Is Skater's Cramp a Task-Specific Dystonia?

Nijenhuis, Beorn

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Muscular and Kinematic Features in Speed Skaters Indicate a Task-Specific Dystonia

Beorn Nijenhuis[12*], MSc., Marina A.J. Tijssen[1], MD PhD, Tim van Zutphen[2,], PhD, Jan Willem Elting[1], MD PhD, Aron H.P. Schalkwijk[1], MD, Egbert Otten[3], PhD, Jeroen van der Eb[4], PhD

[1] University Groningen, University Medical Center Groningen, Department of Neurology, NL-9700 RB Groningen, The Netherlands.
[2] University of Groningen / Faculty Campus Fryslân, Wirdumerdijk 34, 8911 CE Leeuwarden, The Netherlands.
[3] University Groningen, Department of Movement Sciences, NL-9713 AV Groningen, The Netherlands.
[4] Leiden Institute of Advanced Computer Science (LIACS), P.O. Box 9512, 2300 RA Leiden, The Netherlands.

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Abstract

Objective

Skater's cramp is a movement disorder in speed skaters. We investigated whether affected skaters matched the disease profile of task-specific dystonia, specifically whether there was evidence of maladaptive muscle activity occurring simultaneously with aberrant movements (jerking). We further examined different skating intensities, positing no change would be more indicative of task-specific dystonia.

Methods

We analyzed video, kinematic and muscle activity in 14 affected skaters. We measured the angular velocity and electromyographic activity of normalized speed skating strokes using one dimensional statistical non-parametric mapping. Skaters were matched with comparably skilled controls, and filled out a bespoke clinical questionnaire.

Results

Skaters' impacted leg showed over-activation in the peroneus longus, tibialis anterior and gastrocnemius that coincided with higher foot movement compared to their healthy leg and controls. This pattern persisted regardless of skating intensity. Clinical features indicated it was task-specific and painless with common trigger factors including stress, equipment change, and falling.

Conclusion

We showed aberrant muscular and kinematic activity in a movement disorder in speed skaters indicative of task-specific dystonia.

Significance

Understanding skater's cramp as a task-specific dystonia could reduce the damage that misdiagnosis and unsuccessful invasive operations have caused. Our quantitative method has value in testing future treatment efficacy.

1. Introduction

Dystonia is characterized by "sustained or intermittent muscle contractions causing abnormal, often repetitive, movements, postures" (1). Task-specific dystonia (TSD) only occurs during the performance of a complex skill in an isolated body part, rarely generalizing beyond highly related movements (2). TSD has been noted in manually complex occupations like painting and watchmaking (2,3), is quite common in musicians (4) and is prevalent in many sports such as running and golf (5). In all cases, fine motor coordination is compromised and muscular over-activity, jerking and a general loss of motor control is common. Clinically, features of TSD include an insidious onset over a course of weeks or months (6), as well as triggering factors such as stress and equipment change in the period before TSD onset (7,8). The exact pathophysiology of TSD is not known, however it is thought to be partly caused by corrupted motor engrams that arise due to arduous over-practicing (4). Physiologically this often appears as dysregulation of cortical and subcortical networks that drive fine motor control through inhibition (9–12). The resultant corrupted engrams affect a subsection of a skilled movement resulting in patterned and stereotypical jerking movements, muscular over-activation and co-activation at a specific moment while executing the skill (4,6,13,14).

Skater's cramp is a debilitating movement disorder affecting dedicated amateurs and Olympic level speed skaters alike, often after many years of healthy skating. Developing skater's cramp is dangerous, as the loss of motor control can lead to falling and injury, and has caused many, including former Olympians, to quit the sport (15). It was first described as a sudden inversion or eversion of a speed skater's ankle joint, just before skate placement (15). Attempts at treating skater's cramp based on it being a peripheral issue have been unsuccessful, and surgeries resulting from suggested diagnoses such as compartment syndrome and arterial occlusion have led to needless additional risk for affected skaters (16,17). In an attempt to better understand skater's cramp and improve treatment outcomes the clinical diagnosis of task-specific dystonia was proposed by a group of neurologists specializing in movement disorders, in 2014 (16). Importantly, TSD can only be diagnosed clinically, as there is no quantitative means of diagnosis. Despite this, a growing body of research has linked the condition to key muscle and movement features whose presence can support the diagnosis of TSD. These include 1) a task-specific repetitive jerking of a limb during a complex movement combined with 2) muscular over-activation (6).

The first of these two features was identified in a case control study investigating if skater's cramp was a TSD. Five affected skaters and five controls underwent video and kinematic analysis employing inertial measurement unit (IMU) sensors and found a patterned, active and stereotypical jerking of the impacted foot (17). However, missing from this

experiment was measurement of muscle activity, therefore this method could not show whether the aberrant movement resulted from muscular over-activation, another key symptom of TSD. For example, in runners, researchers combined EMG and movement analysis assuming that a combination of repetitive patterned muscle and movement activity needed to be present simultaneously to suggest TSD as a diagnosis (14). We chose this same approach in our current study, and therefore combined EMG and kinematics to investigate our first major research question: 1) Does the involuntary movement in skater's cramp coincide with muscular over-activity in the lower leg at the moment of skate placement?

Furthermore, we tested whether the problem was persistent irrespective of skating intensity, assuming that differences dependent on intensity would be more indicative of a peripheral problem then a central one. We posited this based on evidence that after a peripheral change, (such as muscle damage or neuropathy) muscular and kinematic patterns of a flawed movement can change depending on the intensity of its execution (18,19). For example, if skater's cramp were a mechanical instability eliciting compensatory stabilization in the impacted limb, increased intensity/speed may result in higher activation to maintain control. Conversely, if the problem is a TSD over-activation is less likely to be influenced by increased intensity, as has been observed in golfers and runners (5,20,21). It is thought this is partly because the motor engrams dysregulated in TSD are so complex as to scale to a wide range of different intensities (7). Therefore for our second research question we asked: 2) Does muscular over-activity remain constant despite different skating intensities?

