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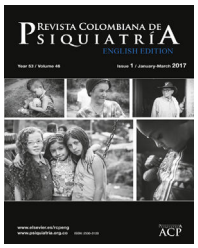

Case Report

"Cotard's syndrome", a description of two cases. Delusion of negation in melancholia versus delusion of negation in paranoia☆

Andrea C. Casas López

Psiquiatría, Universidad del Valle, Cali, Colombia

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ABSTRACT

In 1880, Jules Cotard described a set of delusions in the form of negations that later became his eponymous syndrome. Cotard's syndrome is an uncommon condition characterised by the presence of nihilistic delusions in which the person thinks that "they are dead or that the world no longer exists". This document describes two cases in which a broad and enriching semiology is evidenced from a descriptive point of view, which allows us to review them together with Cotard's syndrome in the light of modern psychiatry. The first case corresponds to a depressive disorder and the other occurs in the context of a non-affective psychotic disorder. A review of the literature is presented from what Cotard described until the current psychiatric classifications.

«Delirios de Cotard», descripción de dos casos. Delirio de negación en la melancolía frente a delirio de negación en la paranoia

Palabras clave:
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RESUMEN

En 1880 Jules Cotard describió un conjunto de delirios en forma de negaciones que luego se denominó síndrome con su apellido como epónimo. Se trata de una condición infrecuente que se caracteriza por delirios nihilistas en los cuales la persona piensa que «se encuentra muerta o que el mundo ya no existe». En este documento se realiza el seguimiento de un par de casos en los que se evidencia una semiología amplia y enriquecedora desde el punto de vista descriptivo, lo que permite revisarlos junto con el delirio de Cotard a la luz de la

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E-mail address: andrea_cl90@hotmail.com

psiquiatría moderna. El primero corresponde a un trastorno depresivo y el otro se da en el contexto de un trastorno psicótico no afectivo. Se plantea la revisión de la literatura desde lo descrito por Cotard hasta llegar a las clasificaciones psiquiátricas actuales.

Introduction

In 1880, Jules Cotard described a set of delusions in the form of negations,[1] which later became known as a syndrome named after him. Cotard's syndrome is an uncommon condition, with a prevalence of less than 1%,[1] characterised by the presence of nihilistic delusions in which the person thinks "they are dead or that the world no longer exists".[2] The delusional content reflects a preoccupation with guilt, despair and death. In its initial course, a feeling of anxiety is described that can last from weeks to years, and the accompanying symptoms can include analgesia and mutism.[3]

Reviewing the literature from the first description in the 19th century to the present day, we found few texts, most of them descriptive, of the case report or case series type, with conceptual differences, various proposals for classification, mention of stages for the presentation of symptoms, and hypotheses about the aetiology. In the articles that provide statistical information, it is suggested that Cotard's syndrome occurs indistinctly in patients with psychiatric or neurological disease, and that psychotic depression or anxious melancholia, which could currently be classified as major depression with anxiety symptoms, tends to be an important factor in the development of this condition.[4]

However, none of the authors managed to establish or define clear criteria for this diagnosis. In addition, Cotard's name has been used as an eponym when referring to delusions of negation in general (of any type), although, from a French nosological point of view, the term also includes emotional and intellectual symptoms. This raises the question of whether Cotard's syndrome should be considered a separate entity.

Starting from this premise, we discuss here Cotard's work and how it has evolved over time according to different authors. We describe two clinical cases as examples, attempting to find the utility and a possible classification of this nosological entity according to modern psychiatry.

Case reports

Case 1

This was a 57-year-old woman, seller of traditional craft liqueur with no history of mental illness. Her symptoms began in November 2016 when she found out that her husband, with whom she had lived for more than 40 years, had a parallel relationship with another woman. As background: drinking traditional craft liqueur ("viche", made from distilled fermented sugar cane juice; 40% alcohol) every two weeks for several days in a row until she became drunk, for over 20 years, the last session was at the beginning of 2017, plus smoking cigarettes, but without clear details of the extent of her habit. Finding out about her husband's infidelity generated feelings of "dreariness and sadness", which she expressed constantly. The patient reported having had symptoms for two months; she initially complained of constipation and abdominal pain of sudden onset. Her next of kin stated that she "said her body had *gone frozen stiff*" and had no strength. They helped her with her activities, and so they had visited different clinics and hospitals, mainly due to constipation, "abdominal discomfort" and asthenia. She then began to report difficulty breathing and a feeling of suffocation. They also reported that she started to suffer overall insomnia, hyporexia, anxiety and restlessness. Medical tests and various diagnostic aids did not show any disease. Five months after the onset of symptoms, she stated she was dying, that she knew it, "she gave us a blessing, she said she was almost dead or dead", for which she was referred to psychiatry in June 2017. At this assessment, the patient said she was dead, her flesh was going to fall off, she insisted her body was *frozen stiff,* and that this happened to people who were dead or about to die; "I'm not alive, with this illness, this accident I had, I went to bed fine and got up ill, *frozen stiff*. I have no appetite. I feel the air isn't getting in, I don't know, I can't breathe, I can't move my bowels". She was admitted to hospital with a diagnosis of a depressive episode, which was initially treated with haloperidol and then clozapine. She finally received quetiapine accompanied by sertraline as an antidepressant, with which there was an improvement in her delusions in general (nihilistic, referential and somatic) and her functionality. During this stay, the paraclinical tests were completed in search of aetiology, which included a brain MRI.

