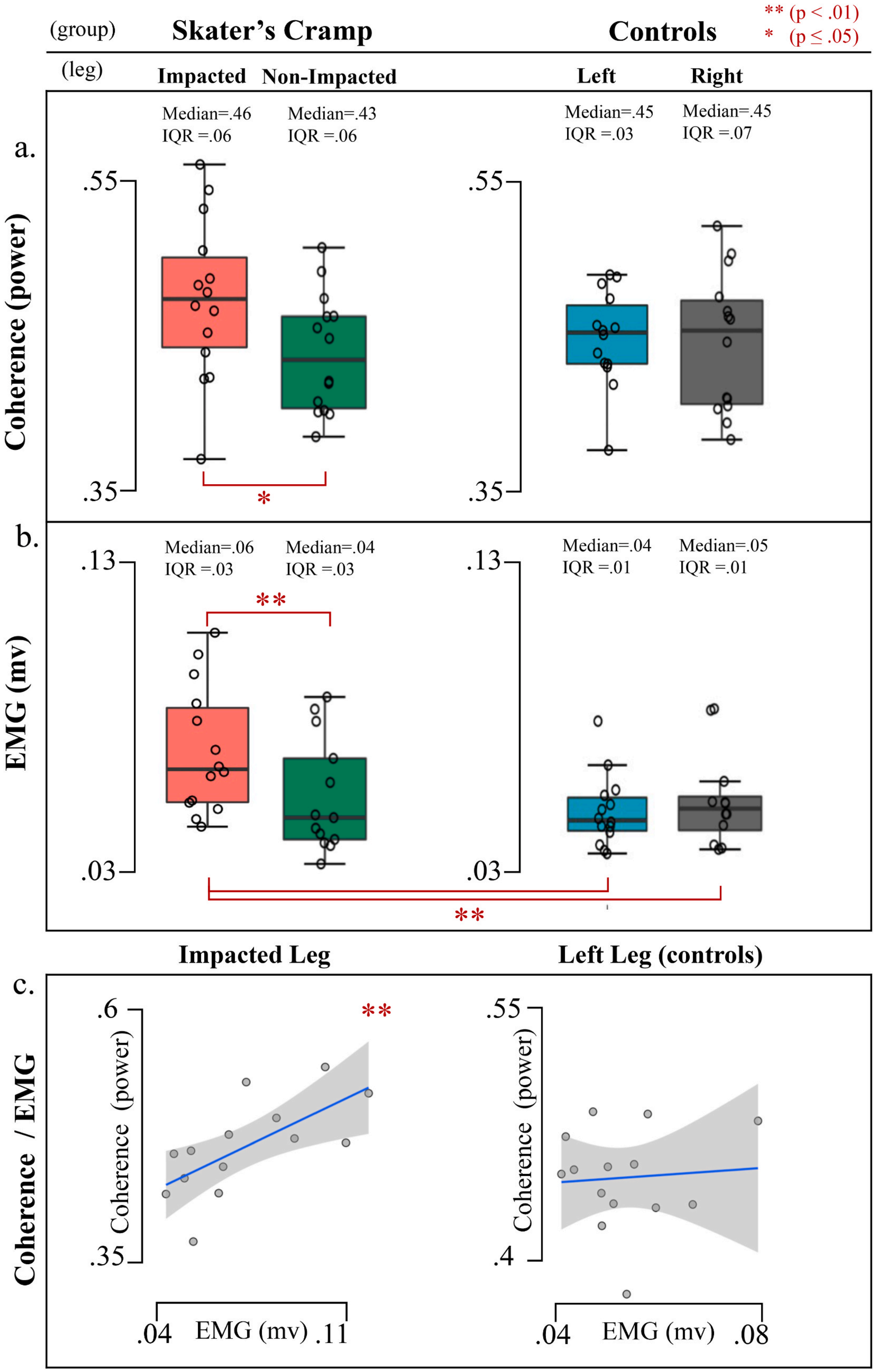

Fig. 2. Depicted is a comparison of the impacted and non-impacted leg of affected participants (left side) and the left and right leg of controls (rights side) for: a) IMC measured as power (top row), b) EMG activity in millivolts (middle row), and c) the correlation between IMC and EMG scores using simple linear regression (bottom row). A and b represent average muscle activity and average coherence from the pooled result of 4 muscles and 4 muscle combinations respectively. Plots show participants as dots. Box and whisker plots show median scores, where the box represents the first inter-quartile range and the whisker is the 90th percentile of the median. The blue line indicates best fit and gray area is the 95% confidence interval.