We aimed to answer these questions for a larger cohort of skaters with skater's cramp (15), comparing them with a control group of healthy skaters matched for skating skill-level. We hypothesized affected skaters would show consistent muscular over-activation in their impacted leg, and that this pattern would occur irrespective of skating intensity.

2. Methods

2.1 Population

Participants were recruited by publication on a popular Dutch speedskating website. Based on 50 initial responses, 15 participants were selected (convenience sampling) and an oral interview was conducted to ascertain whether they fulfilled the inclusion criteria. Inclusion criteria were: more than ten years of speed skating experience with a former practice frequency of minimum twice per week and at least 5 years of unaffected skating before symptom onset. Furthermore, the abnormal movements had to occur only during skating and needed to be described by the participant as an active patterned jerking of the lower limb that occurred at skate placement. Participants were asked to fill out a bespoke questionnaire before they

began their participation in the rest of the experiment. It was based on a previous case-study questionnaire investigating demographic and clinical information on skater's cramp (table 1) (17). They were further submitted to a physical exam by a neurologist (MT and AS). Due to Covid-19 restrictions, 3 of the 15 affected participants could not be examined but reported no further neurological complications. Exclusion criteria: participants who exhibited other neurological disorders during the medical examination and skaters who reported sprains, tendonitis or other current injuries. 15 controls were recruited from the same skating clubs as affected participants through contacting coaches and skaters individually. Controls were matched based on experience (years of skating) and dedication (hours of skating per week). We performed an independent samples t-test to compare experience and dedication with the affected group. Mann Whitney was used when normality (tested with the Shapiro Wilk test) or equality variance (tested with Levene's test) were violated. Controls were further excluded if they suffered from any injuries reported in the questionnaire. The study was reviewed by the Medical Ethical Committee of the University Medical Hospital of Groningen (UMCG) ruled IRB approval was not necessary for this study (M119.241754). Informed consent was obtained from all participants.

2.2 Experiment

2.2.1 Skating Exercise Design

Skaters skated two sets of two laps and two sets of four laps on a 400 meter ice track at 60% and 80% of their approximate maximum speed. Skaters subjectively self-rated their intensity, which was additionally measured in both groups with lap-times calculated using video footage from go-pro recordings. These two intensities were chosen so as to ensure a speed sufficient for normal and natural skating movements, while not incurring overly high levels of exhaustion before data collection was complete. High speed video recordings of all participants were collected from behind skaters at a fixed distance using a head mounted GoPro video camera (GoPro Hero7, 1440p, 120fps). The GoPro was fixed to the head of a high level speed skater (BN) and outcome variables consisted of a qualitative analysis of skaters' movements.

2.2.2 EMG Stroke Cycle Classification

Skaters were fitted with two Inertial Measurement Units (IMUs) (Shimmer3 shimmersensing.com) fixed to the toe-end of their skates to be referred to as "skate sensors"(22), running a bespoke software package, SkateView (23), designed to correctly capture and classify speed skating strokes without the need for prior positional calibration. SkateView detected when a skate landed on the ice, (skate placement), and the moment it lifted off the ice, (take off), forming time-blocks of an off-ice swing phase and an on-ice contact phase (similar to stance and swing phases in a walking gait-cycle). Time-synchronized to these skate sensors, 8 channels of surface EMG sampling at 512Hz (Shimmer3 EMG) were placed on the following leg muscles: peroneus longus, tibialis anterior, gastrocnemius (medial side), soleus, rectus femoris, gluteus medius, semitendinosus and adductor longus. We

chose these muscles as they had the highest likelihood of being involved in the skating movement. The 512Hz sampling rate was chosen to ensure optimal syncing between the 8 Shimmer3 sensors during data collection. EMG stroke cycles were time normalized with SkateView from take-off to take-off by concatenating the individually time normalized swing phase and contact phase so that skate-placement (comparable to heel strike in a walking gait-cycle) was at the center (50%) of every complete (100%) normalized time-block. For a detailed depiction of our method see figure 1a. EMG data was rectified and then filtered using a Journée filter (24) employing a band-pass filter between 10 and 50 Hz (a form of envelope filter). The high-pass filter (10Hz) was required due to the dynamic acceleration and deceleration of speed skating movements. To control for the inter-participant variance in the amplitude of surface-EMG signals caused by external factors (skin conductance etc.), all statistical analyses were performed on y-normalized strokes by taking each individual stroke and dividing it by the average of that stroke (y-axis normalization). We chose this course because the presence of extra activity was interesting *relative* to the rest of the cycle and compared to the healthy leg.

2.3 Statistical Analysis

2.3.1 Does the involuntary movement in skater's cramp coincide with muscular over-activity?

We initially qualitatively compared video footage (see Video, Supplemental Skater's Cramp Example Video 1, 2, 3) with EMG and kinematic data from time normalized strokes (figure 1). To assess how skater's cramp presented on a group level, we employed one dimensional statistical non-parametric mapping (SnPM) (25) (see Appendix A for details) using *MATLAB 2018a (MATLAB, MathWorks, Natick, USA)* to compare differences between the impacted and the healthy leg in skater's cramp, and the left and the right leg in control participants. Using a non-parametric paired samples t-test (SnPM{t}) employing permutation (25–27), we individually compared the normalized muscle activity in 8 muscles in one leg, with the correspondent 8 muscles in the other leg. Additionally we compared the angular velocity of the two IMU sensors (skate sensors) that were attached to the skater's feet. We used only the angular velocity values around the z rotational axis of the sensor (capturing inversion/eversion of the skater's foot) . To account for individual differences in inversion/eversion, the absolute value of the angular rotation in degrees per second (|deg/sec|) was compared for the impacted and non-impacted leg in the region of interest (ROI) of skate placement (40%-50% of the completed gait cycle [figure 1a]) using one dimensional statistical parametric mapping (SPM) (25). We chose this ROI to capture the period pre-skate placement. Because the skating movement is bilaterally symmetrical in the straightaway, we tested the null hypothesis of no difference between legs in muscle and movement activity.

