

SENILE-ONSET TOURETTE SYNDROME: A CASE REPORT

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INTRODUCTION

Tourette's syndrome (TS) was described by Gilles de la Tourette in 1885 and the contemporary definition in the appearance before the age of 18 of multiple motor tics and at least one vocal tic occurring several times a day or intermittently for a minimum of 1 year, with no more than 3 consecutive months tic-free. It is reported that the average age of onset of tics is 7 years, but tics may occur as early as the age of 2 years. In order to establish the diagnosis, the onset must occur before the age of 18 years, although the age at onset is a matter of debate. However there is a limited number of reports concerning the adult onset of TS. Another case is Tardive TS which is characterized by the occurrence of multiple motor and vocal tics in patients on long-term neuroleptic medication (Fountoulakis & Panagiotidis 2011, Fountoulakis et al. 2011).

CASE REPORT

A case report of a female aged 81 is being described who presented to the private practice of the first author complaining of frequent eructation. Clinical examination revealed that in reality it was a complex motor and vocal tick suggestive of TS and more specifically of senile-onset TS (SOTS). The patient had a history of depression since the age of 23 with postpartum onset. For the next 40 years depression had followed a benign course with rare relapses and good overall outcome but after the age of 60 relapses became more frequent and chronicity appeared. At around that age she also developed complex motor and vocal ticks for the first time. She received treatment with various antidepressants without any response. Since young age she was diagnosed with hypothyroidism and was under treatment. During the last few years she is suffering from mild hypertension. She used to be a heavy smoker and unspecified severity of alcohol abuse but quit 15 years ago. Brain MRI showed diffuse vascular subcortical encephalopathy (Figure 1). At present she was receiving duloxetine 60 mg/day and quetiapine 50 mg/day. She was put on 100 mg/day amisulpride on top of ongoing treatment and ticks disappeared after two weeks. Remission was stable after two months. Depression remained unaltered. Antidepressant treatment was changed to escitalopram 20 mg/day but the patient stopped it because of morning headaches.

DISCUSSION

In the literature authors are very careful and often refer to 'tics' instead of TS even when the clinical criteria are met, probably because of the prevailing opinion concerning the young age at onset. Tics that are due to a secondary cause such as drug use, brain insult or neurodegenerative disease could constitute one-third to one-half of adult onset syndrome and the pathogenesis involves a dysfunction at the basal ganglia level. There are only four case series of adult-onset tics not attributed to any other neurologic disorder and not because of recurrence of childhood tics (Chouinard & Ford 2000, Eapen et al. 2002, Jankovic et al. 2010, Erro et al. 2014) and five case reports. Of all these cases only three concern a senile-onset, one in a 81 year black man with unspecified mild brain atrophy who responded excellently to 1 mg haloperidol/day (Sutula & Hobbs 1983) and one in a 66 year old man with delusional depression and no pathological neuroimaging findings, which responded well to clonidine 0.25 mg BID (Fliman et al. 1991). In both cases and underlying vascular brain pathology could be suspected but imaging of the time failed to unveil it. A third concerned a 78 year old man with mild vascular dementia documented by brain imaging, who did well on paroxetine 20 mg/day plus haloperidol 1 mg/day (Sikdar & Paranthaman 2002). However, this kind of pathology is not included in those factors associated with secondary tics. Only a minority (8%) of adult-onset are reported to be female (Robakis 2017).

While TS in children is treated with a combination of education, medication, behavior therapy, botulinum toxin, and, rarely, deep brain stimulation surgery, there are no data concerning the usefulness of these modalities in adult onset TS. There are isolated case reports suggesting a favorable response to verapamil, benzodiazepines, haloperidol, tetrabenazine, and eight to botulinum toxin (Robakis 2017). The current case report is the first to report a SOTS in a female and the second to document an underlying vascular brain pathology. It also confirmed response of this kind of cases to treatment with strong D2 blockers like haloperidol or amisulpride.

