

## BRIEF COMMUNICATION

## Relationship Between Physical Activity, Tic Severity and Quality of Life in Children with Tourette Syndrome

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### Abstract

**Objective:** To examine the relationship between physical activity, tic severity and quality of life (QoL) in children and adolescents with persistent tic disorder and Tourette Syndrome. **Method:** Baseline data was examined from a larger randomized controlled trial (Clinicaltrials.gov NCT02153463). Physical activity was assessed via pedometers with daily step count recorded. Tic severity (assessed via Yale Global Tic Severity Scale or YGTSS) and QoL (assessed via PEDs QL 4.0) were compared between those more physically active ( $\geq 12,000$  steps/day) and less physically active ( $< 12,000$  steps/day). **Results:** Thirteen children participated; four had  $\geq 12,000$  steps/day and nine had  $< 12,000$  steps/day. The active group had a lower total tic severity ( $p = 0.02$ ), and total YGTSS score ( $p = 0.01$ ). The vocal tic severity score was lower in the active group ( $p = 0.02$ ). Motor tic severity was not different amongst the two groups. For Peds QL scores, the active group performed better in physical functioning ( $p = 0.01$ ), social functioning ( $p = 0.03$ ), school functioning ( $p = 0.02$ ), psychosocial functioning ( $p = 0.03$ ) and total PEDs QL score ( $p = 0.01$ ). **Conclusions:** Higher physical activity levels are associated with lower vocal tic severity and improved aspects of quality of life. Further research is needed to determine the utility of physical activity as therapy for tics.

**Key Words:** tics, Tourette Syndrome, physical activity, quality of life

### Résumé

**Objectif:** Examiner la relation entre l'activité physique, la gravité des tics et la qualité de vie (QdV) des enfants et adolescents souffrant d'un trouble de tics persistants et du syndrome de Tourette. **Méthode:** Les données de base ont été examinées d'après un essai randomisé contrôlé plus vaste (Clinicaltrials.gov NCT02153463). L'activité physique a été évaluée par des podomètres enregistrant le compte de pas quotidien. La gravité des tics (évaluée par l'échelle globale de gravité des tics de Yale ou la YGTSS) et la QdV (évaluée par l'inventaire de la qualité de vie des jeunes adultes, PEDs QL 4.0) ont été comparées entre les enfants plus actifs physiquement ( $\geq 12\ 000$  pas/jour) et les enfants moins actifs physiquement ( $< 12\ 000$  pas/jour). **Résultats:** Treize enfants ont participé; 4 avaient  $\geq 12\ 000$  pas/jour et 9 avaient  $< 12\ 000$  pas/jour. Le groupe actif avait un total plus faible de gravité des tics ( $p = 0,02$ ), et un score total moindre à la YGTSS ( $p = 0,01$ ). Le score à la gravité des tics sonores était plus faible dans le groupe actif ( $p = 0,02$ ). La gravité des tics moteurs n'était pas différente chez les 2 groupes. Pour les scores à la Peds QL, le groupe actif a eu un meilleur rendement au fonctionnement physique ( $p = 0,01$ ), au fonctionnement social ( $p = 0,03$ ), au fonctionnement scolaire ( $p = 0,02$ ), au fonctionnement psychosocial ( $p = 0,03$ ) et au score total de la PEDs QL ( $p = 0,01$ ). **Conclusions:** Des niveaux supérieurs d'activité physique sont associés à une gravité plus faible des tics sonores et à des aspects améliorés de la qualité de vie. Il faut plus de recherche pour déterminer l'utilité de l'activité physique comme thérapie pour les tics.

**Mots clés:** tics, syndrome de Tourette, activité physique, qualité de vie

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### Introduction

Gilles de la Tourette's syndrome (TS) is characterized by recurrent motor and vocal tics for at least one year (American Psychiatric Association, 2013) with a prevalence of 0.77% in children aged five to 18 years (Knight et al., 2012). Persistent (chronic) tic disorder (PTD) is classified as having either motor or vocal tics for over one year (American Psychiatric Association, 2013). Tics fluctuate in type, anatomical location, complexity, frequency and intensity, with peak severity occurring between ages 8-12. As such, it is during childhood and adolescence that the most significant impacts from the disease occur. Quality of life (QoL) of young people with tics is lower than that of healthy controls and tic severity predicts lower quality of life (Storch et al., 2007). Studies have shown a negative social perception toward patients with tics (Boudjouk, Woods, Miltenberger, & Long, 2000) and that tic severity is inversely related to social acceptability (Boudjouk et al., 2000). It is increasingly recognized that tic variability can be attributed to contextual factors, whether environmental or intrinsic, which can act as antecedents or consequences to tics (Conelea & Woods, 2008).