2.3.2 Does muscular over-activity remain constant despite different skating intensities?
To test whether intensity of skating at 60% and 80% made any differences to skater's cramp we performed an additional SnPM analysis in the form of a two way repeated measures ANOVA (SnPM{F}) (Appendix A). The dependent variable was EMG activity and the two within subject factors (independent variables) were leg: left vs right leg, and intensity: 60% vs 80%. We focused specifically on the swing phase, ignoring any differences in intensity during the contact phase, as these would be expected in healthy individuals due to differences in pushing intensity between the 60% and 80%. In addition to identification of any differences between 60% and 80% in the swing phase, we also looked for possible interaction effects to evaluate if differences in intensity were dependent on whether a leg was cramping.

3. Results

3.1 Population

We included 15 affected subjects (12 Male ;3 Female) with skater's cramp. One participant was excluded from the affected group due to inconsistencies in responses to the bespoke questionnaire. The participant initially reported task-specificity and a patterned active jerking in the inclusion interview, but later reported task-generality and bilateral instability when filling out the questionnaire, and was therefore omitted from our analysis. The remaining 14 participants had a mean age of 47 (STD 17) (see table 1) and had 21 (STD 9) years of skating experience. Mean age of onset for skater's cramp was 40 (STD 17). Subjects 2, 5 and 10 were excluded from the neurological examination due to health concerns related to the onset of the Covid-19 pandemic. There were no peripheral neurological abnormalities reported in the remaining subjects. Four affected subjects reported pre-existing medical conditions, including a heart defect, knee operation, broken ankle and depression. Controls had a mean age of 36 (STD 16) and 22 (STD 10) years of skating experience. Control participants were not age matched, as they were selected based on experience and level and dedication. Experience in years of skating was the same between affected and control skaters (Mean affected±SD: 23±.11 vs Mean control±SD: 24±12), $t(1,25) = -1.15$, $p=.76$, as was hours of weekly practice (Mean affected±SD: 5±3 vs Mean control±SD: 7±6), $W = 68.5$ $p=.62$.

3.2 Clinical Findings

Symptoms presented only while skating and unilaterally. The rate of symptom onset was insidious, reaching full severity within weeks or months (median: 9 weeks). Subsequently, the condition stabilized, with 12 subjects reporting no remission after onset, even following many years of skating. Subject 3 and subject 13 reported a remission in symptoms, however this was temporary. The persistence of symptoms resulted in 9 subjects quitting skating, and no full recoveries. All subjects reported the condition was painless. Trigger factors were varied, but fell into 3 major groups: high stress, a change in equipment or a fall.

Table 1. Clinical Features

Sub.	Sex	Age	AAO	ROO (weeks)	Duration (years)	Rem	QS	Pain	Leg	TS	Triggering Factors	PMC	NE
1	M	45	34	Sudden	10	0	1	0	R	1	Change in training partner. Focus on new Technique	Heart Defect, (stenosis).	H
2	M	73	62	Sudden	2	0	1	0	R	1	Nothing Reported	0	NA
3	M	52	51	DNR	2	1 temporary	0	0	L	1	Equipment(new shoes)	0	H
4	F	19	17	DNR	1	0	1	0	L	1	Equipment (new blades)	0	H
5	M	61	53	26	4	0	1	0	L	1	Fall (skating)	0	NA
6	M	68	64	Sudden	0	0	0	0	R	1	Stress (general)	0	H
7	M	59	53	Sudden	4	0	0	0	L	1	Broken Ankle	0	H
8	F	41	32	13	9	0	1	0	L	1	Fall during skating	0	H
9	M	52	46	20	7	0	0	0	L	1	Stress (general)	0	H
10	F	52	45	2	6	0	1	0	L	1	Nothing Reported	Meniscus Operation	NA
11	M	48	45	104	2	0	0	0	L	1	Stress at work	Lower lumbar neuropathy (age 12)	H
12	M	19	17	DNR	1	0	1	0	L	1	Equipment: new skates.	Broken ankle (age 14)	H
13	M	48	19	10	28	1 temporary	1	0	L	1	Intensive Training Period	Depression	H
14	M	24	20	9	2	0	1	0	L	1	Equipment: new skates.	0	H
Mean		47	40	17 weeks	6								
SD		17	17	30									

Sub: Subject, NA: not available, F: female, M: male, AAO: age at onset, ROO: rate of onset, Rem.: Remission, QS: Quit Skating, L: left, R: right, TS: Task Specific, PMC : pre-existing medical condition, NE: Neurological Examination, H: healthy, C: complications.

