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CLINICAL CASE

COTARD SYNDROME IN RECURRENT DEPRESSIVE DISORDER: CASE REPORT

SÍNDROME DE COTARD EN TRASTORNO DEPRESIVO RECURRENTE: REPORTE DE CASO

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ABSTRACT

The Cotard Syndrome, also known as delirium of negation or nihilistic delirium, was described by the French neurologist, Julius Cotard, on June 1880. From that time until the end of the 20th century, in scientific literature, approximately 100 cases were reported that presented symptoms of depression, anxiety, nihilist delusions concerning the body and existence, hypochondriacal and immortality delusions, as well as auditory and visual hallucinations. We present the case of a female patient, 73-years old, from Ayacucho, with a history of sexual assault at the age of 12 and suicidal attempts on 2 occasions, diagnosed with depression at 40 years of age, received treatment with quetiapine, trazadone and clonazepam. She remained with fluctuating medical progress, with symptoms worsening with stressors such as the death of her mother and husband and when she underwent surgeries. Currently, she presents sad and anxious mood, daily crying, ideas of guilt, disability and death, insomnia of conciliation, nihilistic delusive ideas concerning the body, referring that her stomach and bladder do not work, reason why she doesn't want to eat or drink water. Due to the total refusal of food intake and persistent suicidal ideation, she was hospitalized to receive psychiatric treatment and nutritional support. It is concluded that there are few cases reported on the use of psychotherapy in Cotard's syndrome. Therefore, this case in which psychotherapy was indicated to reduce anxious-affective symptoms, to avoid isolation of the person and to redirect his personal and social life, is important.

Key words: Recurring depressive disorder; Cotard syndrome; Delusions of negation (source: MeSH NLM).

RESUMEN

El Síndrome de Cotard también conocido como delirio de negación o nihilista, fue descrito por el neurólogo francés Julius Cotard en junio de 1880. Desde esa época hasta fines del siglo XX se ha reportado en la literatura científica aproximadamente 100 casos. Que presentaban sintomatología depresiva, ansiosa, delusiones nihilistas concernientes al cuerpo y a la existencia, delusiones hipocondriacas y de inmortalidad, así como alucinaciones auditivas y visuales. Se presenta el caso de una mujer de 73 años, natural de Ayacucho, con antecedentes de agresión sexual a los 12 años, y episodios de intento de suicidio en dos oportunidades, siendo diagnosticada de depresión a los 40 años, recibió tratamiento con quetiapina, trazadona y clonazepam. Se mantuvo con evolución fluctuante, empeoraron sus síntomas con factores estresantes como el fallecimiento de su mamá, esposo y cirugías a las que fue sometida. Actualmente presenta ánimo triste y ansioso, llanto diario, ideas de culpa, minusvalía y tanáticas, insomnio de conciliación, delusiones nihilistas concernientes al cuerpo, refiriendo que su estómago y vejiga no funciona, por tal motivo no desea comer ni ingerir agua. Debido al rechazo total a ingesta de alimentos e ideación suicida persistente fue hospitalizada para recibir tratamiento psiquiátrico y soporte nutricional.

Se concluye que, existen pocos casos reportados sobre el uso de la psicoterapia en el síndrome de Cotard. Por lo tanto, el presente caso en el que se indicó la psicoterapia para reducir síntomas ansioso – afectivos, para evitar el aislamiento de la persona y para reconducir su vida personal y social, es de importancia.

Palabras clave: Trastorno depresivo recurrente; Síndrome de Cotard; Delusiones de negación (fuente: DeCS BIREME).

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INTRODUCTION

The first clinical presentation of Cotard Syndrome was performed during an encounter in the Psychological Medical Society in Paris on June 28th, 1880[1]. It was a woman 43 years of age, who was named "miss X", who stated she had "no brain, nerves, chest, stomach, intestines, only skin and bones", furthermore, she stated that "her body was in decomposition". Cotard described: "she said she had no soul, for her God nor the devil existed, and affirmed that she was no more than a decomposition, she had no need to eat to live, that she would not die of natural causes and that she would exist eternally" (Translation Cotard 1980)[1,2].