There is conflicting evidence regarding the relationship between physical activity and tics. O'Connor et al (O'Connor, Brisebois, Brault, Robillard, & Loiselle, 2003) had adults with TS keep a daily diary and found that engagement in physical activity was identified as a situation where these patients were less likely to have tics emerge. Jacome (Jacome, 1987) published a case of a patient whose tics worsened with medium distance jogging. Nixon et al (Nixon, Glazebrook, Hollis, & Jackson, 2014) showed a reduced rate of tics following an exercise session, suggesting that acute exercise may have an attenuating effect on tics. Additionally, case reports (Chang, Liu, Yu, & Lee, 2012; Wang, Kuo, & Stern, n.d.) have demonstrated improvements in TS symptomatology after exercise or after a period of sustained practice of activities requiring reflexive responses. Thus, there is reason to believe that physical activity may have effects on patients with TS; the purpose of this study was to examine the relationship between physical activity level, tic severity and quality of life.

### Methods

#### Study Design

We examined baseline data of children with TS and Persistent Tic Disorders collected from a larger pilot randomized controlled trial (Clinicaltrials.gov NCT02153463). Baseline physical activity was assessed via pedometers worn for seven days with daily step count recorded. Each participant was given a Yamax Digi-Walker SW-200 pedometer; a small, lightweight (16 grams), device that counts the number of steps taken. Daily total step count, wear times

and reasons for pedometer removal were recorded on a log sheet.

#### Participants

Eligible participants for the study were identified by AD, the neurologist responsible for caring for the majority of patients with tic disorders and TS at the Children's Hospital of Eastern Ontario (CHEO) in Ottawa, Canada. Inclusion criteria for participants were: a) age 8-16; b) diagnosis of TS or PTD as per DSM V (American Psychiatric Association, 2013); c) ambulatory; d) able to understand English or French; e) function at minimum Grade three level; f) able complete questionnaires; and, g) no changes to tic/psychotropic medications for four weeks prior to and for the duration of the study. Exclusion criteria were participants who: a) cannot ambulate independently; and, b) are not permitted to participate in physical education class.

#### Measures

A patient questionnaire was used to collect demographic information (including age, gender and family structure). Participants were assessed on baseline tic severity via the Yale Global Tic Severity Scale (YGTSS) and on quality of life via the PEDs QL 4.0. The Yale Global Tic Severity Scale (YGTSS) is a clinician rated measure that begins with the completion of a checklist of all tics present in the past week. Current motor and vocal tics are then rated on five dimensions (number, frequency, intensity, complexity, and interference; range, 0-5 each), which are summed to yield separate motor and vocal tic scores (range, 0-25) and a combined total tic score (range, 0-50). An associated impairment scale (range 0-50) assesses tic related disability during the past week. The Pediatric Quality of Life Inventory (PedsQL 4.0) is a 20-item measure that assesses QoL and also provides domain scores for physical, emotional, social and school functioning. The PedsQL has established validity and reliability for youths with and without chronic health conditions (Varni, Seid, & Rode, 1999).

#### Analyses

Tic severity and quality of life was compared between those more physically active (mean of  $\geq 12,000$  steps/day) and less physically active ( $< 12,000$  steps/day) via the Mann-Whitney U-Test. 12,000 steps per day was chosen as this number has been shown to be equivalent to 60 minutes per day of moderate to vigorous physical activity. Sixty minutes of moderate-to-vigorous physical activity is the Canadian (and international) guideline for the amount of physical activity children need to perform in order to optimize health outcomes related to physical activity (Colley, Janssen, & Tremblay, 2012).

Fisher's exact tests were performed to analyze categorical variables; linear by linear associations were performed for ordinal variables; and the Mann-Whitney U test was

performed for continuous variables. For Peds QL scores, Psychosocial Functioning Scales were generated from the average of the Emotional, Social, and School Functioning Scales; and the Total PEDs QL Scales were generated from the average of the Physical, Emotional, Social and School Functioning Scales.

The Children's Hospital of Eastern Ontario Research Ethics Board approved this research.

## Results

Thirteen children (five female) participated in this study (Table 1). Participants were divided into a higher activity group (HAG) ( $\geq 12,000$  steps,  $N = 4$ ) and lower activity group (LAG) ( $< 12,000$  steps,  $N = 9$ ). Of note, the four participants in the HAG were all male. There were no significant differences amongst both groups across all demographic variables.