Figure 1: Muscle and Movement Activity of One Affected Skater

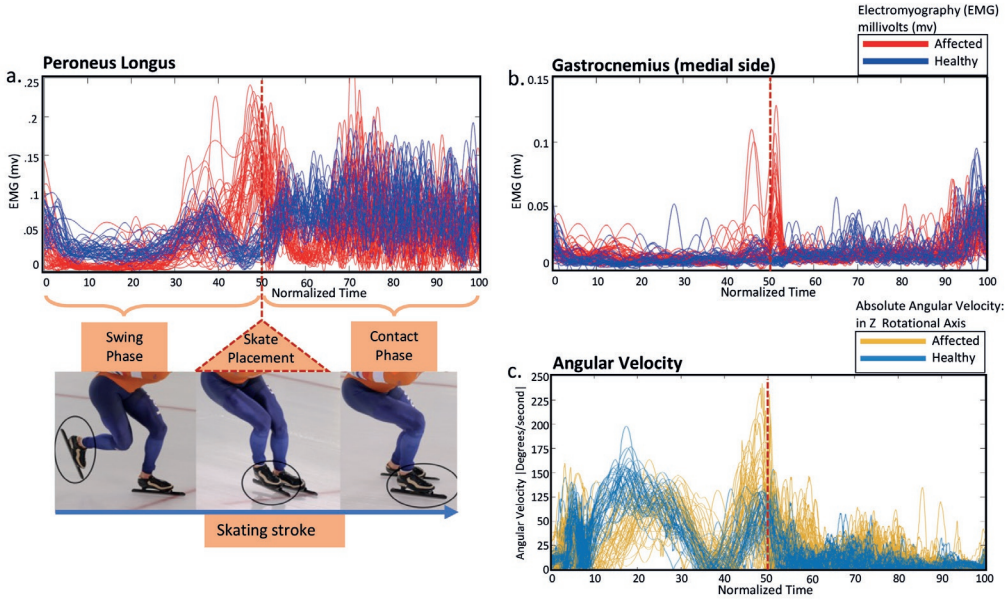

Figure 1a,b: Time normalized electromyography (EMG) of one affected participant over multiple strokes of the straightaway. In orange is shown a visual guide to one skating stroke. Figure 1c shows absolute angular velocity of one affected participant over multiple strokes. As skating is symmetrical in the straightaway, the impacted (red/gold) and healthy (dark and light blue) legs can be directly compared.

3.3 Statistical Analysis of EMG/Kinematics

3.3.1 Does the involuntary movement in skater's cramp coincide with muscular over-activity?

There was evidence of consistent muscular over-activity and maladaptive movements occurring only at skate placement. This was apparent in qualitative analysis of video of an affected skater (see Video, Supplemental Skater's Cramp Example Video 1, 2, 3, which demonstrates skater's cramp), and comparing it with EMG and kinematic analysis (figure 1). Additionally, across the entire affected group, muscles of the impacted lower limb showed higher relative activity compared to the corresponding muscles of the contralateral non-impacted limb only during the swing phase and only at skate placement (figure 2). When comparing group wide muscle activity between the legs in a paired samples t-test using SnPM{t}, the impacted peroneus longus, tibialis anterior and gastrocnemius showed higher activity than contralateral non-impacted muscles, crossing the critical threshold {t} = 4.051, 4.205 and 4.012, forming a suprathreshold cluster with a likelihood of being reproduced in repeated random samplings of p=.002, p=.001 and p=.003 respectively (figure 2). The deviations in these three muscles occurred at 42.1-48.9%, 45.1-50%, 48.5-52% of the normalized stroke cycle respectively. This moment of higher muscle activity corresponded with a moment of higher absolute angular velocity at 49%-50% of the

normalized stroke cycle, where the impacted foot crossed the critical threshold: {t} = 3.311, forming a suprathreshold cluster of p=.048 (figure 2). All these findings corresponded with video analysis, showing jerking that had a consistent pattern and moment of onset (see Video, Supplemental Skater's Cramp Example Video 1, 2, 3, which demonstrates skater's cramp). In sum, kinematic and muscular deviation in the normalized gait cycle occurred simultaneously, and both appeared to occur only during the moment where skater's cramp presented visually. There were no differences between legs in muscle activity or movement activity, and no visual indication of any problems in the control group.

On average there was one instance of under-activation later in the stroke cycle, where the gastrocnemius crossed the critical threshold {t} = 3.991, creating a suprathreshold cluster, p=.003 at the moment equating to 92% of the completed stroke cycle. Despite this lower activation, there were no visual indications of instability and no kinematic differences at that moment.

3.3.2 Does muscular over-activity remain constant despite different skating intensities?

Between 60% and 80% intensity, lap times decreased by 4.6 (SD 3.7) seconds and 5.4 (SD 3.4) seconds per lap for the affected and control group respectively. Over-activity remained consistent at different intensities. There was no difference in muscle activity in the skater's cramp group at 60% vs 80% skating intensity using a two way Repeated Measures ANOVA. In figure 3b the result for the within subjects factor intensity (60% vs 80%) is depicted and shows there was no crossing of the significance threshold {t} = 15.9 for this factor, indicating no difference. Figure 3c shows that despite there being no difference in intensity, the peroneus of the impacted leg was over-active relative to the healthy leg, showing a crossing of the significance threshold {t} = 15.6 at time period 42%-48%, supra-threshold cluster: p=.0004. Figure 3d shows there was no interaction effect between intensity and leg with no crossing of the threshold {t} = 16.6, indicating intensity did not differ based on whether the leg was impacted or non-impacted . Results of this analysis were the same for the gastrocnemius and tibialis muscles (Appendix B, Figure B.1).