In 1882 Cotard used the term delirium of negation to describe the conditions of this case, which was published in the book Maladies cerebrales et mentales, in 1891[1].

According to the data, there were prior cases reported, such as one of the French doctor Charles Bonnet, who reported in 1788 a woman who said she was dead[3], or that of the psychiatrist Jean – Etienne Dominique Esquirol, also French, who in 1838 described five analogical cases[4].

In 1893, Emil Regis termed the name Cotard to this clinical manifestation, but it was Jules Seglas, also a French psychiatrist, who spread the term Cotard Syndrome, consolidating it in wide and universal form since 1897[4].

Berrios and Luque, while studying the Cotard Syndrome historically[3], analyzed 100 reported cases in the scientific literature since 1880 until 1995 and found that 89% of those affected presented depressive symptoms, anxiety in 65% and guilt in 63%. Within the nihilistic delusions, those pertaining to the body were found in 86%, while those of proper existence were 69%, hypochondriacal delusions were 58% and immortality 55%. Within the hallucination spectrum, 22% referred auditory hallucinations and 19% visual hallucinations.

Clínically, it was described:

1. Cotard type I: Hypochondriacal delusions, nihilistic delusions of the body, concept and existence.
2. Cotard type II: Immortality anxiety delusions, auditory hallucinations, nihilistic delusions of existence and suicidal behavior.

Cotard syndrome presents two types of delusion. Type 1 (delusion of negation), which could be expressed by the negation of existence of diverse parts of the patient's body, the negation of their own existence, including the entire world. And it could end up as type 2 (delusion of enormity), characterized by ideas of immortality and immensity[3,5].

In the XIX Century, what Jules Cotard described was named délire de négation (delirium of negation), nihilistic delusional disorder or Cotard Syndrome. Patients with this syndrome manifested having lost not only their possessions, height, strength, but also their heart, blood, intestines. Their exterior world was reduced to nothing. This relatively rare syndrome is considered a precursor to a schizophrenic or depressive episode. Today, with the use of antipsychotics, the syndrome is less frequent[2]. In 1882, Cotard described the delirium of negation, characterized by the patient's belief that parts of their body do not exist or do not function or that they themselves have died. The person may also believe their internal organs have stopped working or they are rotting[6].

The diagnosis is currently not classified within the DSM -5, nor the CIE 10.

The management of Cotard syndrome focuses on the management of the subjacent clinical condition. As has been observed in patients with affective disorders, antidepressants can be effective. However, due to the presence of delusions, electroconvulsive therapy has been strongly suggested by literature. When this syndrome is associated with chronic schizophrenic disorder condition, the prognosis worsens. If it appears as an initial presentation of dementia, the chances for improvement are slim. If the syndrome arises as a result of a state of confusion secondary to an organic condition, it is clear that the treatment of the pathology will lead to the patient's complete recovery. Other measures that should be considered in the treatment of Cotard syndrome is watching the patient, as far as the possibilities of self-aggression and suicide, specially recommended in patients with predominantly depressive symptoms and specially when, once treatment is initiated, the patient in recovery phase recovers their mobility in a more active manner[7].

The prognosis of Cotard syndrome is reserved. The patient's complete recovery may occur promptly and spontaneously, even in the most severe cases. On occasion, when dealing with depressive symptoms, the depressive symptoms may resolve, and the delusional ideas may remain.

When this condition is part of a schizophrenic disease, it may resolve to the extent the other psychotic symptoms resolve. In a similar manner, if symptoms remain for many years, they may coexist with a chronic schizophrenic condition[7].

Table 1. Case reporst of patients with Cotard Syndrome and catatonic symptoms during 2010-2017.

