Cotard's Syndrome And Catatonia: A Case Report

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Cotard's Syndrome is a rare neuropsychiatric condition, initially described by Jules Cotard as a hypochondriacal delusion and then as Delusion of negation, in which the patient denies the existence of parts of his body, his own existence and / or the entire world. The appearance of a Catatonic Syndrome together with Cotard Syndrome is even more infrequent. We present the case of a 72-year-old patient with a psychotic depression, who developed Cotard's Syndrome and later Catatonia. She achieves good response after the addition of Lorazepam and Venlafaxine to the current pharmacological treatment, so the use of Electroconvulsive Therapy is dismissed. Total remission of symptoms and subsequent functional recovery ad integrum was observed, being evaluated through clinical interview, Hamilton Depression Rating Scale, Bush-Francis Catatonia Rating Scale and Barthel Index. In addition, other case reports on this comorbidity are reviewed, and unlike most of these, the favorable evolution of the patient stands out without the need for Electroconvulsive Therapy. The relationship between the two syndromes has not been elucidated, although some authors have proposed the hypothesis of shared neurobiological pathways and others have postulated the appearance of catatonic symptoms such as the progression of Cotard's Syndrome. To clarify these questions, more studies are needed in order to know the etiopathogenesis of this unusual combination.

Keywords: *Cotard Syndrome, Catatonia, Depression.*

Introduction

Cotard's Syndrome was presented in 1880 by Jules Cotard as a Hypochondriacal Delusion in patients with anxious melancholy, to later refer to the same clinical picture as Delusion of Denial[1]. This delusion can be expressed as a range that goes from the denial of the existence of various parts of the patient's body to the denial of his or her own existence, including the existence of the entire world[2]. In addition, it can culminate in a form of pseudo-megalomaniac delusion or "Delusion of Grandeur," which is characterized by ideas of immortality and immensity. It is a rare, serious syndrome that represents a high risk of self-harm and suicide in the patient[3, 4]. Its presence is associated with various conditions, mainly neuropsychiatric, such as Bipolar Disorder, Depressive Disorder, Schizophrenia, Parkinson's Disease, Ischemic Cerebrovascular Disease, Epilepsy, Subdural Hemorrhage, Insular Cortex Atrophy, and Catatonia, among others[5]. Fluoxetine, Paroxetine, Lithium Carbonate, and combined treatments such as Haloperidol and Clomipramine, are included among the reported therapeutic options.

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The use of Electroconvulsive Therapy (ECT) in cases of psychotic depression or melancholy, especially in combination with post-ECT psychopharmacological maintenance treatment, has been suggested in several studies[3].

Conversely, Catatonia, a neuropsychiatric condition initially described by Karl Kahlbaum in 1874, is a psychomotor syndrome in which "inhibited" and "excited" subtypes are identified. The former is more frequent, characterized by immobility, mutism, staring, and rigidity, among others. The second subtype is less prevalent, and it presents periods of psychomotor agitation. The appearance of the syndrome is recognized in a wide spectrum of medical illnesses and psychiatric disorders, especially in mood disorders[6]. Its pathogenesis has not yet been elucidated, although the influence of the gabaergic and glutamatergic neurotransmission systems is recognized. Regarding prognosis and treatment, it is a life-threatening condition for those who suffer from it, and it benefits from low doses of benzodiazepines and Electroconvulsive Therapy (ECT)[7].

The appearance of catatonic symptoms in Cotard's Syndrome is rare, and only a few reported cases that illustrate such coexistence have been found[2, 5, 8].

In order to contribute to the knowledge of this comorbidity and its management, the following is the case of a 72-year-old woman who, in the course of a severe Major Depressive Episode with psychotic symptoms, developed a Cotard's Syndrome and, subsequently, Catatonia.