Table 2 details the baseline tic scores. The mean global tic severity score was 31.5 (SD: 13.1, range = 10 – 58) indicating moderate tic severity in the group as a whole. Mann-Whitney U test results showed that the vocal tic severity score was lower in the HAG (M: 1.5, SD: 2.4 in  $> 12,000$  steps group vs. M: 9.2, SD: 4.3 in  $< 12,000$  steps group;  $p = 0.02$ ). Additionally, total tic severity (M: 11.8, SD: 1.7 vs. M: 21.4, SD: 6.9;  $p = 0.02$ ), and total YGTSS score (M: 19.3, SD: 6.3 vs. M: 37.0, SD: 11.5;  $p=0.01$ ) were lower in the HAG. Motor tic severity was not significantly different amongst the groups. Since the LAG had more students in Grade nine and higher, the analyses were repeated with high school students excluded and the association between motor tic severity score and physical activity level remained non-significant ( $p=0.91$ ). For vocal tic severity, the association remained statistically significant ( $p=0.02$ ), while for total tic severity score, the association was no longer significant ( $p=0.06$ ).

Table 3 outlines the PEDs QL scores. When comparing the HAG ( $> 12,000$  steps) versus the LAG ( $< 12,000$  steps), the active group performed better in the physical functioning, social, school, psychosocial and total QL scores. Emotional functioning in both groups did not vary based on baseline physical activity.

## Discussion

Our results show that total tic severity and total YGTSS was lower in the HAG; this finding was mainly driven by the lower vocal tic severity score. Although motor tic scores were also lower in the HAG, this result was not statistically significant. It is possible that children with vocal tics may participate less in physical activity due to the stigma associated with vocal tics, combined with the fact that physical activity often involves social interaction (i.e. team sports). Studies with medications such as aripiprazole and

guanfacine (Pringsheim et al., 2012) have shown specific improvement in vocal tics and not motor tics; however the reasons for this remain unclear. It is also possible that an insufficient sample size in our study contributed to the lack of a significant difference in motor scores.

Because causation cannot be determined in a cross-sectional study such as ours, it is unclear if patients who engage in more physical activity are those who have fewer tics, or if the physical activity itself directly causes a reduction in tics. Nixon et al have theorized three possible explanations for why tics may show improvement with exercise (Nixon et al., 2014). First, it is possible that tic reduction may occur via a transient activation of executive control circuits through exercise. Second, exercise functions as a distraction task, taking attention away from the tics, leading to tic reduction due to the execution of an alternative purposeful movement. Third, exercise may act as a competing motor response to tics.

With respect to QoL, our trends indicate that students who are physically active at baseline have better physical performance and are better able to get along with their peers. Additionally, these students also tend to perform better at school, with higher psychosocial function. Emotional functioning scores were greater in the HAG, but this result was not statistically significant. Similar to our study, previous research has shown improvements in QoL with physical activity (Bailey, 2006). It is theorized that this improvement may be due to a number of factors, including the process of actually engaging in physical activity (e.g. increased social interaction from group sports participation or time spent outdoors), improved self-esteem (e.g. positive perceptions of self) or biologic factors (increased endorphin levels as a result of the activity) (Anokye, Trueman, Green, Pavey, & Taylor, 2012).

As mentioned, limitations of our study include its cross-sectional nature, as well its small sample size, the latter of which may have contributed to the lack of a significant difference in motor tic severity between the two groups. The high proportion of female participants may make generalization of results more difficult, considering the fact that Tourette Syndrome and tics are seen more commonly in males. Our study also did not examine co-morbidities (i.e. ADHD, anxiety) and their relationship to tics and activity; examination of these relationships would be important in future studies. Despite these limitations, our results suggest the possibility of future longitudinal studies for patients with tics and Tourette Syndrome, where amounts of physical activity could be adjusted in a controlled fashion, to determine if exercise may be a viable intervention for tics.

Table 1. Demographics of study participants.