Author	Age/sex	Cotard síndrome characteristics	Catatonic symptoms	Reported diagnosis	Treatment
Huarcaya. et al[11]	47 / masculine	Nihilistic delusion of existence, auditory hallucinations	Mutism, immobility, rigidity.	Severe depressive episode with psychotic symptoms.	venlafaxine 150 mg/d, aripiprazol 15 mg/d and diazepam 40 mg/d
Peter V. et al[12]	17 / masculine	Nihilistic delusion of existence, depersonalization		Moderate depressive episode	Olanzapine 5mg/d
Lingshuang He. et al[13]	63 / masculine	Hypochondriacal delusions, nihilistic delusión, sadness, anxiety		Moderate depressive episode, hypochondriac	Olanzapine 10 mg/d, TEC.
Grover et al[14]	62/femenine	Nhilistic delusion (parts of the body, outside world)	Mutism, rididity, mild negativity, careful of self	Affective disorder, bipolar depressive episode with psychotic symptoms	ECT, olanzapine, fluoxetine, mood stabilizers (unspecified)

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CASE REPORT

M.H.G, female of 73 years of age, from Ayacucho, widow as of 7 years ago, has 5 children, currently living with 2 of them. Educational level: illiterate and current occupation is housewife. She presents with a history of chronic gastritis and irritable bowel diagnosed 12 years ago. Psychiatric history: she was diagnosed with depressive disorder at age 40, in the psychiatric department of HNAL. She received regular treatment with Quetiapine a 300 mg/d +Trazodone 75 mg/d + Clonazepam 0.5 mg during 6 months. She has two prior suicide attempts. The first was with benzodiazepines 4 months prior to her admission and the second attempt was trying to hang herself with the curtain cord in her home 2 months before her admissions. She suffered sexual assault at 12 years of age by a young man 17 years old. The family obligated her to marry her perpetrator.

From the beginning of her illness until the present moment, the patient feels sad and refers that it is fluctuating and worsens at times. At 43 years of age, her mother died and her sadness increased and was associated to insomnia and loss of appetite. At 48 years of age, she underwent cataract surgery and the patient refers she felt useless and not able to do the same things she did before, feeling like a burden for her family. At 63 years of age, her husband died and she refers feeling lonely, she cried constantly and had insomnia. At 70 years of age, she underwent hemorrhoid surgery and stopped taking her antidepressants. She felt uneasy, worried about her health, sad, cried, she felt she was not worth anything because she couldn't perform the house hold chores and because her daughter-in-law would tell her that she was a burden. Six months prior to her admission

to the psychiatric department, the patient refers she saw her son hitting her daughter-in-law because she " spent ten thousand soles of their savings without his permission" and that impacted her because when she tried to separate them he did not listen to her, that made her feel useless and helpless because she could do nothing except cry. From that moment she began to feel sad and anxious, cry daily mainly at night, ideas of guilt, insomnia of conciliation and hyporexia. Three months before admission, her daughter noticed she was more anxious, she would say she no longer wanted to live, that she wanted them to kill her, the insomnia worsened, she only slept two hours per day. One month before being admitted, she asked her son to her because she was tired of her health state, saying " I don't want to live so that I don't suffer anymore", she referred that she did not want to eat because "her stomach was stuck and did not work" and "her bladder did not work that is why she didn't drink water". As the days passed, the described symptoms worsened. Due to all these ailments, her daughter decided to take her to the emergency room of a psychiatric specialized hospital. While hospitalized, she referred "I feel sad because my son hasn't killed me as I asked him to, I don't want to eat or drink water because my body doesn't work, I want to die".

The patient refused ingesting food, which is why a nasogastric tube and peripheral line was placed to ensure nutrient support.

The physical exam did not contribute to any symptomatic pathology, her weight at admission was 30 kg and height 1.50m. The mental exam at admission: alert, oriented in person, partially oriented in space and time, without a mental disorder, hypoprosexia, with a tendency to avoid eye contact. Brady psychic thinking focused on somatic complains, nihilistic ideas of delusion, structured passive death ideations, depressed and anxious mood. Diminished appetite. Global insomnia.

