Delta-9-tetrahydrocannabinol for the treatment of a child with Tourette syndrome: case report

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ABSTRACT

Background: Tourette syndrome (TS) is a childhood-onset neuropsychiatric disorder characterized by motor and vocal tics. In severe treatment-resistant cases of TS, cannabis-based medicine could be used alternatively as a therapy of last choice.

Case Presentation: We present the case of an 7-year-old boy with severe TS and comorbid attention deficit/hyperactivity disorder (ADHD), who significantly benefitted from treatment with cannabis-based medicine. During an episode with increased tics, he became depressed, developed suicidal ideation, and exhibited separation anxiety resulting in social isolation. As treatment with various antipsychotics and Habit Reversal Training turned out to be unsuccessful, we implemented therapy with oral delta-9-tetrahydrocannabinol (THC) as oil-based drops. Starting dose was as low as 0.7 mg THC/day once a day and was gradually increased up to a maximum dose of 29.4 mg THC/day, resulting in a significant improvement of both tics and behavioral symptoms. Follow-up visits over a period of 4 months demonstrated a sustained treatment effect without any adverse events.

Conclusion: From this single case report, it is suggested that THC is effective and safe in the treatment of tics, depression, and ADHD in children with severe and otherwise treatment-resistant TS.

Keywords: THC, Tourette syndrome, tics, cannabis, children.

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Background

Tourette syndrome (TS) is a childhood-onset neuropsychiatric disorder characterized by motor and vocal tics. In 80%–90% of cases, additional psychiatric comorbidities occur such as attention deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), anxiety disorder, and depression. In a substantial number of patients, TS causes a significant impairment in quality of life. First line treatment for tics in children is either behavioral therapy or pharmacotherapy with antipsychotics. Alternatively, α-adrenoceptor agonists or less established drugs such as topiramate or botulinum toxin are used. In severely affected and otherwise treatment-resistant children, only a few alternatives can be offered. In adults, in addition, cannabis-based medicine such as medicinal cannabis [1], delta-9-tetrahydrocannabinol (THC, dronabinol) [2], and nabiximols [3,4] has been found to be effective in reducing tics. In minors, so far, there is only one single case report available describing the beneficial effects of dronabinol in a 15-year-old male adolescent with treatment-resistant TS and ADHD [5]. We report the first case of a successful treatment with THC in an 8-year-old child with TS.

Case Presentation

The 7-year-old boy diagnosed with TS and ADHD was referred to our TS outpatient clinic for consultation due to general symptom increase. There were no complications during pregnancy, labor, and psychomotor development and he was not affected by any serious somatic conditions, besides a history of recurrent streptococcal infections. His mother had mild tics in childhood and his grandfather suffered from dementia. Patient's father suffered from cluster headache that was successfully treated with nabiximols for many years. Our patient developed his first tics at the age of 6, which followed a typical waxing and waning course and deteriorated significantly at the age of 7. At this stage, the patient was highly impaired by his tics, in particular, by motor tics of his right arm rendering him unable to write and by very loud vocal tics. School attendance was nearly impossible and the patient refused to leave home altogether, which lead to social isolation and a loss of friends. In addition, he became depressed, developed suicidal temptation, and exhibited separation anxiety.

Treatment attempts (alone or in combination) with risperidone (up to 2 mg/day), aripiprazole (up to 30 mg/day), tiapride (up to 500 mg/day), methylphenidate (up to 20 mg/day), and guanfacine (up to 2 mg/day) as well as Habit Reversal Training and occupational therapy were unsuccessful.

We decided to augment risperidone (2 mg/day) and guanfacine (2 mg/day) with oral THC (oil-based drops). Starting dose was as low as 0.7 mg THC/day once a day and was gradually increased over a period of about 2 months. Above a dosage of 3.6 mg THC/day, positive effects on both tics and behavior were reported by the patient, his parents, and teachers. After 2 weeks, a daily dose of 5.4 mg THC once in the morning was reached. According to the parents' report, this resulted in a tic reduction of about 50% lasting for 3–4 hours accompanied by only mild and transient sedation. Therefore, dosage was slowly up-titrated to a dose of 18.2 mg THC/day

Table 1. Clinical measurements before and during treatment with THC.

