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Tic reduction following dehydration in Gilles de la Tourette Syndrome (TS) †

Ashley B. Durbin [1,*], Matthew R. Rodgers [1], and James R. Brasic [1]

[1] Johns Hopkins University, Baltimore, Maryland, USA; mrodge12@jhu.edu (M.R.R.), jbrasic1@jh.edu (J.R.B.)
* Correspondence: adurbin3@jh.edu (A.B.D.)
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Abstract: A 24-year-old man with the syndrome of Gilles de la Tourette experienced a marked remission of 2 years after heat-induced dehydration. Further investigation of further effects of heat and dehydration on TS may be useful.

Keywords: Neurotransmitter; movement disorder; clinical trial; dopamine; scales; non-motor scales

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1. Introduction

Gilles de la Tourette Syndrome (TS)- related tics are challenging to eliminate. Heat has, in some cases, reduced compulsive symptoms in TS. However, prior reports have produced opposing results. Individuals with tics and other symptoms of TS typically experience a decrease in symptoms with dopamine-receptor blocking drugs, while stimulants cause an exacerbation of symptoms. Those who can attain remission likely have a later age of onset as opposed to those who have had an earlier age of onset [1].

We sought to assess the beneficial and adverse effects of heat on a man with TS.

2. Materials and Methods

A 36-year-old man with TS presented for a follow-up research examination to report that he is doing well, working 70 hours a week on two jobs.

At 7 years of age, he experienced multiple motor and phonic tics.

At 10 years of age, he experienced episodes of daily emesis preceded by an uncomfortable sensation of regurgitation in his throat when eating solid foods. He was able to resume eating after the emesis.

At 24 years of age, he was unable to carry bottles for fear they would be damaged due to tics. Then, without the knowledge or approval of his physicians, he deliberately entered a hot bath of 103-104 degrees F for 3-4 hours to induce dehydration. He believed this would favorably alter his dopamine metabolism. After about 2 hours, his tics subsided. After leaving the tub, he had cramps in his arms, legs, and neck. For 2 days after leaving the tub, his urine was pinkish, and he felt weak. He experienced a sustained remission of tics.
At 27 years of age, after a tetanus immunization, he had his worst exacerbation of tics for two weeks. The tics gradually returned to a mild level.

At 29 years of age, he began using Δ9-tetrahydrocannabinol (THC) as an inhaled vapor nightly.

His follow-up examinations 1 month and 11 months after the remission showed market improvements upon his initial evaluation, 6 months before the remission (Table 1. Self-rated instruments, Table 2. Examiner-rated instruments).

He indicated he had not used any drugs, including marijuana, at the time of the assessments. Toxicology may be positive for cannabis 30 days or more after its use.

At 25 years of age, the man with TS underwent heat-induced dehydration and experienced a remission for 2 years.

At 27 years of age, after a tetanus immunization, his symptoms were exacerbated.

Now at 36 years of age, he is not currently taking any medications, but occasionally uses vaporized THC and CBD. He is not vaccinated for COVID-19 but was not severely affected when he contracted the illness.

3. Results and Discussion

3.1. Six months before entering a hot tub

As a 24-year-old man, the participant had difficulty working as he feared he would drop glass bottles due to his tics (table 1, video S1).

Table 1. Subjective measurements of 24-year-old man before entering a hot tub.

Instrument	Range of scores	Seven months before remission
Compulsion checklist	(0, 111)	10
Dental pain/fear/anxiety	(0, 112)	4
Dental pain/fear/anxiety Mid	(0, 112)	4
Dental pain/fear/anxiety Post	(0, 112)	1
Dental pain/fear/anxiety 1 month follow up	(0, 112)	0
Dental pain/fear/anxiety 3 months follow up	(0, 112)	0
Fear Questionnaire	(0, 192)	8
Social Situations Questionnaire	(0, 192)	13
Questionnaire for tic disorders	(0, 23)	21
Tic Symptom Self Report Motor	(0, 60)	24
Tic Symptom Self Report Vocal	(0, 60)	11
University of Miami Modified Maudsley Obsessive-Compulsive Inventory	(0, 60)	11
Wender Utah Rating Scale (WURS) Behaviors	(0, 168)	46
Wender Utah Rating Scale (WURS) Medical problems	(0, 28)	1
Wender Utah Rating Scale (WURS) School	(0, 48)	11
Wender Utah Rating Scale (WURS) Attention-Deficit/Hyperactivity Disorder (ADHD) items	(0, 100)	39

3.2. One month after entering hot tub

After entering the hot tub, the patient had a marked remission from tics (table 2, video S2).

