brought to you by T CORE



**FUNDING:** No funding was received for the development of this article.

**FINANCIAL DISCLOSURES:** All authors report no financial interests or any other potential conflicts of interest.

ADDRESS CORRESPONDENCE TO: Miguel Palma, Psychiatry Department, Inpatient Ward, Hospital Professor Doutor Fernando Fonseca, EPE, Via IC-19, 2720-276, Amadora, Portugal; E-mail: miguel.palmaaa@gmail.com

**KEY WORDS:** Psychiatry, obsessivecompulsive disorder, schizophrenia, cerebellum, hoarding.

## [CASE REPORT]

# An OCD Patient Presenting with a Cerebellum Venous Variant in a Family with a Strong Schizophrenia Loading: A Case Report

## by MIGUEL PALMA, MD; NUNO BORJA-SANTOS, MD; BRUNO TRANCAS, MD; CATARINA KLUT, MD; and GRAÇA CARDOSO, MD, PHD

Drs. Palma, Borja-Santos, Trancas, and Klut are from Hospital Professor Doutor Fernando Fonseca, EPE, Amadora, Portugal; and Dr. Cardoso is from Faculdade de Ciências Médicas, FCM, Universidade Nova de Lisboa, Lisboa, Portugal.

Innov Clin Neurosci. 2012;9(9):18-20

### ABSTRACT

The role of cerebellar pathology in psychiatric symptoms has long been recognized. Cerebellar pathology has been associated with obsessivecompulsive disorder pathophysiology, particularly with compulsive hoarding. Likewise, some cerebellum abnormalities have been described in schizophrenia, as well as in comorbidity between obsessivecompulsive disorder and schizophrenia. The authors report the case of a 32-year-old woman with obsessive-compulsive disorder and a cerebellum development variant in a family with a strong schizophrenia loading. This case emphasizes the probable role of the cerebellum in the pathophysiology of both obsessivecompulsive disorder and schizophrenia, and reconsiders the existence of a so called schizoobsessive subtype of schizophrenia.

## **INTRODUCTION**

The role of cerebellar pathology in psychiatric symptoms has long been recognized.<sup>1</sup> In addition to its involvement in functions related to movement coordination, motor learning, and balance, cerebellum is likely implicated in higher-order mental functions, such as cognition, emotional regulation, and behavior.<sup>2,3</sup> Traditional neuronal models of obsessivecompulsive disorder (OCD), involving dysfunction of frontal-subcortical circuits, have been supplemented by an increasing role of the cerebellum in its pathophysiology,<sup>4,5</sup> and exploratory functional magnetic resonance imaging (fMRI) studies have unveiled its role in the neural mechanisms of decision making in compulsive hoarding.6 Recent structural and functional brain imaging studies revealed that some symptoms in schizophrenia could be associated with different cerebellar abnormalities.<sup>3</sup> Moreover, there is also one reported case of comorbidity between OCD and schizophrenia in a young male patient with a developmental cerebellar malformation (Dandy-Walker variant).7 Several authors have noted the overlap between the two disorders in the past 100 years.8 Nevertheless, the cooccurrence of obsessive-compulsive symptoms in psychotic illnesses has been shown to be more prevalent than was previously recognized, and the

comorbidity rates for OCD and schizophrenia can reach over 14 percent.<sup>9</sup> There is growing evidence supporting that this comorbid population, with its particular demographic and clinical features, might belong to a so called schizoobsessive subtype of schizophrenia with a common pattern of neurotransmitter systems and neuroanatomic dysfunction.<sup>9</sup> Cerebellum may be a common area of neurobiologic dysfunction in both disorders and account for symptom coexpression.

#### **CASE REPORT**

V. F. was a 32-year-old caucasian female patient. By the age of 16, she began worrying about any object she or any family member might throw into the garbage basket, fearing that anything valuable could go along with it (apparently this fear was initially caused by the fact that her sister threw many objects away). During the first couple of years after development of these thoughts, they were not very disturbing, and the patient managed to successfully complete high school and start working on her college degrees in Portuguese and English Literature. However, little by little, she began verifying everything she or anyone in the family threw into the garbage basket and, ultimately, forbade everybody of doing so. The garbage, particularly empty containers, was meticulously inspected for anything valuable and then items were washed, folded, collected in plastic bags, and kept in her room, prompting her to sleep in the living room. The patient reported that it was the only way to gett relief from the obsessive preoccupation of mistakenly throwing away valuable objects. The process of washing, according to family reports, involved strong preoccupations with symmetry. Moreover, other obsessive and compulsive symptoms were elicited. The patient repeatedly washed her hands and kitchen utensils, which she related to the poor hygienic habits of her father, diagnosed with alcohol abuse and dependence, who might have contaminated them.