SYMPTOM	SCALE [RANGE]	BASELINE BEFORE THC	FOLLOW-UP		PERCENTAGE OF IMPROVEMENT (BASELINE *VS.* FOLLOW-UP AFTER 4 MONTHS)
			AFTER 2 MONTHS	AFTER 4 MONTHS	
		MEDICATION: DOSE [MG] OF THC/RISPERIDONE/GUANFACINE			
		0/2/2	19.6/1/2	22.4/0/2	
Tics	– Yale Global Tic Severity Scale Total tic score [0–50]	38	21	17	–28.9
	– Rush Video-Based Tic Rating Scale [0–20]	12	10	11	–8.4
Tics + impairment	– Yale Global Tic Severity Scale Global score [0–100]	68	31	27	–60.3
Premonitory urges	– Premonitory Urge for Tics Scale [0–40]	11	5	3	–72.7
Quality of life	– Gilles de la Tourette Syndrome—Quality of Life Scale [0–100]	42	6	7	–83.3
	– Questionnaire for Measuring Health-Related Quality of Life in Children and Adolescents Parent's Questionnaire [0–100]	65	96	92	+41.5*
Global impairment	– Clinical Global Impression—Severity Scale [0–7]	5	4	3	–40
	– Clinical Global Impression—Improvement Scale [0–7]	–	2	2	–
Depression	– Depressionsinventar für Kinder und Jugendliche, German instrument to measure intensity of depression in children and adolescents [33–80]	53	45	41	–22.6
Stress	– Perceived Stress Scale [0–40]	36	9	9	–75.0
Behavior	– The Strengths and Difficulties Questionnaire [0–40]	40	24	18	–55.0
Autistic traits	– Autismus-Spektrum Screening Fragebogen [0–56]	22	9	15	–59.1
ADHD	– Swanson, Nolan and Pelham Teacher and Parent Rating Scale [0–78]	34	19	20	–41.2
OCD	– Children's Yale-Brown Obsessive Compulsive Scale [0–40]	0	0	0	–

*The higher the score, the better quality of life.

twice daily after 4 weeks. Temporarily, even higher doses (up to 29.4 mg THC/day) were used to control tic intensity without causing any additional side effects. After having started treatment with THC, the patient was reported to be more engaged in family activities, to be able to focus better at school, to attend all classes, to be overall more at ease, and to experience higher acceptance by others. Most importantly, the patient restarted making appointments with friends, left home (for outdoor activities), and was as adventurous as before, resulting in a tremendous quality of life improvement. Table 1 summarizes results of clinical assessments at our clinic before and after 2 and 4 months of treatment with THC demonstrating not only a tic reduction, but also an improvement in ADHD, mood, stress, general impairment, and patient's quality of life. No detrimental effects, besides mild tiredness at the beginning of the treatment, were noted. In parallel, treatment with risperidone could be gradually withdrawn.

Discussion

This is the first case report suggesting that oral treatment with the cannabis-based medicine might be an effective and safe treatment option in otherwise treatment-resistant children with severe and complex TS. Nevertheless, the tics improved only modestly, while the better quality of life was mostly due to an improvement of comorbidities including ADHD and depression. We cannot entirely exclude that symptom improvement was—at least in part—caused by spontaneous fluctuations of symptoms or a placebo effect. However, tics improved only after the addition of THC and remained stable over more than 4 months, while several other treatment strategies failed to improve symptoms. Most remarkably, even relatively high dosages of THC (up to 29 mg/day) were well tolerated; and only mild and transient sedation was reported by the parents at the beginning of the treatment. Beyond that, no other side effects or negative impact on school performance were observed.

Conclusion

The observation made in this case report suggests that children might tolerate treatment with cannabis-based medication, such as THC, even better than adults. However, long-term follow-up is needed for a final evaluation of the efficacy and safety of treatment with THC in this boy.

Acknowledgements

None

List of abbreviations

ADHD	Attention deficit/hyperactivity disorder
OCD	Obsessive-compulsive disorder
THC	delta-9-tetrahydrocannabinol
TS	Tourette syndrome

Consent for publication

Written informed consent was obtained from patient's parents.

Ethical approval

No ethical approval was required to carry out this experimental therapy and to publish an anonymous case report in a medical journal.

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Summary of the case

Patient (gender, age)	1	Male, 7
Final diagnosis	2	TS, ADHD
Symptoms	3	Severe motor and vocal tics, depression
Medications (Generic)	4	THC
Clinical procedure	5	THC as oil-based drops. Starting dose was as low as 0.7 mg THC/day once a day and was gradually increased up to maximum dose of 29.4 mg THC/day
Specialty	6	Psychiatry