Table 2. Objective measurements.

Instrument	Range of scores	Six months before remission	One month after remission	Eleven months after remission
Urine drug toxicology for tetrahydrocannabis	Negative or positive	Negative	Positive	Positive
Abnormal Involuntary Movement Scale (AIMS)	(0, 40)	9	0	1
Clinical Global Impression (CGI) Severity Index (SI)	(0, 7)	4 Moderately mentally ill	3 Mildly ill	2 Borderline mentally ill
Clinical Global Impression (CGI) Global Improvement (GI)	(0, 7)	4 No change	2 Much improved	1 Very much improved
Clinical Global Impression (CGI) Efficacy Index (EI)	(0, 16)	13	5	13
Clinical Global Impression (CGI) Therapeutic effect	Not applicable	Unchanged or worse	Moderate	Unchanged or worse
Clinical Global Impression (CGI) Side effects	Not applicable	None	None	None
Clinical Global Impression (CGI) Attention Deficit Disorder (ADD)	(0, 6)	1 Borderline	0	1 Borderline
Clinical Global Impression (CGI) Obsessive-Compulsive Disorder (OCD)	(0, 6)	3 Moderate	2	2 Mild
Clinical Global Impression (CGI) Tourette syndrome (TS)	(0, 6)	3 Moderate	1	1 Borderline
Clinical Global Improvement (CGI) Rater Global Evaluation (RGE)	(1, 7)	4 Unchanged	2 Much improved	1 Very much improved
Hillside Akathisia Scale (HAS) Subjective items	(0, 8)	0	0	0
Hillside Akathisia Scale (HAS) Objective items	(0, 12)	6	0	0
Hillside Akathisia Scale (HAS) Clinical Global Impression (CGI) Severity of Akathisia (SA)	(0, 7)	3 Mildly akathisic	11 Normal, not akathisic	1 Normal, not akathisic
Hillside Akathisia Scale (HAS) Clinical Global Impression (CGI) Global Improvement (GI)	(0, 7)	4 No change	1 Very much improved	1 Very much improved
Brief Psychiatric Rating Scale (BPRS) Anchors	(20, 140)	26	21	21
Movement Disorders Checklist	(0, 23)	14	8	0
Myoclonus versus tic checklist	(-2, 6)	6	3	0
National Institutes of Mental Health (NIMH) Obsessive-Compulsive Scale (OCS)	(0, 15)	5	2	1
Rating Scale for Acute Drug-Induced Akathisia (RSADIA) Subjective	(0, 9)	0	0	0
Rating Scale for Acute Drug-Induced Akathisia (RSADIA) Objective	(0, 21)	1	0	0
Rating Scale for Acute Drug-Induced Akathisia (RSADIA) Global Rating	(0, 3)	0	0	0
Rating scale for drug-induced akathisia (RSDIA) Subjective	(0, 6)	0	0	0
Rating scale for drug-induced akathisia (RSDIA) Objective	(0, 3)	1	0	0
Rating scale for drug-induced akathisia (RSDIA) Global Clinical Assessment of Akathisia (GCAA)	(0, 5)	1	0	0
Rating Scale for Tardive Dyskinesia (RSTD) Face	(16, 96)	26	17	18
Rating Scale for Tardive Dyskinesia (RSTD) Neck and trunk	(8, 48)	11	8	8
Rating Scale for Tardive Dyskinesia (RSTD) Extremities (upper)	(8, 48)	11	8	8
Rating Scale for Tardive Dyskinesia (RSTD) Extremities (upper)	(8, 48)	8	8	8
Rating Scale for Tardive Dyskinesia (RSTD) Entire body	(4, 24)	4	4	4
Times Stereotypies Rating Scale	(0, 1000)	27	2	1
Tourette Syndrome Diagnostic Confidence Index (TSDCI)	(0, 100)	61	Missing	82
Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)	(0, 40)	4	10	4
Obsessive-Compulsive Disorder through the application of the current criteria to the Yale-Brown Obsessive-Compulsive Scale (Y-BOCS)	(0, 1)	0	1	0
Yale Global Tic Severity Scale (YGTSS) Motor	(0, 25)	19	13	12
Yale Global Tic Severity Scale (YGTSS) Phonic	(0, 25)	9	0	11
Yale Global Tic Severity Scale (YGTSS) Impairment	(0, 50)	27	0	9

3.3. Subsequent Course

The remission after the dehydration continued for 2 years until he received a tetanus immunization that led to the worst-ever exacerbation of tics.