Clinical Case

72-year-old female patient, professional technical education, retired, married, and mother of two children. Under medical care for arterial hypertension, without previous psychiatric history or problematic substance use, with preserved ability to carry out activities of premorbid daily living and without psychiatric illnesses in the family.

Two months before hospitalization, she developed insomnia from early awakening, decreased appetite, less frequent bowel movements, and increased distress.

Then, nihilistic delusions of non-existence, absence of organs, and ideas of ruin appeared; she stated that she had died, that she did not exist in this world, and requested that her doctor issue her a death certificate. She reported that she did not have a stomach and therefore there was no point in eating if she could not swallow, adding that she did not feel her heart beating because she did not have one. Additionally, she presented visual hallucinations: "I see babies hanging like grapes, and the sky is completely red." Short-term suicidal ideation appeared on one occasion, with a feeling of partial control.
In the weeks prior to hospitalization, she presented progressive psychomotor retardation, decreased impulse, and dependence to carry out daily living activities.

She started on Fluoxetine and Olanzapine while outpatient, reaching full dose without response, and entered the Intensive Care Unit for Psychiatric Hospitalization (UCIPH) for study and to manage severe a Major Depressive Episode with psychotic symptoms and Cotard's Syndrome. Rigidity and mild tremor of the upper extremities, difficulty in walking, blocking of thought, time-spatially oriented, recent memory preserved, abstract thinking, and persistence of delusions of denial and melancholic symptoms are observed. Upon admission, she scored 39 points on the Hamilton Depression Rating Scale (HDRS) compatible with very severe depression; and 5 points when applying the Barthel Index concordant with a severe dependency from a functional point of view. During the first days of hospitalization, the case study began with requests for exams to rule out other diagnoses; psychopharmacological treatment was maintained along with supportive psychotherapy. Her general examinations were within normal ranges, HIV negative, a brain MRI showed involuntary microangiopathic cortical and subcortical changes, with normal EEG, normal lumbar puncture, EKG, and a two-dimensional echocardiogram showed left ventricular hypertrophy.

Her neurological evaluation posed a hypothesis of dementia with undetermined etiology, an inconclusive study when there is doubt about the correlation between the patient's symptoms and the findings in the brain images. She evolved with active negativism, verbal and motor stereotypes, little verbal fluency, hypokinesia, and stupor. The presence of a Catatonic Syn-

drome associated with the described Affective Psychosis was proposed, with a score on the Bush-Francis Catatonia Rating Scale (BFCRS) of 7/14 in screening and 15/69 in severity.

Lorazepam 2mg was added to manage catatonic symptoms and Venlafaxine was started to establish combination treatment for severe depressive symptoms. Also, a referral was made to a reference center for Electroconvulsive Therapy (ECT). The patient responded to the treatment of catatonic symptoms 24 hours after starting Lorazepam. The reduction of her depressive symptoms and Cotard's Syndrome delusion began ten days after adding Lorazepam (3 mgs/day) and Venlafaxine (150 mgs/day), achieving a good response at four weeks. After two weeks of achieving this response, Venlafaxine was adjusted to 225 mgs per day and the transfer to the reference center for the performance of ECT became effective.

In the reference center, she was hospitalized for twenty days. The patient registered favorable evolution with pharmacotherapy in epicrisis, dismissing the need for ECT, maintaining the combined antidepressant treatment prescribed with Lorazepam and Quetiapine in low doses, with instructions to continue outpatient monitoring.

When she was controlled at the specialty polyclinic, remission of depressive symptoms was observed; on the other hand, family members reported that she had recovered her level of premorbid functioning. At her six-month follow-up, she scored 0 both on the screening and on the BFCRS severity scale, earned 1 point on the HDRS (not depressed) and 100 points (maximum score on the ability to perform activities of daily living) on the Barthel Index.