Figure 2

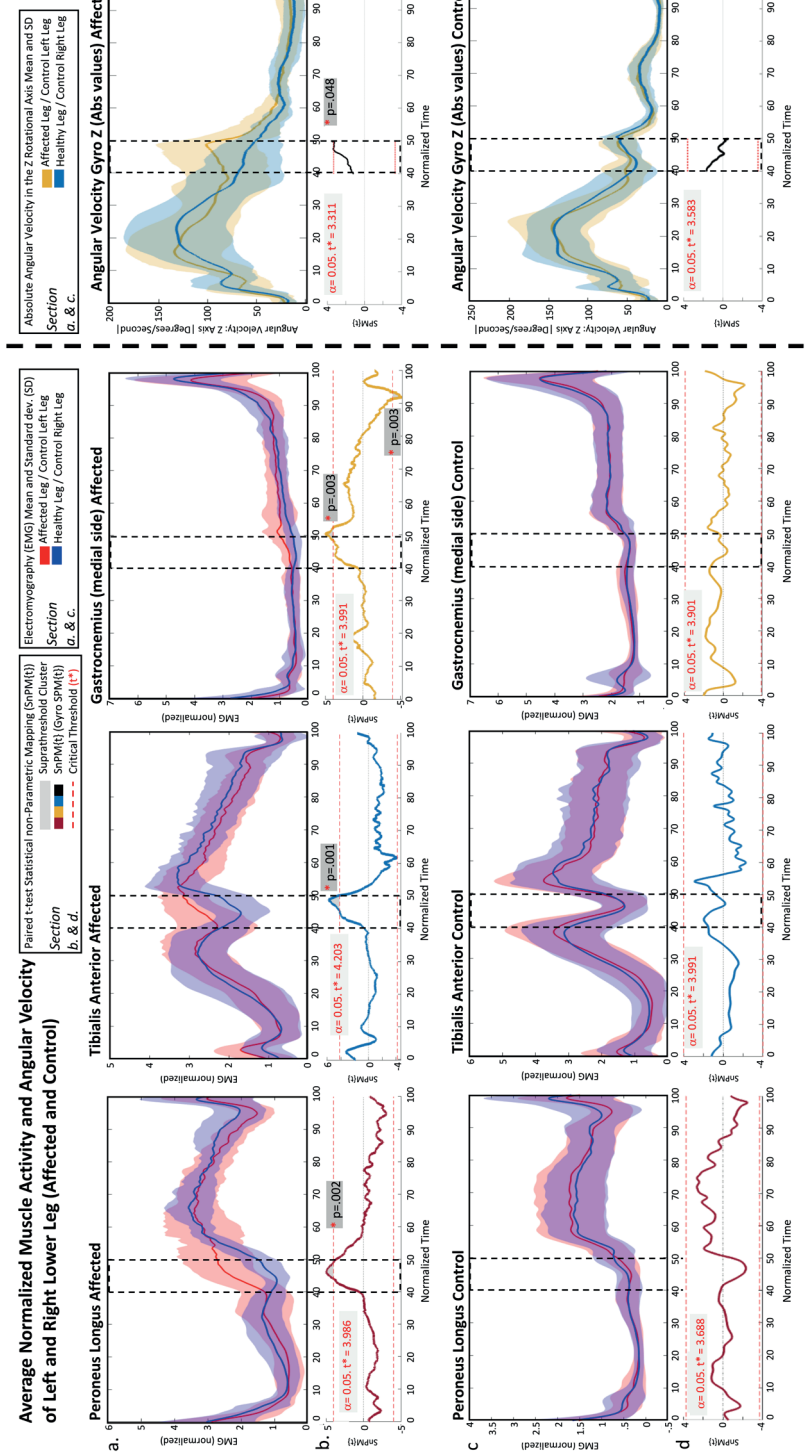

Figure 2a,c: Muscle and movement activity for the affected (above) and control (below) group. The dashed rectangle indicates the 10% of normalized time preceding skate placement. Figure 2b,d: In statistical non parametric mapping (SnPM) paired samples t-test average relative muscle activity in the peroneus longus, tibialis anterior and medial side of the gastrocnemius are higher in the impacted leg at approximately 40-50% of completed gait cycle. Higher absolute angular velocity is shown at approximately 49-50%. Probability (p) values in grey with red star indicate probability that the adjacent suprathreshold clusters was reproduced in repeated random samplings.