Demographic Variables	< 12,000 steps (N=9) n (%)	$\geq 12,000$ steps (N=4) n (%)	p-value*
<b>Gender</b>			
a) Male	4 (44.4)	4 (100.0)	0.11 <sup>1</sup>
b) Female	5 (55.6)	0 (0.0)	
<b>Number of siblings [Median (min, max)]</b>	1 (0, 4)	2 (1, 5)	0.20 <sup>3</sup>
<b>Number of persons in household [Median (min, max)]</b>	4 (2, 7)	5 (4, 6)	0.24 <sup>3</sup>
<b>Current grade in school</b>			0.80 <sup>2</sup>
c) Grade 2	2 (22.2)	0 (0.0)	
d) Grade 3	2 (22.2)	1 (25.0)	
e) Grade 5	1 (11.1)	1 (25.0)	
f) Grade 6	0 (0.0)	1 (25.0)	
g) Grade 8	0 (0.0)	1 (25.0)	
h) Grade 9	2 (22.2)	0 (0.0)	
i) Grade 10	1 (11.1)	0 (0.0)	
j) Grade 11	1 (11.1)	0 (0.0)	
<b>Does your child attend:</b>			0.16 <sup>2</sup>
k) All regular classes	5 (55.6)	4 (100.0)	
l) Regular classes with special help	3 (33.3)	0 (0.0)	
m) All special classes	1 (11.1)	0 (0.0)	
<b>Primary Language spoken by primary caregiver at home</b>			0.37 <sup>1</sup>
n) English	7 (77.8)	3 (75.0)	
o) French	2 (22.2)	0 (0.0)	
p) Other	0 (0.0)	1 (25.0)	
<b>Relationship to child</b>			0.31 <sup>1</sup>
q) Mother	9 (100.0)	3 (75.0)	
r) Father	0 (0.0)	1 (25.0)	
<b>Took part in sports outside of physical education classes that involve adult coaching in the past year</b>			0.50 <sup>1</sup>
s) No	3 (33.3)	0 (0.0)	
t) Yes	6 (66.7)	4 (100.0)	

\*Fisher's Exact Tests were performed for the categorical variables; Linear by Linear Associations were performed for the ordinal variables; Mann-Whitney U tests were performed for continuous variables.

<sup>1</sup>Fisher's Exact Test

<sup>2</sup>Linear by Linear Association

<sup>3</sup>Mann-Whitney U Test

Table 2. YGTSS scores and physical activity level

Variables	Mean (SD)	Median (25%, 75%)	p-value*
<b>Total Motor Severity Score</b>			
>12,000 steps	10.3 (3.0)	10.0 (0.5, 11.0)	0.33
< 12,000 steps	12.2 (5.9)	14.0 (8.5, 17.5)	
<b>Total Vocal Tic Severity Score</b>			
>12,000 steps	1.5 (2.4)	0.5 (0.0, 4.0)	<b>0.02</b>
< 12,000 steps	9.2 (4.3)	10.0 (7.5, 11.0)	
<b>Total Tic Severity Score</b>			
>12,000 steps	11.8 (1.7)	11.5 (10.3, 13.5)	<b>0.02</b>
< 12,000 steps	21.4 (6.9)	22.0 (15.5, 26.5)	
<b>Total YGTSS Score</b>			
>12,000 steps	19.3 (6.3)	21.5 (12.8, 33.5)	<b>0.01</b>
< 12,000 steps	37.0 (11.5)	33.0 (29.0, 47.0)	

Table 3. Peds QL scores and physical activity level

Variables	Mean (SD)	Median (25%, 75%)	p-value*
<b>Physical Functioning Scales</b>			
>12,000 steps	94.5 (5.3)	95.3 (89.1, 99.2)	<b>0.01</b>
< 12,000 steps	65.3 (14.9)	59.4 (53.1, 79.7)	
<b>Emotional Functioning Scales</b>			
>12,000 steps	63.8 (21.0)	67.5 (42.5, 81.3)	0.09
< 12,000 steps	43.9 (19.0)	50.0 (35.0, 57.5)	
<b>Social Functioning Scales</b>			
>12,000 steps	90.0 (8.2)	90.0 (82.5, 97.5)	<b>0.03</b>
< 12,000 steps	64.4 (18.8)	60.0 (47.5, 82.5)	
<b>School Functioning Scales</b>			
>12,000 steps	70.0 (12.2)	72.5 (57.5, 80)	<b>0.02</b>
< 12,000 steps	43.3 (22.9)	50.0 (27.5, 55.0)	
<b>Psychosocial Functioning Scales<sup>1</sup></b>			
>12,000 steps	74.6 (10.7)	77.5 (63.3, 82.9)	<b>0.03</b>
< 12,000 steps	50.6 (18.5)	55.0 (35.8, 65.0)	
<b>Total PEDs QL Scales<sup>2</sup></b>			
>12,000 steps	81.5 (6.5)	82.6 (75.0, 87.0)	<b>0.01</b>
< 12,000 steps	55.7 (13.3)	55.4 (48.4, 65.2)	

\*All p-values were generated from the Mann-Whitney U test. Psychosocial Functioning Scales were generated from the average of the Emotional, Social, and School Functioning Scales. Total PEDs QL Scales were generated from the average of the Physical, Emotional, Social and School Functioning Scales.

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