Discussion

The case of an older adult female patient who, in the context of a melancholic depression, developed intense nihilistic delusions and isolated visual hallucinations, has been presented. When she failed to respond to antidepressant and antipsychotic treatment in an outpatient setting, the patient was hospitalized; she then developed catatonic symptoms that responded rapidly to the addition of benzodiazepine therapy (Lorazepam). Regarding the psychotic depressive symptomatology of Cotard's Syndrome, its decrease was observed ten days after the addition of Venlafaxine and Lorazepam, achieving a good response after one month, so the application of ECT at the reference center was rejected. After her hospitalization, she presented a complete remission of her symptoms, making a total functional recovery that was clinically verified through instruments and the reports of close relatives. Although the clinical case and neuroimaging findings initially pointed to the presence of some type of dementia, this hypothesis was discarded after neurological evaluations and in consideration of the favorable evolution of the patient. Still, it was considered necessary to maintain a longitudinal perspective of the case, with strict monitoring and serial evaluations from the cognitive point of view.
The psychopathological richness with which Cotard described the syndrome is far from its use in recent decades, where it has been reduced to a nihilistic delusion.

An study by Berrios and Luque (1995), in which they characterized Cotard's Syndrome by taking into consideration a hundred cases from the existing literature, is an exception to this. They found depression in 89% of the sample, the most frequent nihilistic delusions were those related to the body (86%) and existence (69%), with a high frequency of anxiety, feelings of guilt, and hypochondriacal and immortality delusions. Upon analyzing the sample statistically, they obtained 3 factors: Psychotic Depression, Cotard Type I, and Cotard Type II. The first group was characterized by a predominance of melancholy with a few nihilistic delusions. Cotard type I patients presented hypochondriacal delusions and nihilistic delusions of the body, concept and existence, which the authors pointed out as the "pure" type of the syndrome; its origin then would be in delusions and not in affective disorders, with the therapeutic implications that this could carry. Lastly, the group denominated Cotard type II brought together anxiety, depression, and auditory hallucinations, constituting a “mixed” type[9].

Longitudinally, Yamada, Katsuragi and Fujii (1999) described three stages for Cotard's Syndrome: A "Germination" stage with depressive symptoms and hypochondria as part of the prodromal manifestations; an "Outcrop" stage,

in which the syndrome develops fully with the presence of the delusions that characterize it; and a "Chronic" stage with two variants: a depressive type that presents persistent affective symptoms and a paranoid type with systematization of delusions[10].

These observations provide valuable information and suggest ways to categorize Cotard's syndrome cases, but we do not yet have definitive criteria and instruments to assess the syndrome. This limits our knowledge of the condition, as well as explains the paucity of epidemiological data, the disparity of neurobiological findings, and the difficulties in terms of establishing differential diagnoses and effective treatment schemes[3].

In the reviewed clinical case, the patient presented symptoms compatible with the first group described by Berrios and Luque (1995), that is, a psychotic depression characterized by melancholic symptoms with nihilistic delusions. She transited only through the Germination and Outcrop stages of Yamada et al. (1999), achieving remission of the condition with the use of combined antidepressant treatment along with a benzodiapine and an atypical antipsychotic, without the need for ECT.

There is some overlap of clinical manifestations between Cotard's Syndrome and Catatonia, so the evidence for this association could be more robust than what is currently found in the literature. Furthermore, Catatonia is often underdiagnosed, as clinicians do not routinely screen for it[8]. Mainly, case reports about this combination were found; the majority corresponding to affective psychosis in adults.