She also meticulously ordered decorative objects in the house, aligning them in a precise fashion. The patient became progressively secluded in her own house: the danger of any family member throwing away precious objects was too great to allow her to leave the house if anyone was left inside.

Even though the ruminations were ego-dystonic, there was only slight resistant effort concerning compulsions. Increasing symptom severity promoted intense social disability. She eventually quit college and became more and more isolated at home.

Diagnosis was reached only at the age of 23, when family members brought the patient to our outpatient clinic and she was observed for the first time by a medical team.

On examination, the patient's appearance was somewhat dishevelled, though she was able to maintain eye contact. Facial expression and mimic were normal. She had a general tone of uncooperativeness, which was sometimes punctuated with cynical remarks. Diverse obsessive thoughts and motor compulsions were evoked, though it seemed difficult for her to spontaneously verbalize them, since these symptoms remained a matter of discomfort and anxiety. No delusional thinking or errors of perception were detected. Mood was euthymic and the affective expression, although quite constricted, was not blunted. Insight to the disease was limited to recognition of undesired thoughts; judgement regarding the morbid nature of her condition and the need for treatment was partially impaired. Neurological examination was unremarkable, and we did not note any motor or posture signs suggestive of cerebellar dysfunction.

After some discussion of her case, the patient accepted referral to the inpatient unit, where she began treatment with fluvoxamine (100mg three times daily).

Routine blood tests, a formal neuropsychological evaluation, and karyotype were normal. Brain magnetic resonance imaging (MRI) revealed a cerebellum variant caused by inferior right venous development abnormality. Because the patient's sister was diagnosed with schizophrenia with some obsessive features, we also requested a brain MRI for the sister, which came back normal.

Our patient was discharged one month later, with partial improvement of obsessive thoughts and compulsions. Soon after discharge, however, the patient discontinued both her medication and the follow-up visits.

We admitted the patient to the inpatient unit four additional times under the Portuguese Mental Health Act and on account of the increasing severity of the hoarding behavior. Symptom clusters were almost the same as in first admission, although she progressively lost insight into her disease. In one episode, the patient presented with slight paranoid ideation. She was treated once with paroxetine (20mg twice daily), which did not worsen her paranoid symptoms, and twice with clomipramine (75mg three times daily) [once in association with flupenthixol, 20mg biweekly intramuscularly]). After every hospitalization, the patient was discharged with partial improvement of obsessive thoughts and compulsions. After the first discharge, we referred her to our day hospital unit, which she refused to attend after four weeks. She was also referred for cognitive behavior therapy (CBT) but dropped out after two sessions. Once home, after each hospitalization, she would stop medication immediately and discontinue follow-up visits within the first month. The patient once explained that the reason she did not adhere to her medication was due to her fear of mismanaging it.

Interesting to note, the patient's mother, though permanently refusing psychiatric care, had a likely diagnosis of schizophrenia without prominent negative symptoms. She exhibited continuous morbid behavior with suspicions about the quality of the food (frequently throwing it away) and of the daughter's medications (telling her not to go to follow-up appointments and to stop pharmacological treatment).

We did not identify classical depressive symptoms in the patient throughout the longitudinal follow-up visits. However, she told us she felt profoundly unhappy about her condition.

During her last admission, we decided to initiate treatment with intravenous clomipramine, building to 75mg in association with risperidone 2mg once daily. During a 10-day period, we noticed a striking transition from generally hostile behavior to one of cooperation with the treatment and remarkable social interaction with other patients and professionals, which was at odds with previous inpatient behavior. However, when we switched her to oral clomipramine (titrated up to 225mg once daily) this outstanding effect was lost, and the patient returned to her baseline behavior.