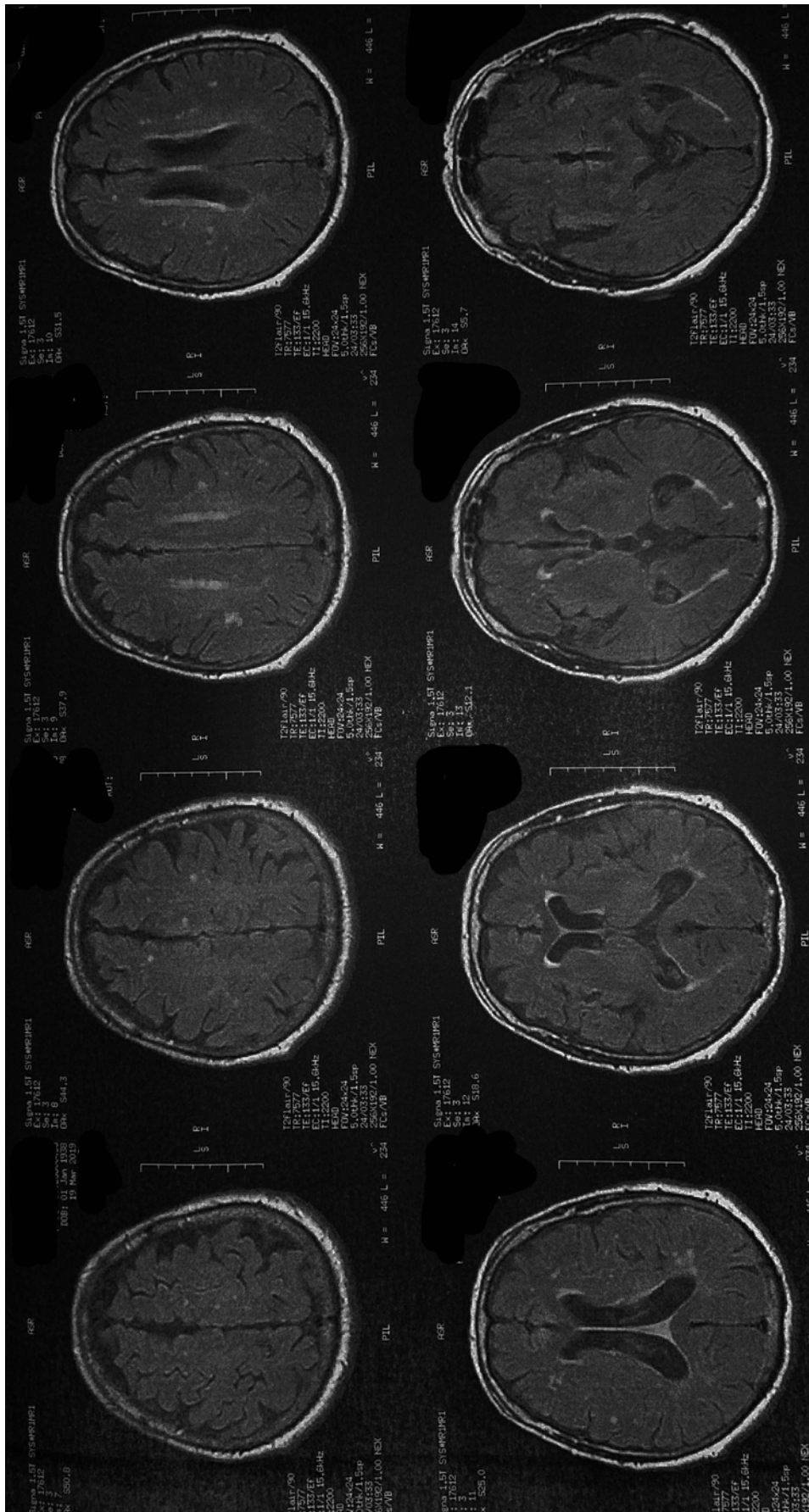


Figure 1. Brain MRI of the 81 years old female patient with senile-onset TS. There are signs of diffuse and focal vascular encephalopathy

CONCLUSION

We certify that this work has the element of novelty since it is the first case report of senile onset Tourette's syndrome (TS) in a female patient. It also confirms the claims of a previous case report on a senile male patient that senile onset is related to vascular encephalopathy and that it responds to treatment with potent and specific D2 antagonists (haloperidol or amisulpride) (Sikdar & Paranthaman 2002). It adds to the literature and specifically to the clinical practice because it identifies a specific type of ticks which respond exclusively to specific antipsychotics at low dosage. The usual treatment approach clinicians have for elderly patients is to attribute ticks to anxiety and treat them with antidepressants (as happened with the reported case). Thus, this case report identifies TS in the elderly, points to the vascular causality and maybe to the high risk of an evolving vascular dementia and suggests a very specific treatment option.

Contribution of individual authors:

Konstantinos N. Fountoulakis conceived the topic, examined the patient, reviewed the evidence and wrote the first and last version of the manuscript.

Maria-Valeria Karakasi reviewed the evidence and contributed to all the versions of the manuscript.

Ioannis Nimatoudis & Vasilios Kimiskidis examined the patient, reviewed the evidence and contributed to all the versions of the manuscript.

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