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CASE REPORT

Late-Onset Obsessive-Compulsive Disorder Associated with Left Cerebellar Lesion

Matteo Tonna · Rebecca Ottoni · Paolo Ossola · Chiara De Panfilis · Carlo Marchesi

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Abstract The onset of obsessive-compulsive disorder (OCD) after age 50 is rare and generally related to an organic etiology. An involvement of fronto-striatal circuits has been strongly suggested, whereas cerebellum remains so far scarcely explored. We present here the description of a "pure" late-onset OCD associated with a cerebellar lesion, neither comorbid with other mental disorders nor with neurological syndromes. To our knowledge, this condition was not previously described in literature. The patient is a 62-year-old woman who developed a late-onset OCD associated with a left cerebellar lesion due to an arachnoid cyst in the left posterior fossa. We debate the possible role of the cerebellar lesion in favoring a transition from a predisposing liability (namely an obsessive-compulsive personality disorder and a depressive status) to the onset of OCD in this woman.

Keywords Obsessive-compulsive disorder (OCD) · Obsessive-compulsive personality disorder (OCPD) · Depression · Cerebellum · Arachnoid cyst

Introduction

The onset of obsessive-compulsive disorder (OCD) presents a bimodal distribution, with a first peak in childhood and the second in early adulthood. The onset after age 50 is relatively rare and generally related to an underlying structural brain damage, such as infections, degenerative disorders, brain injury, or cerebrovascular lesions [1]. The study of such "organic" late-onset OCDs may provide a deeper insight into the complex neurobiology of this disorder.

Although the pathophysiology of OCD remains controversial, increasing evidence supports a role for several functional and structural brain abnormalities in OCD, particularly involving not only the fronto-striatal-thalamic circuits but also basal ganglia, corpus callosum, superior temporal gyrus and hippocampus, amygdala, and cerebellum [2].

With specific respect to cerebellum, congenital or acquired cerebellar disturbances may be associated with ruminative and obsessive behaviors [3], and a decrease in cerebellar gray matter, particularly in the left upper area, has been found in OCD patients [4]. Furthermore, cerebellum has been implicated in the pathogenesis of compulsions by virtue of its involvement in learning motor sequence and stereotyped activities [5].

In this case report, we present a late-onset OCD associated with left cerebellar lesion due to an arachnoid cyst in the left posterior fossa.

Case Presentation

B.B. was a 62-year-old Caucasian woman who sought treatment at the outpatient service at the Psychiatric Clinic of the University of Parma, Italy, for the recent onset of obsessive symptoms.

The patient never got married and had no children. At the time of the psychiatric evaluation, she was working as a primary school teacher and was also actively engaged in voluntary associations. She described herself as an "anxious" person; she acknowledged to be orderly and scrupulous in her

R. Ottoni · P. Ossola · C. De Panfilis · C. Marchesi
Department of Neuroscience, Psychiatric Unit, University of Parma, Parma, Italy

M. Tonna
Mental Health Department, Local Health Service, Parma, Italy

C. Marchesi (✉)
Dipartimento di Neuroscienze, Unità di Psichiatria, Ospedale-Padiglione Braga, Università di Parma, Via Gramsci 14, 43125 Parma, Italy
e-mail: carlo.marchesi@unipr.it

daily life and in social relationships, but at the same time was socially very active. Her medical history was silent.

Four months before the psychiatric consultation, the patient started experiencing marked obsessions, mainly aggressive in content, such as intrusive and persistent memories of famous murderers or criminals and repetitive doubts to commit aggressive acts towards children, as well as to utter blasphemous phrases during religious ceremonies.

These symptoms were totally ego-dystonic and were coupled with a strong attempt to resist them (such as avoidance of "trigger" stimuli or strenuous efforts to engage into "nice thoughts") and with a marked interference with daily life activities, including work and social functioning and domestic occupations. In addition, the intense feelings of anxiety and shame fostered by the obsessive thoughts made it difficult for the patient to spontaneously verbalize her symptoms to the consulting psychiatrist.

The psychiatric history revealed some prior brief and mild depressive episodes, the latest occurring in the preceding year, following the death of the patient's mother. However, the patient never got treatment for any of these depressive episodes.