One month after this last admission, the patient was discharged with a therapeutic plan that included CBT, in which, once again, she refused to participate. Again the patient was lost to follow up.

#### DISCUSSION

To our knowledge, this is the first description of a venous cerebellum development variant in a case of OCD and adds to the understanding of the pathophysiology of this disease. The fact that a cerebellum anomaly has been found in one case of comorbidity between schizophrenia and OCD suggests a relationship between the patient's psychiatric symptoms and her cerebellar variant. Firstly, this patient had one (probably two) first-degree relatives with schizophrenia, one of them-her sister-with obsessive features. Secondly, her OCD was atypical, presenting a continuous and monotonous course, weak resistance to compulsion, passive acceptance of obsessive intrusiveness, isolation, and a striking lack of adherence to treatment. These features remind us of an ancient-yet controversialdiagnosis proposed by several authors: obsessive psychosis-here understood in its original sense, which was related

to a particular experience of being ill rather than to specific psychotic symptoms.<sup>10,11</sup> Moreover, our patient did have, albeit transiently, paranoid ideation. Nevertheless, the impaired judgment this patient undoubtedly presented about her illness has been associated with cerebellar dysfunction in patients with schizophrenia.<sup>3,9</sup> We must take in consideration the fact that schizophrenia is associated with above-average rates of midline cerebral malformations.<sup>12</sup> Therefore, the cerebellar variant disclosed in this case may also be considered as an accidental finding, with no relationship to the clinical description. In addition to the aforementioned evidence supporting the role of cerebellar abnormalities in some symptoms of schizophrenia, the cerebellum itself has been recognized to be involved in the pathophysiology of OCD and, specifically, compulsive hoarding, with which this patient presented.<sup>1,4,6</sup>

In this case, we could hypothesise that cerebellum dysfunction along with some genetic sharing with the schizophrenic relatives, could have contributed to this kind of evolution and account for the symptom coexpression.

#### REFERENCES

- 1. Hamilton NG, Frick R, Takahashi T, et al. Psychiatric symptoms and cerebellar pathology. *Am J Psychiatry*, 1983;140:1322–1326.
- 2. Gillig PM, Sanders RD. Psychiatry, neurology, and the role of the cerebellum. *Psychiatry* (Edgemont). 2010;7:38–43.
- Baldaçara L, Borgio J., Lacerda A, et al. Cerebellum and psychiatric disorders. *Rev Bras Psiquiatr*. 2008;30:281–289.
- Pujol J, Soriano-Mas C, Alonso P, et al. Mapping structural brain alterations in obsessive-compulsive disorder. Arch Gen Psychiatry. 2004;61:720–730.
- Nabeyama M, Nakagawa A, Yoshiura T, et al. Functional MRI study of brain activation alterations in patients with obsessive-compulsive disorder after symptom improvement. *Psychiatry Res.* 2008;163:236–247.

- Tolin DF, Kiehl KA, Worhunsky P, et al. An exploratory study of the neural mechanisms of decision making in compulsive hoarding. *Psychol Med.* 2009;39:325–336.
- Papazisis G, Mastrogianni A, Karastergiou A. Early-onset schizophrenia and obsessivecompulsive disorder in a young man with Dandy-Walker variant. Schizophr Res. 2007;93:403–405.
- Berrios GE. Obsessive-compulsive disorder: its conceptual history in France during the 19th century. *Compr Pychiatry.* 1989;30(4):283–295.
- Bottas A, Cooke R, Richter M. Comorbidity and pathophysiology of obsessive compulsive disorder in schizophrenia: is there evidence for a schizo-obsessive subtype of schizophrenia? J Psychiatry Neurosci. 2005;30:187–193.
- Lanteri-Laura G, Del Pistoia L. Névrose obsessionnelle. Encycl Méd Chir Psychiatrie (Elsevier Paris) 37-370-A-10. 1987:1–20.
- Solyom L, DiNicola VF, Phil M, et al. Is there an obsessive psychosis? Aetiological and prognostic factors of an atypical form of obsessivecompulsive neurosis. *Can J Psychiatry.* 1985;30:372–380.
- 12. Scott TF, Price TR, George MS, et al. Midline cerebral malformations and schizophrenia. J Neuropsychiatry Clin Neurosci. 1993;5:287–293.