He currently continues to function with tics controlled through the used to vaporized THC every night.

3.4. Discussion

A 36-year-old man with TS, experienced a two-year remission at 24 years of age after profound heat-induced dehydration. This remission ended after he received a tetanus immunization which resulted in his worst exacerbation of tics. He has controlled his tics with the use of vaporized THC every other night. He continues to hold several jobs working approximately 70 hours per week.

It is important to note that heat-induced dehydration can have severe consequences. It can lead to fatality and other health-related problems when induced without medical supervision.

The regular use of the vaporized THC may be beneficial to control symptoms and signs of TS.

3.4.1. Limitations

There are limitations of this case report. Because it is a single case, the findings cannot be applied to any general population.

As the symptoms and signs of TS routinely fluctuate, this patient's remission may represent a natural course of the disease and not an effect of the heat-induced dehydration.

Additionally, there may be beneficial effects of aging of people with TS. The patient may be demonstrating decreased presentation of the signs and symptoms because of attaining maturity.

3.4.2. Future Directions

The literature about the effect of heat in TS is conflicting. Results of treatment of TS through heat-induced dehydration are uncertain.

Controlled clinical trials of the effects of heat and dehydration are merited to investigate the possible beneficial effects of this intervention.

There is evidence that CB1 receptor binding, a common association with the presence of tics and other symptoms of Tourette Syndrome, is greatly reduced after THC treatment [2].

Additionally, there is indication that cannabis medicine could be beneficial to the treatment of TS, especially with the approach of starting with low dosages and increasing over time [3].

There exists a radio-tracer available that has been effective for schizophrenia, that might be useful for a future direction in Tourette research. Investigation of people with TS, with and without use of THC could be beneficial [4-6].

We hypothesize that patients with TS also experience down-regulation of cannabinoid receptor subtype 1 (CB_1) with regular use of THC. Clinical trials to investigate the effect of THC agents on CB_1 receptors on people with TS is indicated.

4. Conclusions

Regular use of THC and other cannabis derivatives may be beneficial for the control of tics related to TS.

Supplementary Materials: The following supporting information can be downloaded at: www.mdpi.com/xxx/s1, Video S1: A 24-year-old man was asked to stand still for 2 minutes: Video S2: A 25-year-old man was asked to stand still for 2 minutes.

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Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki and approved by the Institutional Review Board of Johns Hopkins Medical Institutions (protocol code NA_00036810 and date of approval on 2/23/2010; NA_00036828 and date of approval on 4/15/2010).

Informed Consent Statement: Informed consent was obtained from all subjects involved in the study.

Data Availability Statement: All data is provided in the tables and videos.

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Toronto, Canada, June 5-9, 2011 [7]; 1st World Congress on Tourette Syndrome and Tic Disorders. London, UK, June 24-26, 2015. [8]; MD-Washington Regional Movement Disorders Video Meeting, September 28, 2021 [9]; XXVII World Congress on Parkinson's Disease and Related Disorders, Prague, Czech Republic, 1-4 May 2022 [10].

Conflicts of Interest: The authors declare no conflict of interest.

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Tables

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3.1. Six months before entering a hot tub

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Rating scale for drug-induced akathisia (RSDIA) Subjective	(0, 6)	0	0	0
Rating scale for drug-induced akathisia (RSDIA) Objective	(0, 3)	1	0	0
Rating scale for drug-induced akathisia (RSDIA) Global Clinical Assessment of Akathisia (GCAA)	(0, 5)	1	0	0
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Yale Global Tic Severity Scale (YGTSS) Motor	(0, 25)	19	13	12
Yale Global Tic Severity Scale (YGTSS) Phonic	(0, 25)	9	0	11
Yale Global Tic Severity Scale (YGTSS) Impairment	(0, 50)	27	0	9

3.2. One month after entering hot tub

After entering the hot tub, the patient had a marked remission from tics (table 2, video S2).

Table 2. Objective measurements.