Figure 3

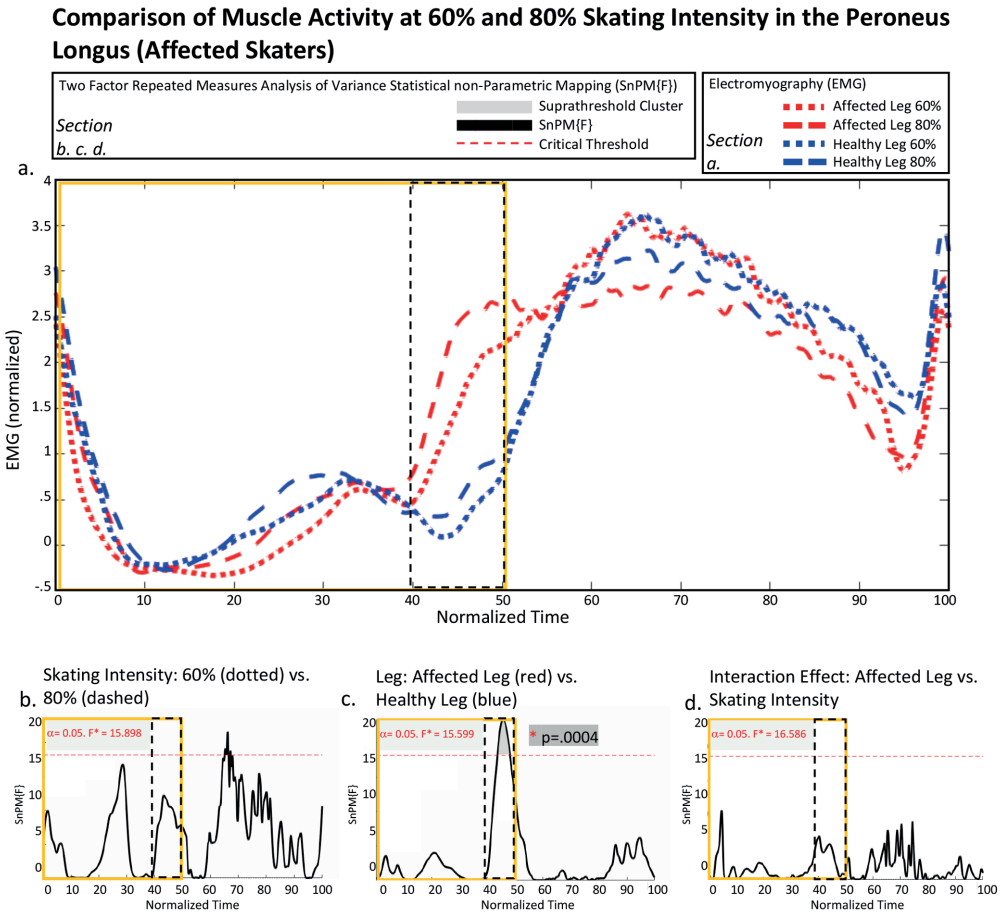

Figure3a: Visual depiction of the factors of the two-way repeated measures analysis of variance (ANOVA) showing within subjects factor leg: Mean relative muscle activity in the impacted leg (red) compared to the non-impacted leg (blue); and within subjects factor intensity: 60% skating intensity (dotted lines) and 80% skating intensity (dashed lines). The gold square indicates the area of the swing phase.

Figure 3b: Results for within subjects factor intensity: muscle activity at 60% or 80% skating intensity showing no difference.

Figure 3c: Results for within subjects factor leg showed the relative activity of the impacted leg was higher than the healthy leg nearing the end of the swing phase as indicated by a supra-threshold cluster at 43-49% of stroke-cycle exceeding the critical threshold 15.599, p=.0004.

Figure 3d: No interaction effect between factor intensity and leg, indicating there was no difference in skating intensity dependent on whether the leg was affected or healthy.

4. Discussion

Our study's results support our hypothesis that skater's cramp is a TSD. As with other forms of lower limb TSD (14,28), angular velocity showed a repetitive stereotyped jerking of the foot that occurred simultaneously with over-activity in muscles of the impacted leg (figure 1), and was task-specific (affected skaters reported the problem did not occur outside of speedskating). Video evidence supported our quantitative findings, showing the same active repetitive jerking symptoms occurred in all affected skaters (video 1, 2, 3), although the severity of jerking appeared to vary per participant. Furthermore, intensity of skating did not influence maladaptive over-activation of the impacted muscles (figure 3), similar to other forms of TSD (20). Finally, the clinical history of the skaters in our study was compatible with TSD (6,29,30) with the rate of symptom onset progressing insidiously over weeks or months, and triggering factors such as stress, equipment change or a fall coinciding with symptom onset (table 1).

In a retrospective case review of task-specific lower limb dystonia, 13 runners and seven other athletes including cycling, dancing and speed walking showed inversion of the foot and maladaptive muscle activation to be among the most suggestive features of TSD (28). Similarly, in a case study of TSD in runners, researchers detected kinematic and EMG evidence of an aberrant repetitive and consistent jerk (14). These studies reported problems arose after many years of healthy performance and often without peripheral injury or pre-existing conditions. Our observations found affected skaters matched these muscular, kinematic and clinical features. We found stereotyped muscular over-activation in the peroneus longus, tibialis anterior and gastrocnemius, combined with patterned jerking (figure 2) that arose suddenly after many years of healthy skating with no neurological issues reported and most skaters reporting no pre-existing medical conditions (table 1). Thus we can conclude muscular, kinematic and clinical features of skater's cramp together form a set of observations that mirror many other forms of TSD. Importantly the combination of these features together is highly elucidative. For example, co-contraction (caused by fear as a result of instability) may produce a muscular over-activation, but would likely not produce an extra movement that was so highly consistent as seen in skater's cramp. As TSD is still clinically diagnosed, our findings cannot unequivocally confirm our hypothesis, even as they provide compelling evidence for it.

A recent model has been proposed to explain certain key features of TSD that matches well with our observations of skater's cramp (7). In it, the primary cause of TSD is suggested to be a sudden peripheral change triggering "sensory motor reorganization" (7) precipitating in the rapid development of task-specific over-activation (8,31–33). This is thought to occur when intense repetition causes the formation of longer sequences of encoded motor engrams whose efficiency causes the loss of intermediate-level representations (34). With

the loss comes less ability to adapt to sudden environmental changes (equipment change or an injury) leading to engram corruption triggering dis-inhibition in motor networks that results in patterned, consistent and task-specific cramping (35). Because corruption may be triggered by myriad factors, TSD can arise without injury, suddenly, after many years of healthy performance and remains persistent and unresponsive to peripheral interventions (4). Within the context of this proposed model, skater's cramp matched these features, as we found it to be a task-specific over-activity in healthy skaters (figure 2), with a sudden unpredictable onset that was persistent (table 1), and unresponsive to attempted interventions including physio guided stretching, dry needling, and surgery for compartment syndrome.

Interestingly, there was also under-activation, with the impacted gastrocnemius showing less activity at 92% of the completed stroke cycle in skaters on average (figure 2). This moment was unrelated to the jerking movement that indicated skater's cramp at skate placement. Higher levels of exhaustion in the impacted leg may explain this finding, as it is known to reduce EMG output. Supporting this suggestion, under-activity occurred at the highest period of relative muscular output in the cycle. Further research is required controlling for fatigue to confirm this. An alternative explanation is y-normalization effects from our signal processing, where the elevated muscle activity at the jerking moment leads to lower relative muscle activity in other parts of the cycle (appearing as under-activation). The visual indicators of lower activity in the peroneus longus and tibialis both before and after skate placement suggests this.