B.B. was administered the Structured Interview for DSM-IV Axis I Disorders (SCID-I), the Yale Brown Obsessive-Compulsive Scale (Y-BOCS), the Hamilton Psychiatric Rating Scale for Depression (HAM-D), and the Structured Interview for DSM-IV Personality (SIDP-IV); such assessment indicated the presence of OCD with severe symptoms (YBOCS total score=26) and mild depressive symptoms (HAM-D total score=12) as well as of an obsessive-compulsive personality disorder (OCPD). Cognitive functioning was normal (Mini-Mental State Examination score=27). Furthermore, the patient complained about persistent "buzzing" and frequent headaches. Therefore, a cerebral magnetic resonance imaging (MRI) was performed, revealing the presence of an arachnoid cyst (26×15 mm) (i.e., a l-cystic malformation characterized by the presence of a substantial CSF collection resulting from active expansion of CSF spaces) in the left posterior fossa, with left cerebellar hemisphere winged inward and reduced in size compared to the right one (Figs. 1 and 2). It is difficult to determine whether such findings were due to hypoplasia, CSF pressure, or both. The cerebellar vermis was normally formed and not rotated. Posterior fossa size was normal. No impaired signal was revealed in the brainstem. No other vascular alterations were observed, with encephalic morphology consistent with the age of the patient.

On neurological examination, oculomotor movements were within normal limits: vertical and horizontal saccades were normal; cranial nerves were normal and her speech was normal and fluent; upper and lower limb tone and power revealed no abnormalities and right upper limb abduction was revealed in the Mingazzini test. Sensation was within

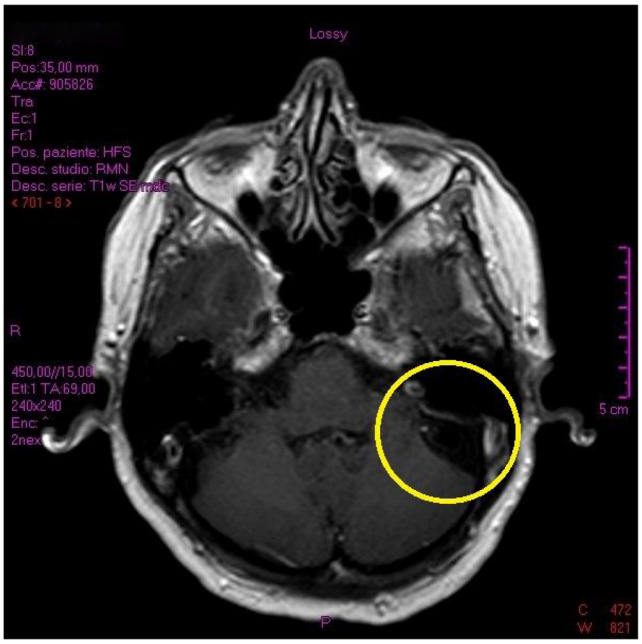

Fig. 1 MRI T2-weighted axial image without contrast showing the presence of an arachnoid cyst in the left posterior fossa

normal limits. The examination of gait and coordination revealed a nod to retropulsion in sensitized Romberg and an impaired tandem gait; tendon reflexes were unremarkable.

A treatment with sertraline was started and its dose was gradually increased to 200 mg daily. Because of the partial response to sertraline observed after 3 months of treatment (as indicated by only a 10 % decrease of the baseline Y-BOCS score), haloperidol (1 mg daily) was added. This resulted in a stable clinical improvement during a 1-year follow-up period,

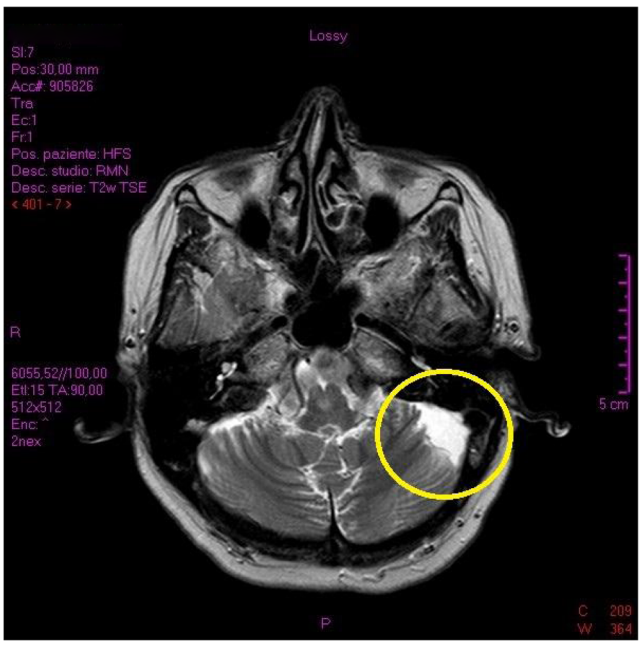

Fig. 2 MRI T2-weighted axial image with contrast showing the presence of an arachnoid cyst in the left posterior fossa

with remarkable attenuation of obsessions (as indicated by a 50 % decrease in the pretreatment Y-BOCS score) and substantial reduction of the patient's subjective distress. The obsessions became less frequent and severe and better controlled by the patient.