Case 2

This was a 77-year-old single, male patient, with no children, diagnosed with chronic mental illness more than 30 years ago, categorised as schizophrenia. He had gone to university, but not completed his studies. He worked as a salesman until he was 40. He stopped working and began to show functional decline, with symptomatic worsening of his mental illness and periods of time living on the street. He had a history of hip and elbow fractures with sequelae, high blood pressure, chronic obstructive pulmonary disease (COPD) and pulmonary emphysema due to heavy smoking from adolescence until the time of assessment, alcoholism for four decades until the start of therapeutic intervention 15 years earlier, when he joined Alcoholics Anonymous (AA). He had intermittent psychiatric treatment, initially with pipotiazine, then with clozapine, and finally with quetiapine, with partial symptomatic improvement taking the medication, although the experiences of passivity, delusions of persecution and grandeur persisted: "I know that I'm dead. The spiritualities killed me because I'm

more intelligent than everyone else. The spiritualities make me urinate sometimes by squeezing my bladder". Because of difficulties living with his sister and the persistence of the symptoms, they decided to consult psychiatry again at the end of 2017. The sister explained that, since he started going to AA in 2002, the nihilistic and persecutory delusions, characteristic of his crises, became more accentuated, "It started with those spiritualities that he says persecute him and don't let him do anything, with the story that he's dead". We found the patient unaware of the disease ("He says the medications they give him make him sick; this pressure is not from him, that the spiritualities surround him want to see him unwell"), with self-care difficulties; he manifested total insomnia and hyporexia, with persistent delusions of persecution, nihilism, thought transmission, and auditory and visual hallucinations, which he described as spirits that spoke to him and persecuted him ("I can't put on weight because I'm dead, the spiritualities took everything away from me. Sometimes they poison my food, they're always persecuting me"). The reduced intake led to severe protein-calorie malnutrition and functional decline, for which he was admitted to the day hospital programme in December 2017. As the patient's symptoms persisted, paraclinical tests were performed, including neuroimaging, with results within normal parameters. Antipsychotic treatment with quetiapine was restarted, which helped regulate his sleep pattern and his behaviour generally improved, primarily in terms of in self-care and appetite.

Review of the subject

At a meeting of the Medical-Psychological Society in Paris on 28 June 1880, French doctor Jules Cotard (1840–1889) described a set of false nihilistic beliefs,[5] delusions of denial, such as the denial of others or of the environment, the denial of oneself and one's own body, and in some cases specifically the denial of one's own existence.[6] The case involved a 43-year-old woman who believed she had no brain, nerves or chest and that she was made up only of skin and bone. She stated the onset was due to a sudden event associated with her corporeal existence ("a kind of internal creaking in her back that affected her head"), said she was a victim of anguish, believed she was cursed, and asserted that she was eternal and would live forever[7] (1). In 1882, Cotard began to use the term nihilistic delusion to name this condition, and expanded the description of the case in the book *Maladies cérebrales et mentales,* published posthumously in 1891 (2). In outlining this new group of symptoms, he incorporated a form called pure, in which he included three elements: the denial of organs; the feeling of immortality; and the denial of the world.[7] He also compared nihilistic delusions in persecutory syndrome, where they could be isolated, and a set of depressive and anxious symptoms associated with nihilistic delusions that he took as a separate entity and called anxious melancholia with nihilistic delusions.[8,9]

Regis registered the syndrome in 1893 with the eponymous "Cotard's delusion" and stated it was not necessarily associated with depression,[10] while Séglas, in his 1897 book *Le Délire de Négation*, consolidated and widely disseminated the condition under the term Cotard syndrome.[2,3] He believed that delusions and especially nihilistic delusions should be classified according to their origin and not by their content, and he proposed a psychosensory, an affective and a motor type. Later, in 1921, Tissot extracted two main components of Cotard's syndrome: one affective, associated with anxiety; and one cognitive, associated with the presence of delusions. Meanwhile, in 1933, Loudet & Martinez sought to clarify the heterogeneity in the syndrome by establishing different types: the first, a non-generalised delusion of negation associated with paralysis, alcoholic psychosis or dementia; and another called "real", which would be found in anxious melancholia or chronic hypochondriasis.[3]