Skaters' cramping remained consistent regardless of whether skaters skated at 60% or 80% intensity (figure 3b). These findings agree with a previous study of skater's cramp where extra weight was placed on a skater's foot, hypothesizing this would cause a higher swing amplitude if the limb were modeled as a torsion spring (36). Similar to these findings, we proposed that increased intensity would have caused higher muscle activation to maintain control if skater's cramp were a mechanical instability. We posit that it was due to the centrally driven nature of skater's cramp that it was not impacted by the added mass or higher intensity. This consistency is typical of TSD, where flawed movements often maintain a persistent pattern despite being performed in differing external circumstances(1). It is thought this is because, despite being highly task-specific, these engrams can likely remain active during a wide range of different limb speeds and masses (37,38). Researchers posit that if these engrams are corrupted, it would result in movement flaws that persist within a wide range of intensities (6). Therefore, we suggest that the consistency in intensity shown in skater's cramp is likely an indicator of a similar centrally driven problem, making task-specific dystonia a more likely etiology. Admittedly, these findings are preliminary and meant to support an approach to differentiate movement issues, in particular those

that are centrally driven. Future research may consider employing a battery of tests using weights and differences in intensity to aid in the diagnosis of new forms of TSD.

4.1 Limitations

Quantitative evidence-based protocols for identifying TSD do not exist, therefore our attempt at diagnosis was based on the best current understanding of TSD, and employing new ways of measuring it with kinematics and EMG. As in other studies, we identified clear kinematic cues of dystonia using accelerometry and related it to EMG measures, however we did not correct for electro-mechanical delay (the early onset of muscle activation compared to force or motion), because the time disparity had no bearing on the strength of our major findings. Furthermore, surface EMG is susceptible to variance in amplitude due to the accuracy of sensor placement, differences in morphology and skin conductance, etc. For these reasons our findings remain suggestive, but not conclusive. A more detailed analysis with sub-dermal EMG looking at individual participants may be required.

4.2 Conclusion

This is the first substantial cohort tested attempting to diagnose skater's cramp as a TSD, a condition that has previously often been misdiagnosed and improperly treated using invasive surgeries. Clinical, kinematic and EMG features support the diagnosis: TSD. The method developed in this study should be used in a future within subjects design to test the efficacy of TSD related interventions to attempt to improve symptoms in skaters affected by skater's cramp.

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Appendices

Appendix A
Statistical *non* Parametric Mapping vs Parametric Mapping.

A statistical non parametric map (SPM) is comprised of a scalar output statistic, SPM{t}, that is calculated separately for each time node in the time-series. In the case of our research, a paired samples t-test was used to find the magnitude of Left leg - Right leg differences for each node in the time series, within skaters for the control group and the affected group in 8 different muscles. However, this magnitude is not, itself, a means of hypothesis testing. For this the SPM1d software package usually calculates the critical threshold where only alpha (a) (5%) of smooth random curves may be expected to traverse, for our purposes to be referred to as *tcrit*. To ensure no biases based on different numbers of strokecycles per-participant, all SnPM tests were performed on the average of all stroke cycles for both legs per participant; which was a strategy successfully employed in other SPM biomechanics research. Due to the data from certain muscles being non-normally distributed, a non-parametric test based on permutation was employed instead of the parametric model. The most aberrant muscle: the Peroneus Longus was normally distributed, and therefore a Gaussian test was permitted in this case, however the soleus, tibialis anterior and gastrocnemius were not, and therefore a non-parametric model was adopted for all muscles.

Importantly, in this non-parametric model, *tcrit* constitutes the major difference in how SPM and S-*non-parametric*-PM (SnPM) are calculated. In a normal parametric test, *tcrit* is produced based on estimates of trajectory smoothness using temporal gradients (1), and based upon this smoothness, random field theory (RFT) expectations of the field-wide maximum (2). In SPM *tcrit* is calculated as the value correspondent to the equation (3) :

Eq. (A.1) $$P(t(q)_{max} > t_{crit}) = 1 - \exp\left(-\int_{t_{crit}}^{\infty} f(x)\,dx - ED\right) = \alpha$$

Here, $P(t(q)max > tcrit)$ designates the probability of the maximum value of t being higher than *tcrit*; $f(x)$ designates the statistical map; a is the level of significance, and *ED* refers to the Euler density function (4). In this model RFT assumptions allow for the calculation of *tcrit* , dependent on it being smooth "n-dimensional Gaussian" (5).

In contrast to this, SnPM uses a permutation technique (6) to arrive at *tcrit* (3). The technique essentially creates a probability density function directly from the input data by randomly permuting n number of times over that data, where maximum n leads to the greatest accuracy (at the cost of computational power and calculation time). Every permuted statistical map is recalculated so the maximum value (for paried SnPM{t} the max t value) is saved. For SnPM *tcrit* is then computed by solving (3):

Eq. (A.2) $$P(permuted\ t_{max} > t_{crit}) = \frac{number\ of\ permuted\ t\ values\ \geq\ t_{crit}}{number\ of\ permutations} = \alpha$$

Here *permuted tmax* is the max value of {t} in the permuted empirical distribution. a is the chosen significance level (such as .05). This approach has been shown recently to provide comparable results with the Gaussian parametric model in biomechanical applications as long as sample sizes are not to small (7); at n=15 for both affected and controls, our n was sufficiently large to employ SnPM{t} as a paired samples t-test.