After clinical stabilization, electroencephalography (EEG) and brain perfusion single photon emission computer tomography (SPECT) were performed. EEG showed a 9-Hz alpha rhythm, which was responsive and symmetrical, and some anterior rapid rhythms. Brain perfusion SPECT revealed a mildly reduced blood flow perfusion bilaterally in fronto-parietal and temporal cortex (Figs. 3 and 4).

Discussion and Conclusions

The potential role of a dysfunction in the orbitofrontal-striatal circuits in OCD pathophysiology has been suggested by the finding of lesions in the frontal lobes and the caudate nuclei in late-onset OCD [6].

However, three case reports also support the role of cerebellum in OCD neurobiology [7–9]. Interestingly, one of these cases refers to a cerebellar displacement caused by a meningioma in the right posterior fossa [8], a neurological condition similar to our case.

However, none of the abovementioned case reports described patients with a "pure" OCD: in one patient, OC symptoms were associated with paranoid ideation [9], whereas in the remaining two patients, OCD was associated with a neurological syndrome, such as dysphasia [7] or startle syndrome [8].

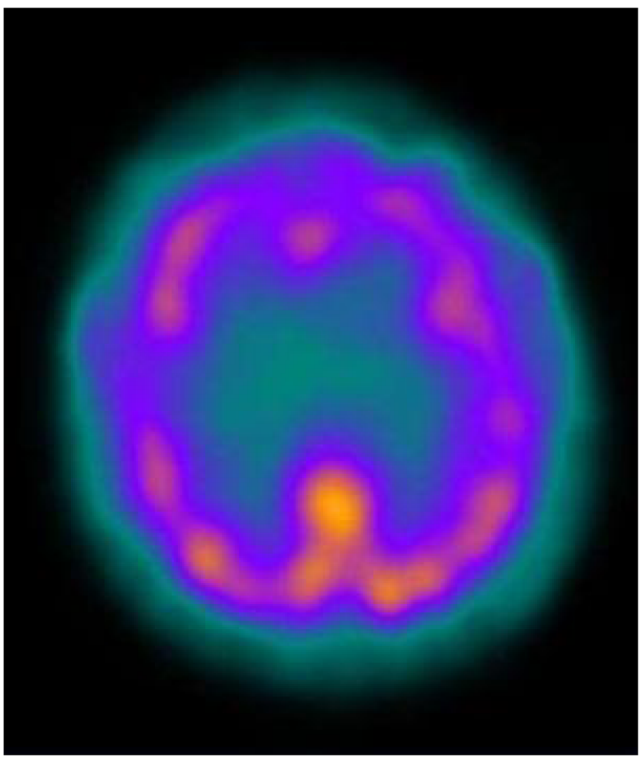

Fig. 3 SPECT axial image showing the presence of reduced blood flow perfusion bilaterally in fronto-parietal and temporal cortex

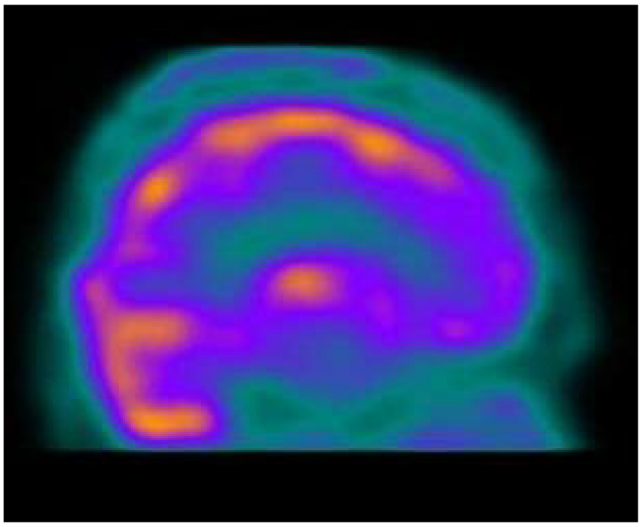

Fig. 4 SPECT sagittal image showing the presence of reduced blood flow perfusion bilaterally in fronto-parietal and temporal cortex

Conversely, in the clinical case described here, a full-blown OCD was neither associated with other mental disorders nor with neurological syndromes or cognitive impairment. Further, MRI did not detect significant cerebral atrophy, vasculopathy, or other cortical/subcortical lesions.