Among the cases described with Major Depressive Disorder with psychotic symptoms,

the report of a 55-year-old male patient who responded to ECT, Olanzapine, and Citalopram was found[11]. Cohen, Cottias and Basquin (1997) presented the case of a 15-year-old woman who was hospitalized for nihilistic delusions about of her own existence and depressive stupor, which was treated with ECT, Mianserin and Amisulpride[12]. McClenahan and Westphal (2006) reported the case of a 52-year-old man with Cotard's syndrome associated with catalepsy, negativism, selective mutism, fixed gaze, and postural changes, who benefited from ECT, Lorazepam, and Venlafaxine[13]. On the other hand, Yamada et al. (1999) presented the case of a 46-year-old woman who showed hypochondriacal delusions and denial of the existence of certain organs, associated with negativism and agitation, who responded to ECT[10]. Similarly, Montgomery and Vasu (2007) published the case of a 48-year-old woman who responded to ECT[14]; and Grover, Shah, and Ghosh (2010), that of a 37-year-old woman who benefited from ECT, Olanzapine, and Venlafaxine[15]. Contrary to what the literature shows in these cases, our patient evolved satisfactorily without the need for ECT.

Continuing with the cases of patients with psychotic depression published in the last five years, Simpson, Kaul, and Quinn (2013) described the case of a 68-year-old man with both hypochondriacal delusions and nihilistic delusions existence who subsequently developed arousal, fixed gaze, echopraxia, impulsiveness, and ambivalence, who benefited from the use of Olanzapine, Mirtazapine, Citalopram, and Lorazepam[8]. Basu, Singh, Gupta, and Soni (2013) presented the case of a 45-year-old woman with nihilistic delusions of existence associated with immobility, stupor, mutism, staring, and catalepsy that responded to Sertraline, Olanzapine, and Lorazepam[16]. A different combination of the conditions was described by Weiss, Santander and Barros (2013) reporting the case of a 22-year-old woman with nihilistic delusions of the existence about her body parts along with immobility, stupor, mutism and negativism, who also presented Neuroleptic Malignant Syndrome treated with ECT, Olanzapine, and Lamotrigine[2]. Finally, Huarcaya-Victoria and Podestá-Ampuero (2018) published the case of a 47-year-old man with delusional nihilistic ideas of existence and guilt, associated with mutism, waxy flexibility, ambivalence, and negativism, who benefited from the use of Venlafaxine, Aripiprazole, and Diazepam[5].

Additionally, cases with Cotard's Syndrome and Catatonia have been found in the context of a Bipolar Disorder with a recent depressive episode, such as those reported by Baeza, Salva and Bernardo (2000) in a 20-year-old man with Cotard's Syndrome along with immobility, mutism, and muscle stiffness, which required ECT and Imipramine[17]; and Grover, Aneja, Mahajan, and Varma (2014) in a 62-year-old woman

with nihilistic delusions of existence and immortality, who later developed mutism, rigidity, and negativism, treated with ECT, Olanzapine, Fluoxetine, and mood stabilizers[18]. Comorbidity was also described in a 50-year-old man with Schizophrenia and nihilistic delusions of existence, guilt, condemnation, immortality, hypochondria and persecution, along with immobility, stupor, mutism, staring, rigidity, and waxy flexibility, which responded to Aripiprazole and Clonazepam after two weeks of treatment[19].

The rareness of the association constitutes a limitation when drawing conclusions regarding the subject; however, high concordance is observed in the treatment of both syndromes in that many of them received benzodiazepines and ECT therapy[8]. Weiss et al. (2013) have pointed out that from a neurobiological perspective, both clinical conditions involve common pathways (orbitofrontal and motor), which could explain certain similarities in their symptoms and response to treatment[2]. As a hypothesis, Grover et al. (2014) state that the development of a Catatonia in the context of a patient with Cotard's Syndrome could be due to a process of progression of the latter and the psychotic depression of the case they presented[18]. The elements posed by Weiss et al. (2013) and Grover et al. (2014) are of great interest when establishing hypotheses that clarify the way in which the two syndromes appear jointly; thus, both positions could be complementary.

More studies are required to clarify this relationship, which implies a more complete knowledge of clinical and therapeutic pathogenesis of both syndromes. In this sense, it is essential to have solid knowledge of psychopathology and to search actively for both clinical entities, in order to establish diagnostic criteria that will lead us to investigate their etiopathogenesis with greater precision.

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