hypothesis, this finding is not surprising, as there is scarce and conflicting evidence that IMC is higher in TSD in a non-task-specific setting. Of the four studies looking at this in writer's cramp, only one, Choudhury et al., found a clear difference [15], while the other three did not [8,13,14]. One possible reason for the conflicting findings is that Choudhury et al. mimicked both the hand position of writing and simultaneously employed a motor-control task, unlike the other studies. Perhaps both position and motor-control of the impacted limb need to be sufficiently engaged to elicit a flawed descending drive. This may also explain our null result, as we were unable to completely replicate the position of the foot of a speed skater, nor provide participants with a motor-control task during exertion. Exact positioning may be an important factor in future IMC experiments. Studies adjusting hand positioning in pianists with musician's dystonia observed improvement to symptoms, suggesting exact posture is a requisite for TSD symptoms [34]. Therefore, even in generic tasks, testing IMC may require task-specific posture.

4.2. Limitations

The rarity of skater's cramp resulted in a small sample size of only 14 affected, and a sub-group of 6 severely affected, participants, reducing statistical robustness of group-wide comparisons. The apparent diversity of our case-mix and these small sample sizes are inevitable when studying rare movement disorders. Using higher EMG as a proxy for disease severity is an assumption that, though supported, requires more research. For the stationary experiment, finding a task that initiated co-activation of the distal leg equivalent to the forearm activity of the pinch-grip tasks in Choudhury et al. was challenging. It required a stationary device that could mimic simultaneous exo-rotation and plantar flexion of the foot at skate placement [6]. As no such device existed, we constructed an in-house solution to elicit co-activation of the requisite muscles. This remains a novel experimental solution, and future studies may consider a different approach. Surface EMG of the soleus is less accurate than fine-wire EMG (more susceptible to crosstalk [35]) due to its deeper position [36]. We consulted experts to optimize placement accuracy but it remains a limitation.

5. Conclusion

Previous research showed skater's cramp is a probable TSD [6]. Our study looked at inter-muscular coherence in skaters with skater's cramp as an indicator of a centrally driven problem, and evidence of a TSD. We measured skaters in a stationary resistance task unrelated to skating based on previous findings showing higher IMC in writer's cramp using a similar design [15]. Additionally, using an approach involving wavelet coherence (novel in TSD research) we measured skaters while speed skating (task-specifically). Results suggested higher IMC in the impacted leg of skaters compared to their non-impacted leg. Additionally, tentative evidence showed that the most severely affected skaters had higher IMC in their impacted leg while skating compared to controls. This was not the case across the entire cohort, possibly due to a diverse case-mix. We found no differences in IMC in a generic stationary activation task. Our results of higher IMC while skating is preliminary evidence that skater's cramp is centrally driven, making the chances it is a TSD higher.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.parkreldis.2022.105250

Author's roles

BN-designed the study, collected the data, analyzed the data, wrote the manuscript.

MT-designed the study, reviewed and critiqued the manuscript.

TZ-designed the study, reviewed and critiqued the manuscript.

EO-designed the study, reviewed and critiqued the manuscript.

JWE-designed the study, analyzed the data, reviewed and critiqued the manuscript.

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Declaration of competing interest

None.