Therefore, the late-onset OCD showed by the present patient could be specifically associated with a cerebellar lesion secondary to the presence of an arachnoid cyst.

The description of the cerebellar cognitive affective syndrome (CCAS) highlights the role of cerebellum in emotional regulation. The CCAS manifestations cover a wide range of behavioral and emotional alterations, including OCD and depressive disorders [3]. The lack of cerebellar control (predominantly vermal-fastigial) on motor and cognitive functions "may hamper the individual's ability to smoothly and automatically maintain the homeostatic, context-dependent responses that govern behavior" [3]. Thus, behavioral and emotional dysregulation associated with cerebellar lesions might result from either excessive (hypermetric) or reduced (hypometric) responses to the external or internal environment. The "dysmetria of thought hypothesis" [3] may explain the wide heterogeneity of neuropsychiatric manifestations due to similar cerebellar lesions in different patients, since a lesion occurring in the "limbic cerebellum" may impair its function in controlling cognition, behaviors, and emotions, making latent psychopathological dispositions or vulnerabilities to manifest at the symptom level.

In this regard, the patient presented here was also diagnosed with an OCPD. Almost a quarter (23–32 %) of OCD

patients have comorbid OCPD, a higher rate than that reported among healthy community controls (1–3 %) [10]. Therefore, it could be hypothesized that the interaction between the lack of homeostatic control due to the cerebellar lesion and the premorbid OCDP might have contributed to the late-onset of OCD developed by this patient. In other words, the cerebellar lesion might have facilitated the transition from OCPD to OCD.

Interestingly, in this clinical case, OC symptoms developed 8 months after the occurrence of a mild major depressive episode. Depression had only partially remitted before the patient's first psychiatric consultation, since depressive symptoms (Ham-D score of 12) were still present at that time. Thus, at the time of the onset of obsessions, the patient was still mildly depressed. This parallels a previous report by Gonzales and Philpot [8] also indicating that obsessions developed during a depressive episode in partial remission. Since OCD is frequently comorbid with depression and dysfunctional fronto-striatal circuits have been implicated in both disorders [11], the present patient's depression could possibly have increased her vulnerability to OCD. Interestingly, depression itself might be due to the underlying cerebellar lesion since the evidence of the involvement of cerebellum in depression and, particularly, the suggestion of an altered prefrontal-cerebellum functional connectivity in geriatric depression [12].

Finally, we cannot exclude that in our patient, the cerebellar lesion due to the arachnoid cyst may represent an occasional neuroradiological finding without any etiopathogenetic link with the onset of OCD. Suggesting this possibility, in this patient, the arachnoid cyst was not associated with an impairment of cerebellar perfusion, as demonstrated by the SPECT. However, the reduction of flow in fronto-parietal and temporal cortex, revealed by SPECT study, might support the hypothesis of a functional impact of the cerebellar lesion on cortical functioning through a disruption of cerebello-cerebral connections [13]. Unfortunately, a tractography was not performed because of the refusal of the patient. Most arachnoid cysts are probably present at birth or develop soon after and are thought to remain stable over time once they are formed [14]. Moreover, reports of rare late-onset OCDs are described without any specific underlying cerebral lesions [15]. Nevertheless, the symptoms caused by an arachnoid cyst may first present after many years and may vary over time [14]. The finding of arachnoid cysts has been associated with different psychiatric conditions in adulthood, such as late-onset psychosis [16], emotional instability, and behavioral disturbances [17]. Interestingly, Heidrich's report refers to an arachnoid cyst located in the posterior fossa adjacent to the cerebellar vermis. Thus, we cannot exclude that in the present patient, the long-standing presence of the cyst may be also involved (together with other risk factors such as OCDPD and life events) in the vulnerability to depression.

In conclusion, the present case report may suggest that the apparently silent arachnoid cyst, thorough its effect on cerebellum, interacted with a predisposing liability (OCPD and depressive status) to determine the onset of OCD at the age of 62 years, even though it cannot be excluded that the cerebellar lesion due to the arachnoid cyst may represent an occasional neuroradiological finding.

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Conflict of Interest No authors received funding for the research and had financial involvement that could represent potential conflicts of interest.

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