Neurología. 2020;35(1):46—72

SOCIEDAD ESPAÑOLA DE NEUROLOGIA

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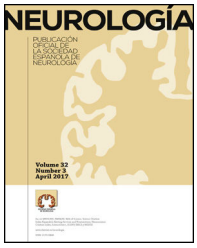

LETTERS TO THE EDITOR

Pollakiuria as a complex motor tic in a patient with Tourette syndrome☆,☆☆

Polaquiuria como tic motor complejo en un paciente con síndrome de Tourette

Dear Editor,

Tourette syndrome (TS) is a condition with onset in childhood, characterised by motor and phonic tics, and such associated psychiatric disorders as obsessive-compulsive disorder (OCD) and attention-deficit/hyperactivity disorder; patients with TS present a wide variety of phenotypes. It is believed to be caused by a dysfunction of different cortical circuits, especially in the fronto-parietal cortex and basal ganglia, which explains the clinical heterogeneity of the syndrome. The current theory is that TS is caused by alterations in the brain's development and maturation.[1] Pollakiuria is defined as an abnormal urinary frequency, and has occasionally been described as a tic in children with TS or paediatric autoimmune neuropsychiatric disorders associated with streptococcus.[2] There are no reported cases of pollakiuria in adults diagnosed with TS. We describe an adult case of TS and pollakiuria.

Our patient was a 46-year-old man with OCD, hypothyrodism, and TS diagnosed according to the DSM-IV criteria since childhood, with simple and complex motor and phonic tics. He responded well to tetrabenazine (125 mg/day). He had been treated for depression or anxiety disorder by the psychiatry department; at some point during this treatment, he presented benzodiazepine overuse. In January 2012, as he presented good emotional control and no tics, the patient himself decided to progressively discontinue all medication (tetrabenazine at 125 mg/day and dipotassium clorazepate at 30 mg/day). In November 2012, he consulted due to a history of pollakiuria of several months' duration, reporting a urinary frequency of up to 50 times a day, which incapacitated him. The patient reported perineal and left inguinal pain, which was alleviated when he urinated. Pain was diffuse, dull, of short duration, and paroxysmal, and was exacerbated by sitting. The symptoms incapacitated him fully, preventing him from going outdoors or living independently. He reported no symptoms suggesting depression or anxiety recurrence. Months before his follow-up appointment, he had already consulted with the urology department, which requested the following tests: lumbar MRI scan, urinary tract and abdomen and pelvis ultrasounds, cystography, urine culture, blood analysis, urodynamic study, and EMG of the pelvic wall; all tests yielded normal results. At the time of consultation, he reported having used antimuscarinic drugs with no benefits. The neurological examination identified no abnormalities. This was a case of pollakiuria as a response to several months' history of genital pain with no bladder, prostate, or genital lesion. Since no alterations were found in the neurological examination, lumbar MRI scan, or EMG, we also ruled out lumbosacral neuromuscular disease. Considering the reported symptoms, we considered the possibility of a motor tic or compulsion. We believed the pain to be a premonitory sensation and urination to be the motor pattern alleviating pain; therefore, our initial diagnostic hypothesis was a complex motor tic, to be treated with aripiprazole at 10 mg/day. One month after treatment onset, the patient was asymptomatic and reported no adverse effects.

Obsessive-compulsive disorder, or anakastic personality disorder, shares many features with TS: both mainly affect young patients, present a remitting course and premonitory sensation prior to motor tic, and may be associated with complex motor rituals. Between 25% and 50% of patients with TS present OCD and 20%—30% of OCD patients have tics. Therefore, in a patient with TS presenting complex motor rituals, a differential diagnosis of tics and compulsion should be performed.[3] The function of compulsion is to alleviate an obsession or the anxiety caused by an unpleasant intrusive thought. However, tics involve a premonitory sensation which can be defined as a sense of urgency to perform the movement, although it may also be a physical symptom localised in the area where the tic occurs. In our patient, the genital pain, alleviated by urination, may be considered the premonitory sensation that appears by definition before the tic. Urinating would be the complex motor tic.