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Appendix A

Methods

1. Excluding Beta-band

Beta-band coherence was not analyzed due to concomitant factors influencing its interpretation in our study. Age has been shown to reduce beta-band coherence [20,21]. The affected group was matched with controls based on experience and dedication to speed skating, but was older (47 STD17) compared to controls (35 STD16), therefore the possible effect of higher coherence due to skater's cramp may be canceled out by lower coherence due to increased age. Also physical exertion increases beta-band coherence [20,22] and higher motor-cortical demand decreases it [23]. Because skater's cramp poses natural challenges to both, it would be impossible to distinguish differences in coherence as an indicator of skater's cramp from differences as a cause of it.

2. Comparing Impacted/Non-impacted to Left/Right Legs of Controls

We compared the impacted and non-impacted leg of affected skaters to the right and left leg of controls. These groupings were based on the fact that all our experiments were bilaterally symmetrical, where we assumed the left and right leg would normally perform identically in the control group. This was true of our stationary task, but also of skating (where our analysis was confined to the straightaway). For this reason we compared 14 left and 14 right legs of controls against the pooled results of the impacted (11 left and 3 right) and non-impacted (3 left and 11 right) legs of affected skaters.

3. Calculating Inter-Muscular Coherence

Inter-muscular coherence analysis was performed. A zero phase high pass 4th order Butterworth filter was applied at 10Hz to all EMG activity. Auto and cross-spectra were estimated with the Welch method using 50% overlap on 1-s windows after applying a Hanning window with no spectral smoothing. Coherence significance-level was calculated using the Halliday et al. [24] method on the basis of the number of segments, using the formula: $\text{signlevel} = 1 - (0.05)^{1/(n-1)}$. The upper limit of segments in the coherence calculation was $n = 50$, where $n = 45$ led to a significance limit 0.0658 (adjusting for overlapping segments). Average coherence scores were calculated in the theta-band: 3–7Hz band based on previous research [15] for agonist-antagonist muscle combinations: PERO/SOL and TA/GAS.

4. Calculating Wavelet Coherence

We calculated wavelet coherence by taking selected EMG segments and converting them to zero mean signals by mean subtraction, rectifying and filtering them at 10Hz with a high-pass 4th order zero phase-shift filter. We used a Complex Morlet Wavelet [37] with FS ratio 4 (containing 4 complete cycles of the scale under analysis) as the mother wavelet. We calculated wavelet coefficients every 25 ms. The wavelet spectrum was 50 bins with an upper limit of the frequency analysis at 50Hz. We calculated coherence values at each time-point based on previous methods [38]. We performed time domain smoothing as a weighted moving-average function, where weight was defined by a Gaussian function and width was equal to wavelet size in the time-domain. Frequency domain smoothing used a boxcar filter with width equaling the scale decorrelation length. Coherence significance limit was based on Monte Carlo simulations based on two uniform white noise time series as applied in previous research [27] These simulations resulted in a significance threshold for this study at 0.425.

5. Calculating Muscle Activity

We calculated the magnitude of muscle activity while skating by creating time-normalized (interpolation using piecewise cubic hermite interpolation ‘pchip’) stroke cycles using SkateView where skate placement was at the center (50%) of every completed stroke cycle (100%). We first used SkateView to organize time points into activity blocks of off-ice and on-ice activity that resembled the stance and swing phase in walking. We then demarcated strokes from take-off to subsequent take-off, by time normalizing the swing phase and contact phase separately and concatenating them, so that skate placement (heel strike in walking) was at 50% of every full (100%) stroke. EMG filtering used a Journée filter [28] with low/high band-pass of 10 and 50Hz. Only straightaway strides were examined, as it allowed direct comparison of impacted and non-impacted leg in affected participants, because straightaway strides in skating are bilaterally symmetrical in terms of propulsion. We calculated time-normalized stroke cycles for four muscles: PERO, TA, SOL, and GAS. We then calculated average EMG activity at two different time windows within these stroke cycles:

1) Skate placement: between 35% and 50% of normalized stroke cycle.
2) Entire Stroke: between 0% and 100% of normalized stroke cycle.