After the Second World War, Perris pointed out that Cotard's intention was to describe a symptom (a hypochondriacal delusion) that occurred in anxious melancholia, adding that, once the nihilistic delusion was established, it dominated the clinical condition and made it chronic, which challenges the notion of a syndrome to give way to the concept of a separate entity. In 1956, De Martisen postulated that it could be a separate form of psychosis when describing the case of an adult woman in the postoperative period for a possible ovarian cancer,[9] which Enoch & Trethowan supported, arguing that, even if other symptoms accompanied Cotard syndrome, nihilistic delusions dominated the clinical picture.[8] In 1968, Saavedra made a new categorisation based on the previous ones, where he described three types, depressive, mixed and schizophrenic, and established a difference between a genuine Cotard's syndrome, which occurs in depressive states, and something he called pseudo-Cotard syndrome, pseudo-nihilist syndrome or cenesthetic schizophrenia, where there are psychotic symptoms without mention of disturbances in affect.[3]

In 1995 Berrios et al.[4] carried out a study in which they described three profiles of presentation: a psychotic depression, a Cotard type I and a Cotard type II. The psychotic depression included patients with melancholia and few nihilistic delusions; patients with Cotard type I showed no loadings for depression or other illness symptoms and its nosology was closer to delusional than affect disorders; and in Cotard type II, patients showed anxiety and hallucinations and represented a mixed group. This was refuted in an article published by Arieti, who claimed the condition belongs to a type of paranoia in psychosis.[9]

Berrios et al.[4] established that the syndrome occurs in all age groups (from the age of 16 to 81), although it seems to be rare in adolescence, with no difference in gender distribution. They also found common thought alterations, such as: nihilistic delusion related to the body in 86%; denial of one's own existence in 69%; guilt in 65%; hypochondriacal delusions in 58%; and delusion of immortality in 55%; in addition to affective symptoms, such as depression in 89% of the subjects and anxiety in 63%. This does not mean that most patients with major depression, psychotic depression or depression with anxiety have nihilistic delusions or Cotard's syndrome. Added to the fact that a small percentage of patients do not have depression, although it may be the most common presentation, having depression is neither a prerequisite nor sufficient in itself to arrive at a diagnosis of Cotard syndrome.[6]

These statistics are complemented by those from the study carried out in China in the mid-1990s, which evaluated

349 older patients with mental disorders, two of whom had Cotard's syndrome (prevalence of 0.57% of the total population studied and 3.2% of patients with major depression[11]) (1). In the 2013 review by Stompe & Schanda in Australia of 346 patients with schizophrenia, three patients had Cotard's syndrome (prevalence of 0.87%).[12,13]

In Latin America, there was a Mexican study carried out in 2010, where four patients had Cotard's syndrome out of a population of 1321 neurological and psychiatric patients (total prevalence, 0.3%).[1] Of 479 patients with mental illnesses, three had psychotic depression and Cotard's syndrome (prevalence of 0.62%)[13] (6).

As far as the clinical course is concerned, in 1999 Yamada et al.[14] made a proposal for staging of Cotard's syndrome and defined three stages: germination, blooming and chronic. The germination stage is characterised by hypochondriasis, cenesthopathy and depressed mood, and is difficult to diagnose. In the blooming stage, nihilistic delusions and delusions of immortality occur along with anxiety and negative thoughts. The chronic stage is differentiated into two: one form with persistent emotional disorders (depressive type); and a second in which depressive symptoms become less prominent (paranoid type).[3]

Cotard's is not currently described as a separate entity in our psychiatric practice and it does not have a definition in the main diagnostic categories of the Diagnostic and Statistical Manual of Mental Disorders (DSM) or the International Statistical Classification of Diseases and Related Health Problems (ICD). It is typically represented as a delusion or symptom that may be part of other disorders. Nihilistic delusions are reported as an example of mood-congruent delusions, mainly in depressive episodes with psychotic symptoms[15] (3). However, it has also been associated with schizophrenia and bipolar affective disorder, in addition to being described during hyperthermia caused by infectious diseases, and after head trauma and other neurological diseases.[5,16] Recognition of this set of symptoms therefore continues to be important, particularly in terms of taking into account the mechanisms, prognosis and treatment.[3]

Discussion

In his writings on so-called Cotard's syndrome, the author draws a parallel between those who have nihilistic delusions in the context of depressive symptoms and those who have them in the context of persecutory symptoms.

