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Cannabinoids in functional tic-like movements☆

Interest in the therapeutic use of cannabinoids in medicine with a focus on neuro-psychiatric disorders has significantly increased. In tic disorders, for example, positive effects of cannabinoids have been reported not only for the repetitive motor behaviors [1–4], but also for relevant psychiatric comorbidities, such as impulsivity, attention deficit/hyperactivity disorder (ADHD), and obsessive-compulsive behaviors [1]. To date, there has been no report on the effects of cannabinoids in functional tic-like disorders- a rather rare presentation of functional movement disorders [5–7]. We recently encountered five patients with functional tic-like movements in our clinics, who reported a drastic effect of cannabis on their abnormal movements. Most importantly, although these patients had received a previous diagnosis of chronic tic disorder or Tourette syndrome (TS), specific features of clinical history and examination pointed towards the diagnosis of a functional tic-like disorder [5–7]. We provide a detailed description of an illustrative case (case 1; previously included in[7]) that is accompanied by video material, as well as a clinical summary of all patients, and discuss the relevance of this observation in clinical practice.

A nineteen-year old male (case 1) presented to our clinic due to repetitive involuntary movements and sounds. Initially, movements such as tongue protrusion and/or mouth opening and shrieking had appeared suddenly ("overnight") at the age of 13. A subsequent dramatic increase in the severity of the unwanted movements over a few days with bouts of complex self-injurious behaviors led to the diagnosis of TS. Several medications, including aripiprazole, risperidone and lorazepam were unsuccessfully attempted (listed in Table 1). At age 15, the shouting of obscenities began to occur. Since that age he has been using cannabis (smoking; ca. 3g/day) on a daily basis. He reported feeling calmer and being able to keep the unwanted behaviors under control (Video 1A).

Supplementary video related to this article can be found at https://doi.org/10.1016/j.parkreldis.2018.09.027.

Clinical examination was unremarkable, but for the presence of the aforementioned complex motor behaviors (Video 1B). According to the patient, the abnormal motor behaviors were prompted by a gradually rising sensation ("energy pulse") perceived in his chest. Allowing the motor behaviors to emerge provided temporary relief from the somatic experience. Additional information, as well as a summary of the clinical characteristics of all five cases is provided in Table 1. In all cases, relevant clinical and paraclinical investigations, including laboratory tests, brain imaging and electroencephalography were normal.

We here present five patients with tic-like movements that we believe fall within the functional movement disorder spectrum. Although, we appreciate that the distinction between primary tics and functional tic-like jerks is challenging for reasons highlighted previously [5–7], we believe that the cases we present here fit within the latter diagnostic category. Indeed, in all cases the abnormal behaviors were complex and extremely unusual for a primary tic disorder (e.g. case 1: bouts of long episodes of painful tensing and grunting; case 2: continuous hand jerking; case 4: biting husband's neck). Moreover, clinical history, as for example the abrupt onset in cases 1, 3 and 5, and the associated phenomenology (e.g. case 1: holding onto objects in order to suppress tics; case 5: experiencing malaise and nausea during tic suppression) clearly differed, in our view, from the presentation of patients with TS. Interestingly, all patients reported a substantial and sustained improvement in their motor behaviors and overall well-being with the use of cannabis. In fact, access to medicinal cannabis was the predominant reason of their presentation in our clinics.

Currently, there is a strong scientific interest to explore the effects of cannabinoids in the treatment of several movement disorders. In the case of tic disorders, several randomized controlled trials are currently underway. The cases we present here are important, therefore, for the following three reasons. First, they demonstrate that the distinction between primary tics and functional tic-like disorders could be notoriously difficult and that misdiagnoses are not uncommon [7]. Indeed, all our patients carried a previous diagnosis of either chronic tic disorder or TS and were actively seeking cannabinoid treatment. Although, we cannot exclude that some patients may also have had mild primary tics at some point, the predominant clinical presentation and the reason for cannabinoid intake was due to their functional tic-like behaviors. Second, our cases highlight one potential pitfall of clinical trials in tic disorders. In the absence of any clear biomarker to distinguish tics from functional tic-like movements, correct patient recruitment for therapeutic trials in TS is crucial and should be performed in centers with expertise in the diagnosis and treatment of tic disorders. Third, we note the consistency of reported improvement with the use of cannabis in our patients. Typically, patients with functional movement disorders, and specifically functional tic-like movement disorders, report either no or only temporary effects of medication on their abnormal motor behaviors and well-being [7,8]. However, in the cases reported here, positive effects of cannabis were consistently maintained for as long as 5 years (case 1). Although, the mechanisms behind the reported improvement are certainly complex, we do note the unusual efficacy of this medication in this group of patients.

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Table 1
Clinical characteristics of patients with functional tic-like movements.

Nr/Sex/Age	Age at onset of functional tic-like movements	Temporal evolution/precipitants	Symptoms at onset	Current clinical presentation	Previous Medications/Effect	Previous Dx
1/M/19[a]	13	Abruptly while watching TV	Tongue protrusion, gagging sounds	Whole body movements, tensing, arching and twisting of his back, holding on to objects; self-injurious behaviors. Sensation of "energy pulse" prior to bouts of abnormal movements.	Risperidone, Sulpiride, Lorazepam, Clonazepam, Chlorprothixene, Methylphenidate, Aripiprazole/All no effect. Symptom-free when smoking cannabis	TS + ADHD
2/M/24	21	Insidious onset	Inability to sit still; need to shake hands repetitively	Cannot leave house because of need to repetitively shake legs and hands; "ehm" "ha" vocalizations. No sensory premonition.	Lorazepam, Clonazepam, Aripiprazole, Risperidone/All no effect. Symptom-free when smoking cannabis	TS; ADHD; Restless Legs; Borderline personality disorder; Addiction to benzodiazepines
3/F/19[a]	16	Abruptly whilst admitted in psychiatric ward due to depression, panic attacks, and self-injurious behavior	Head jerking	Predominantly complex copro- and echophenomena, mostly vocalizations, self-injurious behaviors	Atomoxetine, Aripiprazole, Tiapride, Risperidone/All no effect. Notices improvement with Nabiximols.	TS + ADHD
4/F/49	7	Insidious onset in childhood; age 47 abrupt onset of complex motor behaviors	Grimacing, simple vocalizations	Whole body jerks. Biting (e.g. bit once her husband's neck), long coprolalic phrases, need to sing, barking. Word "Abilify" can only be whispered. Behaviors are temporarily suppressible and associated with an unpleasant sensation of pressure.	Aripiprazole/No other effect, but nausea. Smoking cannabis leads to a perceived 80% improvement.	TS.
5/M/50	49	Abrupt onset	Pressure in umbilical area leading to leg jerks	Rising pressure in abdomen relieved through sudden jerking of legs whilst walking; Voluntary suppression of movements leads to more pressure, malaise, nausea and sweating; gait disorder	Tiapride/No effect. Symptom-free when smoking cannabis	Chronic tic disorder + ADHD

[a] Previously published in Ref. [7].

Conflicts of interest

All authors report no conflicts of interest.

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