Appendix B

Results

1. Participant Characteristics (Affected)

Sub.	Sex	Age	SE	AAO	Duration (years)	Quit S	Impacted Leg
1	M	45	23	34	10	yes	R
2	M	73	32	62	2	yes	R
3	M	52	11	51	2	no	L
4	F	19	8	17	1	yes	L
5	M	61	22	53	4	yes	L
6	M	68	42	64	0	no	R
7	M	59	44	53	4	no	L
8	F	41	17	32	9	yes	L
9	M	52	19	46	7	no	L
10	F	52	21	45	6	yes	L
11	M	48	26	45	2	no	L
12	M	19	13	17	1	yes	L
13	M	48	30	19	28	yes	L
14	M	24	10	20	2	yes	L
Mean		47	23	40	6	62%quit	
SD		17	11	17			

F: female, M: male, SE: skating experience, AAO: age at onset, L: left, R: right, Quit S: quit skating.

2. In-Skating Task Full Stroke

a. Mixed Design ANOVA n = 14 affected/controls		Coherence	EMG
Average over Entire Stroke Cycle.	Between Subjects: Group	$p = .13$, $\eta^2 = .08$	$p = .15$, $\eta^2 = .046$
	Within Subjects: Leg	$p = .63$, $\eta^2 < .001$	$p = .42$, $\eta^2 = .009$
	Interaction Effect: Leg/Group	$p = .63$, $\eta^2 < .001$	$p = .21$, $\eta^2 = .023$

b. Linear Regression	Coherence vs EMG	
Average over Entire Stroke Cycle.	Impacted Leg	$p = .75$, $R^2 = .008$, $R^2_{\text{adjusted}} = -.074$
	Non-Impacted Leg	$p = .38$, $R^2 = .067$, $R^2_{\text{adjusted}} = -.01$
	Control Left Leg	$p = .54$, $R^2 = .03$, $R^2_{\text{adjusted}} = -.049$
	Control Right Leg	$p = .27$, $R^2 = .10$, $R^2_{\text{adjusted}} = .02$

a: Depicted are non-significant p-values for 2x2 Mixed ANOVA for summary scores of IMC and EMG across the whole stroke cycle. Between subjects factor group: affected skaters vs controls skaters, within subject factor leg: impacted (left for controls) vs non-impacted (right for controls), and interaction effect: group*leg. Post-hoc observations: Impacted and Non-impacted: skaters with skater's cramp, Left and Right: controls.
b: Depicted: simple linear regression predicting IMC of skaters from EMG over the whole stroke cycle.

3. Regression Analysis

EMG activity did not explain a significant amount of the variance in IMC at skate placement for the control group of skaters for their left leg, $F(1,13) = 0.07$, $p = .80$, $R^2 = 0.006$, $R^2_{\text{adjusted}} = -0.08$, regression coefficient (B=.17, 95% CI[.-1.3, 1.65]), or their right leg, $F(1,13) = 0.28$, $p = .61$, $R^2 =$ ^{0}.023, $R^2_{\text{adjusted}} =$ -0.06, regression coefficient (B=.41, 95% CI[.-1.3, 2.12]).

Across the whole skating stroke muscle activity scores did not explain a significant portion of the variance for the affected group or the control group in either their left or right leg. For IMC of the affected group in the impacted leg: $F(1,13) = 0.58$, $p = .75$, $R^2 = 0.008$, $R^2_{\text{adjusted}} = -0.074$, regression coefficient (B=-0.11, 95% CI[.-0.89, 0.66]), non-impacted leg, $F(1,13) = 0.84$, $p = .38$, $R^2 = 0.067$, $R^2_{\text{adjusted}} = -0.01$, regression coefficient (B=.3, 95% CI[.-0.41, 1.01]). And for the control group, Left leg: $F(1,13) = 0.4$, $p = .54$, $R^2 = 0.03$, $R^2_{\text{adjusted}} = -0.049$, regression coefficient (B=.-0.23, 95% CI[.-1.02, 0.57]), and right leg, $F(1,13) = 1.34$, $p = .27$, $R^2 = 0.10$, $R^2_{\text{adjusted}} = 0.02$, regression coefficient (B=-0.52, 95% CI[.-1.49, 0.46]).

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