There were no alterations found in the blood count (hemogram, vitamin B12, folic acid, liver panel, lipid panel, clotting factors, glucose, urea, creatinine) and urine test. The computerized tomography (CT) of the brain did not present significant findings. Cerebral SPECT found mild hypoperfusion in the frontal, parietal and temporal lobes. Electrocardiogram without alterations. Abdominal ultrasound with simple hepatic cyst of 18mm in the right lobe. She was diagnosed with a recurrent depressive disorder, current severe episode with psychotic symptoms ((CIE-10: F33.3))- After discharge with Quetiapine 300 mg/d, Mirtazapine 15 mg/ night and alprazolam 0.5 mg at night, her progress was favorable. Her sleep improved, her sadness diminished, ideas of death disappeared and her appetite improved. Anxiety symptoms still persist but in less intensity.

DISCUSSION

In this case, the disease initiates with affective anxious and depressive symptoms from 33 years ago, insidious start with fluctuating evolution. The affective symptoms were incrementing during periods of stressors such as the death of her mother, her cataract surgery, the death of her husband, and the hemorrhoid surgery she underwent, this associated with the irregularity of treatment led to the increase of affective symptomatology and appearance of psychotic symptoms, later manifestations nihilistic delusions and guilt. This evolution is consistent with that reported by Yamada et al, who proposed three stages: 1) germination, where hypochondria is frequently observed, 2) blooming, where nihilistic delusions and/or immortality appear, together with anxiety and negativism, and 3) chronic, with two results: a) persistent emotional symptoms, b) systematization of deliriums (paranoid type).

The chronicity in these patients can be explained by the nosological origin of each case, since these may be due to affective symptoms (psychotic depression), hypochondriacal delusions, nihilistic delusions of the body, concept, and existence (Cotard type I) or a combination of both, anxiety, delusions of immortality, auditory hallucinations, nihilistic delusions of existence and suicidal behavior (Cotard type II), as in this case.

Cotard syndrome in the reported case appears within a recurrent depressive disorder which according to literature, the age of once, severity, duration and frequency of depressive episodes can vary. Generally, the first episode appears later than bipolar disorder, with an average onset age of the fourth or fifth decade. Recovery is usually complete, but a small percentage may remain chronically depressed, especially if it relates to people of advanced age as is in this case, and frequently vital stressing situations may precipitate episodes and have a fluctuating course as in the reported case.

The trigger stressor in the last episode was being a spectator of the violence between her son and daughter-in-law, from that moment she began to become sadder, had emotional lability. The

symptoms that worsened included delusive ideas of the stomach that did not work ("it is blocked") and deathly ideation, progress goes according to what is published in other clinical cases[8,9].

Functional studies using cerebral single photon emission computerized tomography (SPECTc) in patients with Cotard syndrome showed bilateral hypoperfusion in the dorsolateral front lobe, the medial frontal-parietal cortex, basal and thalamic ganglions, very similar to this case where hypoperfusion was found in the frontal, parietal and temporal lobes[10].

Regarding treatment, we should consider Cotard syndrome as a severe pathology due to the increased risk of self-aggression and suicide due to the intense affective symptoms and delusions, which is why hospitalization and antidepressants and antipsychotic medications should be indicated, such as in this case. Due to the presence of delirium, electroconvulsive therapy has also been suggested as treatment.

According to the revised literature, when Cotard syndrome is presented in an acute form, the prognosis is Good, which is the case presented, which once treatment was begun there was considerable improvement evidenced[11].

Regarding the use of psychotherapy in Cotard syndrome, there are few reported cases, Bott et al, informed of a case of a patient with schizophrenia that developed Cotard syndrome, and had a Good clinical response with pharmacotherapy and behavioral cognitive therapy[12]. In this case, psychotherapy was indicated to reduce anxiety-affective symptoms, avoid isolation of the person and redirect their personal and social life.

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