Initially, we find that Cotard described the "accursed" with nihilistic delusions, and explained that in these delusions, the destruction of the organs was completed, the body was reduced to an outward appearance. In these patients, the delusions described were characterised by being of denial, of being alive, of its characteristics, and even went as far as being metaphysical denials.[1] The disappearance ranged from the "moral" to the physical, beginning with "losing" their mental faculties and then their body. The French doctor said that changes in sensitivity were common in these patients; for example anaesthesia, which reaffirmed the "perishable" state, "being dead". He explained that auditory hallucinations were rare and when they occurred they were consistent with delusions, so the patients saw themselves as maintaining a constant monologue, repeated as a litany; in contrast, visual hallucinations were common.[7] He also defined that they showed characteristics close to those of anxious melancholia, as depressive manifestations were previously called; where there were states of intense anguish and anxiety, with constant complaints, the patients begged for permanent help. He specifically said their hypochondriacal ideas seemed to be delusional interpretations of the sensations experienced by depressed people, aggravated by the conviction that there was no cure. It was as if that anxiety were attributed to or associated with fears, ideas of guilt, perdition and condemnation, for which the patients accused themselves, believed themselves incapable and unworthy, thought misery and shame were the origin of "the evils"; being more severe with themselves than with others or their environment and generating propensity to suicide and mutilations, to the ideas of not being able to die because they were possessed or cursed.[7] They also ended up not eating, completely refusing food, punishing themselves and arguing that they did not have a stomach. This made it clear that the depressive state was resonant with their actions, as well as permanent, with them in a true depression. The picture described was one of sudden onset, in people whose health was seen as "good" previously, with an equally sudden recovery. However, the condition was likely to come back over time, particularly in people with so-called chronic "melancholics".[7] Our conclusion then is that these melancholics with nihilistic delusions could be considered patients now referred to as depressed with psychotic symptoms. Our case 1 is an example of this, where the patient developed symptoms that she herself described as sudden ("I went to bed well and when I got up to move my bowels I couldn't"), with motor restlessness, sad affect, overwhelming anxiety, speech resonant with affect and ideas of disability, for which she was diagnosed with a depressive episode. Also referring to alterations in her sensitivity, she said that "her body was *frozen stiff*", along with various somatic complaints, until finally denying her existence.

Cotard also explained that the nihilistic delusion was seen differently in the persecuted[1] or "monomaniacs", who would be considered as those who had paranoia or a non-affective psychotic disorder (including schizophrenia). In these patients the organs were attacked in a thousand ways, either by mysterious procedures or by "pernicious" influences that came from various external elements, such as places or people; but the organs were not destroyed, as if they were reborn. The truly persecuted went through phases in the delusion, from hypochondriasis to megalomania, having a variety of types of delusions, including of grandeur; revealing that they denied out of mistrust, for fear of being deceived, or because they ended up living in their imaginary world. In contrast to the melancholics, the disappearance went from the physical to the mental, with them maintaining a good opinion of themselves and making self-harm rare. He also stated that in these patients, it was rare for the "complaining anxiety" or deep depression of the melancholics to appear; so they were not found to accuse themselves, but rather they accused others or the external environment for what happened to them, making themselves look like objects of persecution.[7] He mentioned the development of mistrust before the onset

Table 1 – Comparison of nihilistic delusions.

	Melancholic	Persecuted
Affect	Depressive	Variable, tendency to flattening
Anxiety/anguish	Overwhelming	Rare
Mutilations and suicide	Common	Rare
Sensory changes (anaesthesia)	Common	Rare
Auditory hallucinations	Rare but consistent with the delusions	Vital signs
Visual hallucinations	Common	Rare
Delusions	Primarily of negation	Multiple, including of grandeur
Ideas of guilt	Common	Rare
Onset and development	Sudden	Early, with slow development and progression

of symptoms in these patients, and that they were more severe in judging others than themselves. Cotard also said it was not common to see changes in sensitivity, but that the auditory hallucinations were constantly present as a dialogue, independent of the delusions; being a continuous delirium with paroxysms, with an early onset and slow and progressive development lasting a lifetime (Table 1). This is reflected in case 2, where a long-standing, progressive picture is described, characterised by multiple delusions, including nihilistic and delusions of grandeur. These are consistent with the constant auditory hallucinations presented by the patient ("I know I'm dead. The spiritualities killed me because I'm more intelligent than everyone else"), in addition to having flat affect and being the central object of persecution ("The spiritualities took everything from me. Sometimes they poison my food, they're always persecuting me").

Conclusions

When describing the cases according to contemporary nosology based on epidemiological reports and clinical descriptions of the presentation, without losing sight of the original nineteenth-century comparison between nihilistic delusions in persecutory syndrome and melancholia with nihilistic delusions, we see that these proposals do not fit any current classifications, and are in the middle of a still unresolved debate regarding Cotard's syndrome. This set of delusions could therefore be considered a separate entity with its own characteristics, as Cotard initially believed. In that case, an evaluation of its biochemical, physiological and aetiological mechanisms would have to be made to determine an accurate prognosis and treatment, as was thought in previous studies.

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