Values of SnPM{t} surpassing the critical threshold indicated that the EMG time-series (of the left and the right leg) were significantly different. Because of waveform-smoothness, and inter-dependence of neighboring points, many adjacent points along the SPM{t} curve will exceed the critical threshold together as a group, and this is referred to as a supra-threshold cluster (7) Unlike the calculation of *tcrit,* SPM and SnPM both use RFT to calculate the supra-threshold cluster. 1dSPM uses RFT expectations on supra-threshold cluster size to calculate clustered p-values indicating the probability that these clusters could have arisen from a random field of the same temporal smoothness(3).

For all SnPM{t} tests, we performed the maximum number of permutations to ensure maximum accuracy (<33 000). Furthermore, we ran the parametric alternative in parallel to all non-parametric tests in muscle groups where normality was not violated such as the peroneus longus. In these analyses there were no meaningful difference in amplitude or location of suprathreshold clustering, thereby supporting our assumption that the non-parametric tests were robust and confirming previous assumptions about SnPM/SPM equivalence (3).

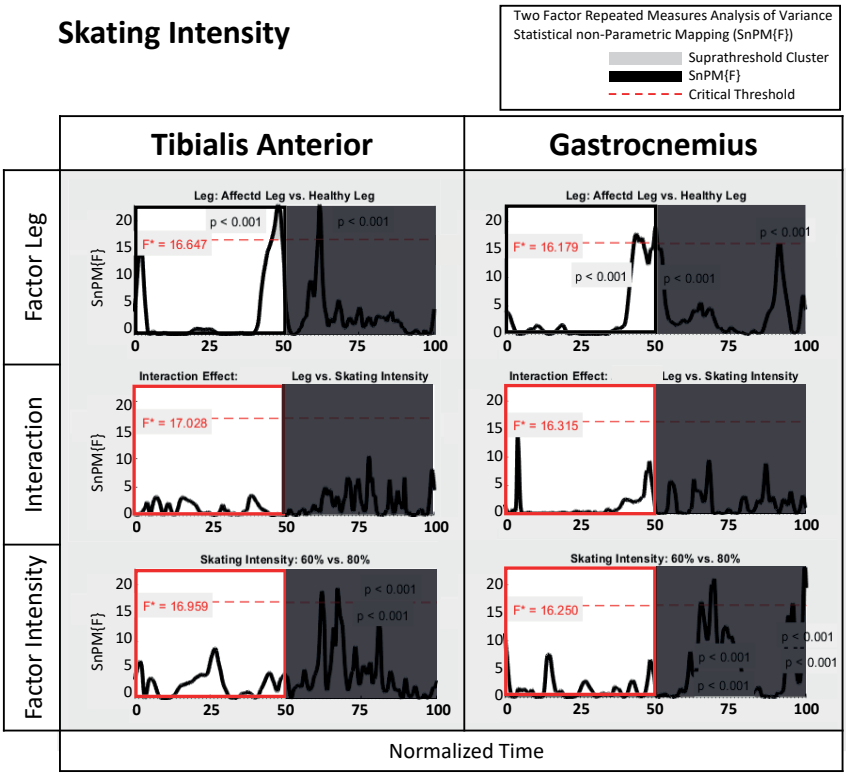
Additionally we performed an SnPM{F} test, or a 2x2 repeated measures ANOVA with two within subjects factors. We compared skating intensity at 60% and 80%, to which leg, either non-impacted or impacted, using a non-parametric 2x2 repeated measures ANOVA design. Here the scalar output statistic forming the statistical parametric map was S*n*PM{F}, where any suprathreshold clusters indicating a difference between 60%/80% skating intensity, or an interaction effect between intensity and which leg, would suggest that intensity played a role in Skater's Cramp. As the maximum number of permutations in such a model is more than a conventional computer can process, we followed the guidelines of set out in previous research: 100 000 permutations.

Appendix B
Consistency in Cramping

In the supplemental figure (Fig. B.1) is depicted the results of a SnPM 2X2 Repeated Measures ANOVA that looked at muscle activity of the impacted and non-impacted leg at 60% and 80% skating intensity. In figure 3b of the main text, results showed there is no difference in activity based on intensity for the peroneus longus. This analysis looked only at the swing phase, because differences in intensity in the push phase are unimportant when looking at skaters cramp, as they are expected at different speeds/intensities in healthy skaters (higher speed will inevitably equal harder pushing and hither muscle activity).

Below in the supplemental figure (Fig. B.1) is the results showing the same pattern of activity in two additional muscles of the impacted lower leg. There was no differences in activity between 60% and 80% skating intensity, even though there are differences in activity between impacted and non-impacted leg irrespective of intensity. Furthermore, there is no interaction effect between which leg and intensity.

Fig. B.1



Video Legends

Videos						
Title	Author	Video-grapher	Participant	Length (Seconds)	Size (MBs)	Summary of Content
Skater's Cramp Example Video 1.mp4	Beorn Nijenhuis	Beorn Nijenhuis	Participant 1	7	.71	Video depicts affected skater showing an exhortation of their impacted left skate as it nears the ice after a completed stroke.
Skater's Cramp Example Video 2.mp4	Beorn Nijenhuis	Beorn Nijenhuis	Participant 2	9	9	Video depicts affected skater showing an exhortation of their impacted right skate as it nears the ice after a completed stroke.
Skater's Cramp Example Video 3.mp4	Beorn Nijenhuis	Beorn Nijenhuis	Participant 3	6	5.4	Video depicts affected skater showing an exhortation of their impacted left skate as it nears the ice after a completed stroke.

Videos: Supplemental videos are available at https://doi.org/10.1016/j.clinph.2023.02.168

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