Interoception is the capacity to perceive sensations originating in the internal organs of the body. Increased perception of the need to urinate may also be considered as heightened interoception in our patient. Increased interoceptive capacity has been related to increased perception of the premonitory sensation of tics, which in turn has been associated with more severe tics.[1] Our patient's pollakiuria may be considered a severe complex motor tic causing a significant limitation to his daily life.

☆ Please cite this article as: García-Ramos R, López Valdés E. Polaquiuria como tic motor complejo en un paciente con síndrome de Tourette. Neurología. 2020;35:46—47.

☆☆ This study was presented in poster format at the 2013 Annual Meeting of the SEN.

It is important to identify the symptoms of patients displaying complex motor rituals in order to select an appropriate therapeutic approach, since compulsion and tic require different treatments. Aripiprazole is a well-tolerated drug which achieves adequate control of tics.[4]

Pollakiuria in an adult patient with TS should raise suspicion of the possibility of a complex motor tic, which would require an adjustment of the therapeutic approach.

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https://doi.org/10.1016/j.nrleng.2017.02.013
2173-5808/

Scurvy and Wernicke's encephalopathy: An underdiagnosed association?☆

Encefalopatía de Wernicke-escorbuto, ¿una asociación infradiagnosticada?

Dear Editor:

Wernicke encephalopathy (WE), first described in 1881, is caused by thiamine deficiency.[1] The classic triad of symptoms is cognitive alterations, oculomotor dysfunction (nystagmus, ophthalmoplegia), and ataxia. Korsakoff syndrome is regarded as the final stage of WE and typically presents with anterograde amnesia and confabulation.[2] WE may be associated with other deficiency diseases, such as scurvy, which require a high level of clinical suspicion and early treatment with vitamin supplementation.

We present the case of a 50-year-old female patient, a smoker, who came to the emergency department due to vertigo upon waking and gait instability, with a tendency to veer to the right. According to the patient's family, she had begun to display behavioural alterations a week previously, in the form of repetitive behaviour, confusion, and frequent memory deficits. She was afebrile and presented no other systemic symptoms. Examination of the patient revealed altered mental state (disorientation and confusion), anterograde amnesia, complete ophthalmoplegia of abduction bilaterally, multidirectional nystagmus, ataxic gait, and abolished stretch reflexes in the lower limbs. Suspecting WE, we administered thiamine supplementation and ophthalmoplegia resolved in 24 h. A brain MRI scan revealed hyperintensities in the floor of the fourth ventricle, around the aqueduct of Sylvius at the level of the midbrain, the superior colliculus, and also the hypothalamus and medial region of both thalamic nuclei, the mammillary bodies, and the pillars of the fornices; the lesions showed contrast uptake. These findings are compatible with acute/subacute WE. The patient acknowledged regularly consuming large amounts of alcohol and a diet including no fruits or vegetables. A blood analysis revealed hypertransaminasaemia (GOT 454 U/L [normal range, 4-32 U/L]; GPT 315 U/L [5-33]) and low prealbumin levels at 12 mg/dL (normal range, 17-34 mg/dL), indicating malnutrition; we decided to intensify treatment with intravenous thiamine dosed at 500 mg every 8 h.

During hospitalisation, the patient was assessed by the dermatology department due to corkscrew hairs and perifollicular purpura on the pretibial area in both legs. The oral cavity showed generalised gingivitis, with the patient reporting frequent bleeding. Two skin biopsies revealed that the infundibula were dilated and filled with keratin plugs surrounded by fibrosis and chronic inflammation. Although we could not determine the plasma ascorbic acid level, we diagnosed scurvy based on the patient's clinical symptoms and started intravenous treatment with 1 g vitamin C every 24 h for 7 days, followed by oral vitamin C dosed at 200 mg/day.

One month after treatment onset, the skin lesions had disappeared and gingival bleeding had stopped; the patient was eating a varied diet and abstaining from drinking alcohol.

At the 3-month follow-up consultation with the neurology department, ataxia had improved but anterograde amnesia persisted.

☆ Please cite this article as: Villacieros-Álvarez J, Chicharro P, Trillo S, Barbosa A. Encefalopatía de Wernicke-escorbuto, ¿una asociación infradiagnosticada? Neurología. 2020;35:47—;49.